PSYCHOSOCIAL OUTCOME IN WOMEN WITH
CONGENITAL ADRENAL HYPERPLASIA

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ABSTRACT

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The thesis reports research examining the psychosocial effects of congenital adrenal hyperplasia (CAH) for a group of adult women. This chronic condition is a genetically transmitted endocrine disorder resulting in high pre- and post-natal levels of androgens, together with possible salt balance problems. For chromosomal females the abnormal androgen levels result in masculinisation of the external genitalia. Life-long medical management necessitates daily administration of replacement hormones, very commonly surgery to feminise the genitalia, and regular hospital outpatient visits. A qualitative methodology was used to investigate the experiences and their meanings that arose from growing up with CAH for N=19 women aged between 18 and 34 years. Comparisons were made with those of an age-matched sample of women with early-diagnosed diabetes (N=17), a chronic condition with many similar demands as CAH. Concepts and relationships between them, identified from interview data, are proposed as either contributing to, or as resulting from the women's perceived need to maintain the 'secret' of their condition. The contributing concepts to this need were communication issues, the perceived value of the hospital clinics, and the level of understanding and the demands made by the condition. The areas of education and employment and social and sexual relationships were identified as effected areas. The findings of the main study were supported by those from a questionnaire study of a group of parents of young girls with CAH; concepts identified from this data reflected those of the main study. The contributions of two cultural discourses – 'the opposite sex' and 'under doctors orders' were considered. The implications of the findings for the future management of those with CAH were considered: these included long-term access to multidisciplinary counselling services with the aims of both providing information and, most importantly, addressing the cultural discourses that effect understanding.
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INTRODUCTION

Having a chronic condition, which requires regular medical supervision from early childhood, is an experience that might be expected to show effects across many areas of an individual's life. Such a proposition suggests a number of related issues, issues such as the identification and extent of effects, and the factors to which such effects may be attributed. Factors to which effects may be attributed might be those related to particular characteristics of the condition or the more generic experience of growing up with the demands of a chronic condition. These alternatives imply different comparative outcomes: the first would predict that different effects would occur for different chronic conditions, whereas the second, that similar effects would occur across different chronic conditions.

The research reported here examines these issues. An opportunity to investigate the effects of a chronic condition was presented by the identification of a group of women with congenital adrenal hyperplasia (CAH) who had reached adulthood and who as children, had been medically managed at The Hospital for Sick Children in London. The issue of the condition specific or general effects was examined by comparing outcome for this group, with that for a group of women with diabetes mellitus.
CAH and diabetes, while having some differences between them are similar in that they are both chronic endocrine disorders and require life-long medical management. This includes regular outpatient visits, the need to provide blood for chemical analysis, and daily medication that may require adjustments during episodes of illness. In addition, there is some evidence that the sexual response may be effected in both chronic conditions (Bancroft, 1988, 1989; Zucker et al, 1996).

The underlying pathology of the bio-chemical mechanisms that result in the two conditions are, of course, dissimilar. CAH is due to a genetic enzyme (21 hydroxylase) defect which affects the production of hormones in the adrenal glands, and this results, for girls, in the masculinisation of the external genitalia before birth. Diabetes is the result of an impairment of insulin production, which leads to excessively high levels of blood glucose. In order to understand the effects of the experience of growing up with CAH or with diabetes, it is necessary to consider in more detail the role of the endocrine abnormalities.

THE ROLE OF THE ENDOCRINE SYSTEM IN CAH AND DIABETES

The endocrine system in pre-natal sexual development

The endocrine system plays an important role in the prenatal differentiation of males and females. A mother’s ova and a father’s sperm each contain 23
chromosomes in which all the genetic information is coded for the resulting embryo with forty six chromosomes. One of these chromosomes is known as the 'sex' chromosome- X in the ova, and either X or Y in the sperm, and the combination of these in the fetus, resulting in either XX or XY, will determine chromosomal sex, either female or male respectively. At birth the classification of the sex of the neonate as 'a boy' or 'a girl', is based not on chromosomal analysis but on the appearance of the external genitalia. The congruity of the classification of the infant as 'boy' or 'girl' with the chromosomal sex depends upon clear differentiation of the external genitalia; where there is some ambiguity, gender misclassification can occur. It is the endocrine system, under the control of the presence or absence of the Y chromosome that determines the classifying characteristics of the external genitalia.

Up to about seven weeks gestation, the normally developing fetus is not obviously either male or female, both having the same pair of gonadal structures. The presence of an area on the Y chromosome (Sry) is responsible for initiating the differentiation of the testes in the male, with the absence of this area resulting in development into ovaries (Koopman et al, 1991). From the third month of gestation the testes begin to secrete testosterone, an androgen hormone, which locally stimulates the development of the male internal reproductive ducts - the Wolffian system. A second hormone secreted by the testes, the Mullerian-inhibiting hormone, causes the female internal ducts to degenerate (Goy and McEwan, 1980). In the absence of a Y chromosome and the development of testes, these hormones are not produced and the Mullerian ducts develop to form the uterus, cervix, fallopian tubes and upper vagina.
Testosterone also controls the development of features of the external genitalia. Initially there is a single external opening, the uro-genital opening that leads to both the bladder and the internal genitalia. The precursors of the external genitalia are at this stage common to both chromosomal sexes, and consist of the genital tubercle, the urethral folds, the lateral bodies and the labio-scrotal folds. Testosterone enlarges the genital tubercle to form the head of the penis, and the lateral bodies to form the shaft with the urethra extending so as to pass through it. In the absence of testosterone, this enlargement does not occur and the tubercle remains as the clitoris, with the lateral bodies forming the clitoral hood. In the chromosomal male the urethral folds fuse to become the scrotum into which the testes descend shortly before birth. In the chromosomal female the folds remain divided as the labia and the gonads develop internally into ovaries. The functions of androgens in the sexual differentiation of the external genitalia has been supported by animal research; injections of androgen hormones into female embryos during the critical period have been shown to reverse the external genitalia to a male pattern, and castrated male embryos develop female external genitalia (Jost, 1972).

Further actions of the endocrine system

There are two main classes of gonadal hormones - androgens and oestrogens - testosterone being the most common androgen and estradiol the most common oestrogen. Although both classes of these gonadal hormones are present in males and in females their relative levels differ. The release of more oestrogens than androgens by the female ovaries, and vice versa by the male testes, has led
to the misleading practice of referring to androgens as the 'male sex hormones' and to oestrogens as the 'female sex hormones'. The relative amounts and proportions of these 'male' and 'female' sex hormones vary a great deal between individuals. Ellis (1982) has reported a five to ten per cent overlap between androgen levels in normally developing male and female fetuses, and this hormone is secreted both during gestation and after birth by the adrenal glands of both sexes.

Negligible amounts of sex hormones are secreted by both sexes in the early years after birth, but the maturation of the reproductive organs at puberty depends upon complex changes and large increases in production. The greater rise in oestrogens in the female results in development of the breasts, regulation of the changes relating to menstruation, ovulation, and the lubrication of the vagina. It is the 'male' androgen produced by the adrenal glands and ovaries that are central to the growth of body hair, and the normative range of the extent of this growth in the female is wide. McKnight (1964) in a self-report study of 400 randomly identified female students found that 26 per cent reported facial hair, 17 per cent hair on the chest and/or breasts, and 35 per cent on the abdomen. Hair on the upper arms and legs was reported by 70 per cent with 84 per cent having growth on the lower limb areas. Androgen has an affect on the functioning of the sebaceous and apocrine glands; blocking of these is partly responsible for the acne that is often a characteristic of adolescence. The muscular development occurring at this time, especially that of the neck, chest and shoulders areas (Glucksmann 1974) is also in response to the increased androgen production. The female sex characteristics of the rounding of the
hips, development of the breasts, re-distribution of fat, and menarche, all of which are important in sexual attraction, are under the control of the endocrine system.

In addition to androgens, the adrenal cortex produces two other important hormones: aldosterone and cortisol. The action of aldosterone is on retention of sodium by the kidneys, and this hormone is essential for the maintenance of normal amounts of body fluids and salts. Cortisol, a glucocorticoid, acts with other hormones particularly the adrenaline released by the core or medulla of the adrenal gland, and is responsible for mobilisation of energy stores during stress.

Aldosterone, cortisol and androgen are produced by the action of enzymes. Cholesterol is firstly converted into pregnenolone and then either into cortisol, aldosterone, or via 17-progesterone, into androgens. If the endocrine system is functioning normally, feedback loops maintain the aldosterone, cortisol and androgen hormones at optimum levels. However, in the commonest form of CAH the deficiency of the 21-hydroxilase enzyme results in the abnormally high levels of androgens that causes the abnormalities of the female external genitalia.

**Effects of the hormonal system dysfunction in CAH**

CAH is one of a group of endocrine disorders due to recessive genetic errors. The most frequently occurring form, about 90 per cent of cases, is that involving 21-hydroxylase enzyme deficiency (New, 1985). The deficient activity of this enzyme, necessary for the synthesis of adrenal hormones, leads to the accumulation of high levels of androgens during fetal development. At birth,
the genitalia of chromosomal males with this condition show very little abnormality, however chromosomal females are usually born with a degree of genital abnormality. The genitalia of these females are said to be 'masculinised' by the excess fetal androgen production and, depending on the levels of excess, the effects can range from a mild enlargement of the clitoris to a fully masculinised penis and scrotum.

For both sexes, the natural course of CAH results in further penile or clitoral enlargement due to the continued high levels of androgens. Although there is early excessive bone growth in males and females, the bone maturation becomes advanced and this results in short adult stature (Klingensmith et al, 1977). This is accompanied by the early appearance of the secondary sex characteristic of pubic hair growth, and for females, early breast development. For both sexes there are a number of further abnormalities caused by the CAH endocrine dysfuctions. In a proportion of cases the enzyme deficiency also results in a failure of salt and fluid retaining mechanisms with resulting dehydration. Although the structural aspects of the internal sex organs of chromosomal females are mainly unaffected by the excess androgens and therefore the uterus, ovaries and fallopian tubes develop normally, further functional problems may develop in adulthood, such as hirsutism, amenorrhea, and infertility.

**Diagnosis of CAH in the neonate**

Although it is possible to establish a pre-natal diagnosis of CAH by measurement of the steroid hormones in the amniotic fluid and by examination of DNA, these are only considered feasible if there has been a previously
affected child. More usually the diagnosis of CAH is made post-natally by tests including the measurement of the blood levels of the various steroid hormones made by the adrenal cortex. These, together with the identification of the chromosomal sex by examination of blood white cells grown in tissue culture, and a family history of incidence of CAH, are the major diagnostic indicators where there is ambiguity of the external genitalia. The need for these tests may result in some delay in clarifying the sex of the neonate and stabilising the hormonal dysfunction. Delay of diagnosis may be even greater for those neonates where the genital abnormality is less severe. Here investigation may not commence until during the first weeks of life the hormone imbalance produces dehydration, or until there is development of features of precocious puberty. In severe levels of CAH, where diagnosis is delayed or there is a poor response to treatment, there is a risk of death from an adrenal crisis (Cutler and Laue 1990).

**Effects of the hormone system dysfunction in diabetes**

Diabetes mellitus is a chronic disorder of carbohydrate, lipid and protein metabolism. Current understanding of the cause of insulin-dependent diabetes suggests that it arises from an autoimmune attack on the pancreas (Atkinson and Maclaren 1990). This process is selective and is restricted to the insulin-producing beta cells in the islets of Langerhans in the pancreas. Insulin aids most of the body in the utilisation of biological fuels, its action being crucial for the efficient synthesis and storage of energy from carbohydrate foods, for protein synthesis, for the storage of fats and the laying down of energy reserves in the liver, muscles and adipose tissues. As the pancreas ceases to produce
insulin, glucose (the end product of complex carbohydrate metabolism) accumulates in the blood, and the body becomes dehydrated as the kidneys filter out this excess into the urine. As the conversion of glucose into energy fails, the body attempts to utilise fat as an alternative fuel, with the result that if the breakdown of fat continues unchecked, ketones and acidic by-products develop. These, combined with the dehydration, can induce coma. As a result of this metabolic pathology, the untreated diabetic shows signs of weakness, weight loss, and constant thirst and urination as the kidneys attempt to deal with the pathological blood composition. If treatment is not instigated the condition progresses to diabetic coma and possible death.

It is clear that control of the endocrine dysfunction is essential in both diabetes and CAH. This is usually achieved by the administration of replacement hormones to return the abnormal levels to as near normal as clinically possible.

THE MANAGEMENT OF CAH AND DIABETES

The medical management of CAH

The goals for the medical management of CAH are the control of circulating adrenal hormones to levels as near normal as possible and the surgical correction of any genital abnormalities. The first of these is achieved by administration of glucocorticoids and where necessary, mineralcorticoids (fludrocortisone) to control salt loss. These replacement hormones will minimise the risk of poor growth, further virilisation and dehydration. As the replacement hormones do not remain in the body for long periods, it is
necessary for them to be taken at regular intervals; failure to do so will result in the re-occurrence of abnormal levels of circulating adrenal hormones and further masculinisation and dehydration.

Currently, early surgical correction of the external genitalia is advised when the endocrine balance is corrected and the infant stabilises. In the mildest of virilisation, where the clitoris is only slightly enlarged, surgical repair may not be considered necessary. In the past for more severe virilisation resulting in a larger penis-like clitoris, total clitoridectomy (removal of the clitoris and glans) was commonly performed so as to avoid the problems of a painful mass following the re-siting of the glans. More recently, re-positioning of the glans and recession of the clitoris (clitoroplasty) has been a more favoured procedure, with the labia being normalised during the same operation. In such severe cases it may be necessary to open the entrance to the vagina, and in extreme cases the vaginal passage may have to be constructed; with this level of masculinisation it may be necessary to re-locate the urethra. Minor repair procedures in the teens may be necessary and are usually related to the stretching of the vagina.

The effectiveness of treatment during childhood is routinely assessed at regular outpatient clinic visits, these visits commonly being made four times a year throughout the developmental period. Indicators of management effectiveness during childhood consist of blood hormone levels, rate of growth and the skeletal bone age, this latter being assessed regularly by X-ray of the wrist. Post-operatively, and at further intervals during childhood and adolescence, the external genitalia are examined and internal sexual organs may be investigated.
by ultra-scan. The hormone replacement treatment carries its own risks; reducing the production of adrenal androgens to near normal childhood levels often requires high doses of glucocorticoid, and such high doses can result in side-effects such as obesity, features of Cushing's syndrome - the 'moon face'-, striations and bruising. The control of hormone levels is often disrupted during periods of illness, stress or trauma, and during these times changes in medication may be needed, occasionally requiring inpatient treatment for intravenous administration of fluids. Attendance at the clinic to enable effective monitoring of the corrective medication is continued throughout life, although typically the visits are reduced to once a year in adulthood.

**The medical management of diabetes**

Levels of blood glucose higher than the normal range (hyperglacaemia) are in the long term associated with diabetic complications; there are increased risks of blood vessel damage, heart disease, stroke, pathology of the eyes, nerve damage and kidney failure. Low blood glucose levels have more immediate effects, and these effects - known as hypoglaecemia - may include poor muscular coordination, cognitive confusion and, if remedial action is not taken, unconsciousness. The medical management for the individual with diabetes is therefore focussed on the administration of injected insulin at levels that control the blood glucose within approximately normal range. The goal of maintaining blood glucose within this range results in a number of requirements. Blood glucose level is dependent upon a number of factors; these include the timing, amount and type of insulin; the amount and type of food intake; the timing and level of exercise; and possibly levels of stress (Bradley, 1978). The accuracy of
subjective estimates of blood glucose levels by diabetic individuals has been shown to be unreliable (Gillespie, 1991), and it is therefore a requirement that regular objective measures are made by testing at home. Such tests most commonly involve a finger prick to obtain a blood sample, the glucose level of which is then measured either by reactant slips or electronically by machine. The function of blood glucose testing is, of course, to enable the level to be brought within the normal range. Low blood glucose requires an immediate administration of glucose, and high blood glucose a review of the regimen to identify insulin, exercise, and dietary or stress adjustments. Although the day-to-day management of the diabetic control is firmly in the hands of the individual, regular attendance at hospital or general practitioner clinic is required for specialist advice and medical supervision.

**RESEARCH ISSUES**

**The sequelae of chronic conditions**

Strauss *et al* (1984) have discussed possible effects attributable to the experience of chronic medical conditions. These authors considered that all such conditions would be:

- a) long term in nature
- b) uncertain in a variety of ways, e.g. in prognosis and through being episodic
- c) dependent upon life-long medical supervision
- d) intrusive in the lives of the patient, his/her family’s life style and the day to day activities of all family members
- e) likely to lead to social isolation and differentness
- f) dependent upon the development of basic strategies and skills to handle key problems
The first three of these attributes are clearly fulfilled by both CAH and diabetes. Both are long-term, with the focus of the medical supervision on the management of the hormonal imbalances rather than on a cure, that is a reversal to a pre-condition health status. There is some degree of uncertainty in terms of prognosis; diabetic complications such as heart and circulatory problems and neuropathy are well documented although the important variables resulting in these complications are often not clearly understood. The long-term prognosis for CAH is also unclear, as satisfactory management strategies for this condition are still too recent to allow assessment of very long-term outcomes. However some outcomes, such as adult height and fertility, are known to be problematic.

There are aspects of both conditions that are uncertain in the short term; the hormonal balance attained by replacement medication is generally variable in both conditions. The hormonal balance is affected by many factors: by developmental changes in physiology, by medical problems that for those without chronic conditions are generally considered to be minor, for example colds and infections, and by other more serious medical conditions. In addition to these uncertainties, there are episodic uncertainties for those with diabetes. Short-term factors such as varying amounts of exercise, changes in diet and emotional responses to environmental stresses may alter blood glucose levels and create periods of imbalance.

The requirement for life-long medical supervision has already been discussed; the remaining three attributes and requirements suggested by Strauss et al
(1984) are central areas of investigation of this study and will be considered as part of the discussion.

**Research into chronic condition effects on childhood development**

Research in the area of possible psychosocial effects of chronic conditions in children is complex and requires a sophisticated methodology. There are large numbers of variables which are often difficult to define and measure, and which are related to many factors. The age at onset, speed of development and specific characteristics of the condition, the timing and effectiveness of the required medical intervention, the characteristics and life experiences of the individual together with the psychosocial, medical and economic support systems available to them and their family, may all affect outcome.

Another important variable is the source of the data obtained on the children; studies have frequently examined information about the child gained from only one source, usually the parent or teacher. Assessment of a child's behaviour may be influenced by background biases, the demands of the situation, and the expectations of the observers including their knowledge of the child's illness and its consequences. The importance of these factors is emphasised by the often-discrepant ratings obtained on the same child by different raters (Rutter *et al*, 1970, Rutter, 1978; Glow, 1981).

These factors, while not all-inclusive, indicate the breadth of variables that may strengthen, weaken or spuriously identify outcome relationships between
chronic conditions and later characteristics. It cannot be assumed that variables will be related in a linear or additive fashion. It is usual, given such difficulties, to fall back on the premise that similar outcome results from different populations and methodologies support conclusions about the strength and direction of associations. Two conclusions based on this strategy have been made about the relationship between chronic conditions and emotional/behavioural outcomes; firstly that individuals with chronic conditions are at risk, and secondly that the outcome is a general one rather than that specific chronic conditions result in specific difficulties. Both of these assumptions need careful examination.

In their 1986 paper entitled ‘Emotional correlates and consequences of Birth Defects’, Nolan and Pless identified four types of studies published over the previous 20 years in peer-refereed journals. The conclusion from their examination of eighty-two studies was that:

the presence of a birth defect or chronic disorder significantly increases the risk of emotional problems in childhood. (p.202).

However examination of the results section for the list of studies shows that less than twenty five percent show significant levels of difference between groups with chronic conditions or birth defects and comparison groups, and one of these was in the opposite to expected direction. There are other studies not included in the Nolan and Pless review that also do not indicate significant increases in risk for these children (e.g. Tavormina et al, 1976; Drotar et al,
The general association between chronic conditions and outcome characteristics remains to be clearly supported by published research evidence.

In considering their conclusion that specific chronic disorders do not lead to specific types of problems, Nolan and Pless (1986) stated:

> Increasingly over the years, the evidence suggests that the specific nature of the disability or defect is not an important determinant of its psychological consequences. Although on purely developmental grounds it would appear reasonable to assume that the age at onset of a condition might help to predict the likelihood of these disturbances, relatively little solid evidence has been produced to support this view. (p.201)

Cassileth et al (1984) came to a similar conclusion. These investigators compared the emotional impact of six different disorders: cancer, diabetes, arthritis, renal disease, dermatological disorders and depression. They concluded that the emotional impact of chronic illness may be quite uniform across the range of disorders, and across a variety of populations. No one group, apart from the depressed patients, differed significantly on measures of anxiety, depression, positive affect, emotional ties, control, and global mental health. They concluded that their results cast doubt on the notion that emotional traits are unique, either in cause or effect to a particular illness. Stein and Jessop (1992), and Pless and Perrin (1985) took similar positions. The one exception to this 'general outcome' conclusion are neurological conditions involving the brain. Howe et al (1993), in their review of the studies aimed at clarifying the general v. specific debate concluded that it was impossible to make a choice between the alternative positions. They also stated that there was consistent evidence that conditions involving neurological impairment put children at much
greater risk for some types of adjustment problems. In conclusion they suggested that the general factor position must be modified, that the answer to the general/specific question must be 'it depends' (p.1167). They proposed that the effects of chronic illness on children may depend on three general sets of factors: the presence of brain based conditions, the specific domain of functioning being studied, and aspects of the subjects other than the health status being studied, for example, age and gender.

There are then, a multiplicity of factors relevant for an understanding of the outcome effects of chronic childhood conditions. As well as physical development, such conditions may have the potential to affect the child's social, cognitive and emotional development, all of which may have implications for later adult functioning. An adequate explanatory model would need to take account of the transactions between various facets of experience and functioning; the social systems of the hospital and school, family, friendships, the medical condition with its treatment demands, and the communication effectiveness of the team responsible for management of the condition. Children are not passively affected by the condition but affect its course by their behaviour; behaviour may be a function of understanding and this, in turn, may be a function of cognitive development. Such an ecological approach would conceptualise the child's biological system as 'nested' within its psychological system, which in turn is 'nested' within its social world (Bronfenbrenner, 1979). Questions as to the appropriateness of a generic model or a series of specific disease models or whether there are outcome characteristics attributable to
chronic conditions, remain largely unanswered and clarification will require a complex methodology.

**RESEARCH ON HORMONES AS EXPLANATIONS FOR BEHAVIOUR**

Research on outcome effects of chronic conditions continues, and is still frequently based on a linear, reductionist, biomedical model. Such a model seeks explanations of dysfunctions in biological structures and processes, conceptualising 'disease' as a departure from an optimum, normal state (Atkinson 1988). It follows from this paradigm that an understanding of an outcome characteristic attributed to a chronic condition such as CAH or diabetes, would be sought in the internal biochemical imbalances. In the case of CAH, particular behaviours have been explained in terms of the high levels of androgens.

Ehrhardt *et al* (1968) reported findings that they attributed to the hormonal abnormalities of girls with CAH. Although the girls in the study were described as having developed a female gender identity, a high proportion of them were also described as exhibiting 'tomboyism'. This was defined as a preference for outdoor active play over indoor less active play. In addition to the methodological problems of this study where the behaviour observations came from parents and teachers who knew of the girls' condition, and CAH girls' differences in experiences from 'normal', there are other more fundamental sources of concern. The assumption that certain behaviours are intrinsically masculine or feminine and provide independent criteria for some biological
reality known as 'masculinity' or 'femininity' ignores the current evidence that gender categories are socially and culturally constructed, that both change as ideas about appropriate gender behaviour change, and that they show cross-cultural differences (Bem, 1993; Fausto-Sterling, 1992; Ussher, 1989).

Similar attributions have been made for the relationship between high androgens and sexuality in individuals with CAH. Money and Erhardt (1972) reported 37 per cent of their sample of CAH females rated themselves as homosexual or as bisexual. As Quadagno et al (1977) pointed out, a high proportion of the CAH girls in the study received late corrective surgery of the genitalia, and this may make the group non-representative. A later study by Mulaikal et al (1987) failed to replicate this high incidence of homo- or bi-sexuality. Again, homosexual women have not been found to have a particular level of hormones; Downey et al (1987) failed to find significant differences in blood levels of testosterone, androstenedione or cortisol between homosexual and heterosexual females. In summary, sexuality is a complex phenomenon and the term includes sexual preferences as well as behaviour; both of these are more likely to be multi-determined than linearly related to hormonal levels.

The observation of possible childhood gender differences in levels of aggression have also been explained either in terms of socialisation, or biological factors in which case the action of testosterone has been proposed as an explanation for differential findings. An associated gender difference is that boys' play involves more 'rough-and-tumble play', that is, play-wrestling and tumbling accompanied by signs that convey that such aggressive behaviour is 'in fun' (Smith and
Connolly, 1972). In contrast, the female gender stereotype of aggressive behaviour is believed to be that of verbal aggression. The evidence for these gender differences is equivocal with the results of some studies reporting findings which support such differences (Archer and Westerman, 1981, Durrett, 1959), and others not (Whiting and Edwards, 1973). The methodological problems and conflicting findings, together with possible alternative causes of reported differences, make recent reports (e.g. Hines 1994) of increased engagement in aggression and rough-and-tumble play by children with CAH difficult to interpret.

Socialisation differences may affect the differential willingness of males and females to engage in aggressive behaviour. Cultural stereotyping influences parental responses to the behaviour of their children; aggressive behaviour by girls may be met with disapproval, whereas it is an important component of social status in boyhood and disapproval would be therefore less likely. This differential view of behaviour is transmitted to the child by parents, teachers and peers through direct and vicarious reinforcement, and by modelling others' behaviour (Bandura, 1977). The media also presents the child with information on the description and value of stereotyped behaviour (Durkin, 1984).

Aggression in adulthood has been studied by the use of different methodologies. In the laboratory a variety of tasks have been used to estimate levels of aggressive behaviour, and outside the laboratory, questionnaires and the use of ‘stooges’ in varying social situations have produced data purporting to measure aggressive and hostile feelings as well as behaviours. The use of such different
methodologies and the conceptualisation of aggression as unitary, have made the results in this area confusing. The use of the statistical technique of meta-analysis has resulted in the suggestion that the degree of support for gender differences in aggression depends upon the circumstances surrounding the collection of data. Stronger support has been found from naturalistic than from experimental studies, and the age of the subjects used has been shown to be important, with children showing greater gender differences than adults (Glass et al, 1981; Hyde and Linn, 1986). If sources outside social psychological research are examined, for example statistics for violent crime, there is again the tendency to claim support for a higher frequency of male aggressive behaviour, in the U.K. in one period almost ten times as many men than women were convicted for violence against others (Social Trends, 1984). However, such statistics may be influenced by reporting factors and classification biases based on legal rather than behavioural criteria (Pollack, 1950. Marsh, 1981). In addition engagement in aggressive behaviour is affected by a variety of social and cultural factors including gender and role expectations (Campbell 1993).

The findings from attempts to demonstrate the activation effects of testosterone on the aggressive behaviour of humans have been inconsistent. Olwens et al (1980) found a relationship between levels of testosterone and questionnaire measures of aggression and hostility. Meyer-Bahlberg et al (1974) selected 'aggressive' and 'non-aggressive' groups on questionnaire scores but did not find any significant differences in testosterone levels between the two groups, a result supported by Kranz and Rose (1972) with prisoners convicted of either violent or non-violent crimes. A more recent study (Langevin, 1991) examining
a similar hypothesis, reported that when alcohol abuse was controlled, there was no significant relationship between the levels of eight hormones, which included testosterone, and aggressive tendencies. A study of women attending a neurobehavioural clinic, reported that those with histories of violence showed higher levels of testosterone. However the direction of causality here must be questioned; there is cross-species evidence that changes in testosterone levels can follow events rather than precede them (Archer, 1979; Rose et al, 1972).

The validity of the belief that research reports support adult males as more aggressive than adult females must be questioned. Conceptually there is a difference between 'feeling aggressive' and aggressive behaviour. Frodi et al (1977) concluded in review that higher levels of adult male aggression reflected higher levels of inhibiting factors in females, factors such as anxiety or empathy for the victim, rather than higher levels of male aggressive feelings per se. Engaging in aggression may be a function of opportunity, availability of means, assessment of outcome and/or other circumstantial or environmental variables, all of which may contribute either singly or in some additive or transactional way to the engagement in aggressive behaviour. In summary, a more complex methodology is required before the male stereotype of greater aggressiveness per se can be understood, and at present hormonal attributions for this behaviour have very little research support.

The research on human sexual behaviour also fails to provide support for direct hormonal control. Dabbs and Mohammed (1992) examined the short-term effects of testosterone on sexual behaviour and reported that levels of early
evening testosterone measured in saliva, did not predict sexual activity that night for sexual partners who were living together. Testosterone levels were raised after sexual activity, a finding that is in line with those showing that short bursts of activity raise the level of this hormone. Low testosterone levels in hypogonadal men do not appear to affect erections in response to erotic stimulation or personal fantasies (Bancroft, 1988). A survey of heterosexual college students by Alexander et al, (1990) failed to find any relationship between women's testosterone levels and their sexual activities.

It would seem that a direct link between sexual behaviour and testosterone has not been demonstrated despite such repeated attempts. This lack of support has led to the qualified suggestion by Bancroft (1989) that the level of testosterone necessary to fuel sexual desire is low and levels above this have no measurable effect. 'Threshold levels' are limited in their explanatory power for differences in behaviour, only permitting a dichotomous classification; below the threshold levels predicting absence and above threshold predicting presence. Alternative explanations for such observed differences in sexual behaviour must therefore be considered.

The work of Udry has emphasised both the direct and indirect effects of hormones on adolescent sexual behaviour (Udry, 1988; Smith, Udry and Morris, 1985). Although Smith et al (1985) found significant differences between the cultural norms for sexual behaviour between black and white teenagers, Zabin et al, (1986) reported that within a group of black adolescents there was a significant association between age at menarche and first sexual experience.
This association may be misleading however, as although age at menarche is under the control of hormones, the decision to engage in sexual behaviour may be more related to social factors. Menarche is a sexual characteristic which acts as a signal of physical sexual maturity and it may be the perception of this 'readiness' that is the crucial, indirect factor, rather than a direct hormonal factor.

If levels of androgens do not directly affect sexual behaviour, there seems little reason to propose high levels of androgen as an adequate explanation for the sexual behaviour of girls with CAH. An interactional model combining cultural and peer effects with maturational factors would be more appropriate in gaining an understanding of the sexual behaviour of these women. Abnormally high levels of adrenal androgens in women with CAH may, in fact, effect sexual behaviour in a negative way. Such levels may put the females at risk for the development of a number of secondary sexual characteristics that are usually held to be characteristic of males. Hair growth on the body and face, over-development of the muscles of the chest, shoulders and neck, and acne are all signs that are commonly seen at the clinic and are reported as troublesome by adolescents girls with CAH. Characteristics considered abnormal are associated with difficulties in acceptance by peer groups and increase the self-image of being 'different', a self-image that may become established at a young age in girls with CAH.

In summary, there does not appear to be support for hypotheses that hormonal levels are directly the cause of a variety of behaviours. It is possible that
hormones may have indirect effects by causing advances and/or changes in physical characteristics which may interact with social variables to effect behaviours. Direct hormonal effects for behaviours in females with diabetes have not been an area of research interest, except to suggest indirect effects of blood glucose levels on such psychological variables as emotional state and locus of control (Zeltzer et al, 1980; Dunn, 1987). An alternative approach to a direct hormone/behaviour relationship would be to ask the question 'what is it about chronic conditions in general, or about a particular condition, that may have implications for the development of the child?

THE IMPLICATIONS OF SOME CHARACTERISTICS OF CAH AND DIABETES ON THE DEVELOPMENT OF CHILDREN

Implications of some characteristics of CAH

The birth of any abnormal child is a traumatic experience for the parents. When there is some uncertainty about the sex of the neonate, the implications can be wide reaching (Slijper, 1984). In CAH there is the possibility that a severely masculinised chromosomal female, who presents with a penis that is under-developed and an absence of testes, may be incorrectly classified as male. This error in classification may not be recognised until a salt-losing crisis develops, usually during the first four weeks of life, and further diagnostic tests are made. A diagnosis of CAH at this stage may involve two classes of difficulties for the parents. Firstly, difficulties arising from considerable social embarrassment and psychological re-adjustment for the family members when a 'son' becomes a
'daughter'. Secondly, a difficulty in providing clear gender appropriate child rearing practices because of retained doubts about the sex of the child.

Individuals may hold very different concepts and views about all aspects of health and illness; about the causes and mechanisms of the condition, and the rationale underlying the treatment regimen (Pill and Stott, 1982; Warwick et al, 1988). Frequently individuals do not passively accept presented medical ideas relating to aspects of their health status. Where ideas do not fit into a pre-existing schema, they may be ignored or rejected (Blaxter, 1983; Calnan, 1984). The endocrine pathology which underlies the observable developmental abnormalities of neonates born with CAH is complex and involve concepts which are at variance with those within many adults' belief systems. It is commonly conceptualised that males and females are discrete, mutually exclusive categories, each of which develops along separate pathways. Those without specialist knowledge may be unaware of prenatal developmental events relating to similarities and differences of male and female foetuses: the common initial developmental pathways, the anatomical similarities between the male and female external and internal sex organs, or that final genital differences are more apparent than real. In the male, the penis develops from the female clitoris, the scrotum from the fusion of the labia, and the testes from the ovaries. Failure to understand the mechanism of these developments may make it difficult to comprehend how, as it seems to be the case, that a baby who is born a 'boy' may become a 'girl'. This difficulty may remain unresolved and have effects over time. Money and Ehrhardt (1972) reported that parents in whose minds there are doubts about the sex of their baby tended to monitor the child's behaviour.
with super-vigilance 'looking vainly for signs to resolve their doubt' (p.153). Confused parenting behaviour and expectations about the appropriate behaviours of their child may accompany this super-vigilance.

In most cultures parents hold very different stereotyped expectations for appropriate behaviour for boys and girls, and from birth the responses of adults are influenced by the expectations held for perceived sex of the baby (Condry and Condry, 1976; Frisch, 1977). In a laboratory study, six-month-old babies were dressed as either a boy or a girl, and mothers of similar aged infants were invited to play with them. The sex of the baby was given as either male or female according to dress, and the perception of the sex influenced the mothers in both their verbal behaviour and their responses to the physical actions of the baby. When the baby was presented as male, the mothers responded with physical action, but with comforting and soothing behaviour when it was presented as female. Thus the response to unfamiliar babies was determined by gender stereotype (Smith and Lloyd, 1978). This early response difference is continued throughout childhood with parents looking more at infants of their own gender and providing them with more stimulation in play (Rebelsky and Hanks, 1971). Fathers are particularly more boisterous in their play, especially with their sons, choosing to initiate rough-and-tumble activities (Lamb, 1981). By the time the infant is twelve months of age the parents are encouraging their children to play with sex-typed toys and to avoid playful activities that are considered more appropriate for children of the other sex (Snow, Jacklin and Maccoby, 1983). These differential expectations and interactions of parents with very young children begin the process of gender socialisation.
Differentiation is continued throughout childhood by the responses of teachers, peers, and parents and continues into adulthood with the outcome of very clear differences in gender roles. However the variations between cultures and historical times in the tasks and stereotyped perceptions of the characteristics of males and females suggest that these are socially constructed rather than biologically determined (Oakley, 1985; Bem, 1993). The perception of men and women as fundamentally different from each other is accompanied by a subtle use of these perceived differences as organising principles for the social life of the culture. Male-female differences are super-imposed on many aspects of society from styles of dress to occupational roles and ways of expressing emotional and psychological distress. It is of course possible that parental and medical doubts about the sex of the neonate with CAH may result in ambiguous expectations about appropriate behaviour. Unclear expectations about appropriate behaviour may encourage the development of some behaviour perceived by society as more appropriate for those of the opposite sex.

The difficulties referred to above together with absences from school because of illness and necessary visits to hospital may lead to the development of feelings of 'being different' from the other members of the child’s peer group. For children with CAH these feelings would be likely to escalate and become the focus of attention at adolescence. This is likely to arise because of factors such as the feelings of invasion of personal privacy consequent upon vaginal examinations and possible further surgery, and the re-evaluation of the ‘self’ commonly occurring at this time.
The implications of some characteristics of diabetes

For children with diabetes absences from school due to hospital visits or to illness may have similar implications. In addition, dietary needs and the necessity throughout childhood of eating at set times, together with the daily injections and blood tests are factors which might make identity issues salient and reduce opportunities for spontaneous social activities, activities which become particularly important for peer group acceptance during adolescence. The parents' tendency to be extra vigilant towards the child, perhaps motivated by the fear of and need to prevent hypoglycaemic attacks, may reduce the growing child's levels of independence (Tattersall and Lowe, 1981). Finally, as with CAH, the life-long necessity of hospital investigations, and the required sharing of developmental issues during clinic visits may result in feelings of invasion of privacy.

PARADIGMS FOR INVESTIGATING THE OUTCOME EFFECTS OF CHRONIC CONDITIONS

In conclusion, both CAH and diabetes appear to have characteristics that may be risk factors affecting development during childhood. Research investigating the issue of outcome effects attributable to chronic conditions has not, however, shown clear-cut support for such effects. A number of authors have found high rates of psychological problems in a variety of conditions such as asthma, diabetes and renal failure, with increased effects where brain dysfunction is part of the condition as in epilepsy (Rutter, Tizard and Whitmore, 1970; Hoare, 1984; Breslau, 1985; Garralda et al, 1988). Other studies have failed to find increased psychopathology in children with chronic conditions (Bywater, 1981;
Drotar et al, 1981). Discrepant findings could of course reflect differences in a number of variables. The nature, stage and severity of the illness, as well as parental factors, the availability and quality of support networks and medical services and methodological differences in studies, are all likely to effect outcome results for both children and adults who experience chronic illness. The investigation of chronic condition effects based on a linear paradigm does not allow research into the interaction of such multiple variables.

The paradigm underlying research into the effects of chronic conditions has recently become more complex. It is now recognised that illness outcome, particularly that of chronic illness, is not simply attributable to either the nature of the condition - be it biologically or socially determined - or to the individual coping mechanisms. Outcome is also affected by the social and cultural context in which the experience occurs (Freidson, 1970). More recently, this complex collection of factors has widened even further to include the individual's cognitive construction of the meaning of their experiences. The body, diseases, and the acquisition and application of medical knowledge have all been considered from the viewpoint of social constructionism (Foucault, 1976). It has been argued by Duden (1991) that such an analysis would suggest developments in medical knowledge do not involve the gradual unfolding of an objective truth about the body and its health status, but that such knowledge is contingent upon the social, cultural and historical context. Disease and the body, therefore, are not stable realities but are socially constructed and influenced by cultural discourses. The traditional expert/patient interaction during a medical consultation is now also seen as open to similar influences. As
society accepts new ways of representing the world, such changes will result in changes of social practices; the direction of changes will be determined by the direction of discourse changes. As society changes its attitudes towards control of the human body, it is to be expected that there would be a parallel change in the doctor/patient role during medical consultations. The expert/patient interaction would move towards one based upon a more egalitarian, shared transaction of two 'experts', both of whom have viable knowledge about their own areas of relevant experiences which contribute to the problem. Research which has as its aim the investigation of such a complex collection of meanings, cultural contexts, and relationships would necessitate a methodology that allows the collection of data that are dynamic accounts of individuals' experiences. The research aim for such an investigation would become *the study of ordinary people's methods of... making sense of everyday life* (Potter and Wetherell, 1987. p18).

THE RESEARCH AIMS OF THE CURRENT STUDY

Writing on the psychology of the female body in 1989, Ussher proposed that

'We need to look at both the wider social context in which the female body is understood and at the context of the individual woman and the influence which her own conceptualizations of her body have on her experience, status and identity'. (pp.12-13)

The implications of this proposal for understanding the effects of illness, especially those of a chronic condition, are that the transaction between the individual, their immediate context and the wider social and cultural issues in which their experiences are embedded, must be included in the research domain. It is the investigation of such broad areas that is reported here.
There were two research aims:

a) to develop an understanding of the experiences of women with CAH which related to their chronic condition, and
b) to understand the extent to which the women's experiences were specific to CAH or similar to or different from the experiences of women who had another chronic condition.

In the case of each of these aims consideration was given to the extent to which the use of quantitative or qualitative methods was appropriate.

The first aim, being experiential and descriptive, was deemed best addressed by using qualitative methods. The use of such methods would minimise the extent to which the data gathering process would be influenced by pre-judgements as to what constituted important aspects of the women's experience as might happen with, for instance, the use of standard scales or questionnaires; qualitative methods also would allow the women to express themselves as freely as possible on issues identified by them as being the most important in the context of their chronic condition.

The second aim, essentially a comparison of the experiences and constructions of women with CAH and those with another chronic condition, was addressed through both qualitative and quantitative methods. As in the first aim, qualitative methods were used to explore the experiential and descriptive aspects. In addition, where elements were identified which could be meaningfully quantified statistical reliability of difference was demonstrated by the use of quantitative analysis.
Consideration needed to be given to the effect of combining qualitative and quantitative methods and techniques in the same study. The key question concerned the extent to which the flow of those parts of the interview designed to elicit qualitative data would be disrupted by asking quantitatively related questions. In the study reported here this would be particularly important given the extremely personally sensitive nature of some of the areas being discussed. These possible effects could be minimised through an awareness of the need for sensitivity to the timing and placement in the interview of any quantitative components. However such components could also be used in a constructive way in the interview process. For example there might be points at which the interviewee would be in difficulties because of the extremely delicate and emotional nature of the subject matter. At such points offering the interviewee the opportunity to quantify a relevant aspect of related experience could facilitate a continuation of constructive discussion. Further, this technique could help the interviewee subsequently to return to the relevant issue and offer a fuller and more personal account of the experience under discussion.

In addition, there can be a benefit in obtaining data about experiences using different routes. Obtaining both qualitative statements and numerical data on an issue could lead to an increase in confidence that the interviewer had a consistent and valid understanding of the interviewee's communication about a particular experience.
Qualitative research is a term that covers a variety of techniques and methods. In this study the procedure and techniques used are those of grounded theory (Strauss and Corbin, 1990). Rather than beginning with a number of hypotheses which are derived from a theory and are to be tested by collection and analysis of data, a grounded method begins with the identification of the area of study. Rather than being predetermined therefore, the main issues of such a study emerge from the process of the collection of the data. As these issues emerge and relationships between them are theorised, they form the basis for a constructed theory that has been inductively derived from the study of the phenomena under investigation. The area of study of the present research was identified as the understanding of the experiences of women with a chronic condition. It was the data collected in the form of the descriptions of those experiences by the women themselves, from which the key issues or elements of importance to them were developed and which in turn formed the basis of the theoretical formulation.
study were twofold: to examine the ways a group of women with CAH had come to conceptualise - to make sense of - their experiences, and to investigate the effects of such conceptualisations. Investigation of the 'experiences' of any group of individuals would seem to demand as its starting point, the description of those experiences by the individuals concerned. These descriptions would require analysis to enable the identification of the individual's constructed 'meanings' and how these transact with the relevant social and cultural beliefs and discourses. The nature of such research, which aims to identify and understand the concepts of experience, would require the use of a qualitative rather than a quantitative research methodology (Strauss and Corbin, 1990).
METHODOLOGY

The research aim outlined above was a broad one requiring a methodology that went beyond questions of 'How many?' or 'How often?' to examine ways in which the women had come to understand themselves in their social world.

ASPECTS OF RESEARCH METHODS

A number of aspects of the current debate about research methodology were seen as relevant to the selection of methods used for this study. These aspects are discussed below.

Quantitative research methodology

Historically, psychology has adopted a positivist position modelled on the natural and physical sciences, and has favoured a linear model that is reductionist in style. The collection and analysis of data have attempted to meet the criteria of generalisability, validity, replicability, and objectivity. These have involved the use of probability levels, and adequate sampling of subject groups via randomisation and the stratification of what are believed to be important intervening variables in order to show statistical support for, and inductive interpretation of, the experimental hypothesis. It has also required objective, valid, and reliable measuring instruments with which to obtain data.
The belief that quantitative methodology is objective and therefore involves the minimum subjective interpretation and the maximum facts, is increasingly recognised as being ill founded. The problem is that this methodology starts with the selection by the researchers of a particular theory. This inherently subjective selection forms the basis of, and therefore contaminates with such subjectivity, all subsidiary selections of experimental hypotheses and procedures that follow. This problem can be exemplified by the case of factor analysis where the outcome factors are dependent upon the subjectively selected items initially entered into the analysis. The same criticism applies to questionnaire studies, where the collection of questions will be controlled by the directing theory. The P.M.S Questionnaire (Moos 1969) which purports to examine women's emotions during the pre-menstrual period provides an example. On the basis of the theory that gave rise to their hypothesis, the constructors of the questionnaire selected and described forty-seven emotions that form the basis of the subjects' descriptions of their emotions during this period. However, these descriptions will be heavily skewed towards a conclusion that negative emotions are predominant because, of the forty-seven emotions, only five are positive in character. One may therefore question if the data collected by this questionnaire allows an objective (unbiased) conclusion.

Furthermore, as in all research, valid interpretation of quantitative research rests upon the application of the reader's critical judgements. Judgements of the adequacy of the experiment design and procedures, and whether the experiment was validly conducted at every stage from conceptualisation to report writing.
Therefore valid interpretations are not firmly rooted in objective statistical issues, but are substantially determined by subjective experience.

Additionally, quantitative research methods necessitate large samples so as to maximise the validity of the retention, revision or rejection of the experimental hypotheses. For example, analysis using multivariate statistical techniques require large sample sizes to allow the effects of different combinations of variables to be examined, and that which uses statistical modelling techniques needs both large subject samples and long-term programmes of data collection (Breakwell, 1994). In addition to sample size, quantitative methods rely on statistical principles for the selection of the sample. Careful sample selection of subjects who will be representative of the total target population, is believed to allow generalisation from the experimental results. Obtaining such a representative sample is frequently difficult, either because the important population characteristics are unknown, or because subjects who would be representative of such characteristics are not available. These requirements create particular difficulties when the population size is small.

The final aspect of quantitative research to be discussed is the limitations of the requirement to pre-determine experimental design. Quantitative research is concerned with variables that are perceived from the theory to be important, possible relationships between them, and how they may cluster together. As noted by Kerlinger (1969), the major criteria of well-constructed research questions for quantitative method are:
a) that the problem should be stated clearly in the form of a question.
b) variables of interest must be defined both conceptually and operationally.
c) the expected relationships between variables should be stated.
d) the statement of the problem must imply the possibility of statistical testing.

The implications of these criteria are that for quantitative research methodology the analysis is planned before the study begins, the variables must be identifiable and definable, and the level of measurement used - nominal, ordinal or interval - must be appropriate for the pre-decided statistical analysis.

**Application to the current research project of the requirements of quantitative methodology**

These aspects of quantitative research raised important issues for the choice of methodology for the present study of CAH women.

First, the breadth of the area to be investigated would necessitate the inclusion of a large number of variables. The testing of possible relationships and clustering of these, together with the application of appropriate statistical analysis, would demand a very large sample of women with CAH, very clearly this would not be possible as the incidence of CAH in the general population is low. Although there is considerable variation across different population groups, among Caucasians - the ethnic group investigated in the study - the incidence is about one in five to fifteen thousand live births (Pang *et al* 1988). In addition to this low incidence, there was the practical problem that the
available sample was limited to the records of one hospital. In sum, a sample size large enough to justify statistical analysis was not an option.

Other areas of concern were the implications of the breadth of the chronic illness experience, the lack of published unambiguous outcome research that related to this, and the difficulties of conceptually and operationally defining the variables perceived as relevant. The inappropriateness of 'setting hypotheses' added to the dissatisfaction with a quantitative research methodology for this project.

Another determining factor concerning the appropriate methodology was the research aims of the study. These were to acquire an understanding - rather than the identification - of experiences, and the suggestion made by Koeske (1980) seemed pertinent to this aim. Koeske suggested that a way should be found to re-integrate the whole person from the jigsaw of parts created by an adherence to the traditional linear, quantitative research. She proposed that what individuals say about their lives and perceptions should be listened to with care, and their views treated as data, rather than restricting their choice of feelings so as to construct 'objective' measurements. Such methods that aim at the participants' subjectivity are now recognised as legitimate areas of investigation for the discipline of psychology (Henry et al, 1997).

The choice of qualitative methodology for the current research

Over the last decade, the use of approaches such as those suggested by Koeske (1980), have been increasing in published research. Of these qualitative
approaches, some have been developed outside the discipline of psychology; for example ethnomethodology has been developed in the discipline of anthropology. Other qualitative methods have been developed within circumscribed areas of psychology, including market research where they are widely employed. The objective of qualitative methods is to gain an understanding of the meaning of the statements of those interviewed or the actions of those being observed.

The debate as to the 'scientific validity' of this type of research still continues (Henwood and Nicholson, 1995; Gillett, 1995; Henwood and Pidgeon, 1995; Morgan, 1996; Stevenson and Cooper, 1997; Sherrard, 1997). As part of this debate, a number of writers (Gabriel, 1990; Ashworth, 1987; Hirschmann, 1986) have pointed out that qualitative methodology is as open to critical evaluation as is quantitative. Critical analysis in qualitative methodology is achieved through the use of a collection of techniques known as 'critical relativism' (Anderson, 1986). The essence of this is to ask what were the ideologies and values behind the research, how the findings have been produced, and by what criteria they have been and need to be judged. The development of credible answers to these questions depend upon maintaining three 'information trails'. The first records aspects of the planning and collection of the data, the second the interviewees' statements, and the third the methods used in the analysis of the data. These records should be open to investigation by others so that they may make an informed judgement as to how the conclusions of the study were arrived at.
In quantitative approaches, critical analysis is considered under the criteria of reliability, objectivity and validity; in qualitative approaches the humanistic criteria of credibility, confirmability, transferability, and dependability are seen as more appropriate. Presenting the interviewees with an interpretation of their statements so as to obtain feedback from their reactions, tests credibility and information from feedback can then be incorporated as additional data. Confirmability involves checks by others of the records and interpretations made by the researcher, while transferability refers to the similarity found in different groups or individuals. Similarity is of course a subjective judgement as no two individuals or groups will be identical in their analysis of their experiences, and the conclusion that similarity or a consistency of findings has been achieved is an interpretation that should be open to the other validity checks. Finally, dependability is the qualitative equivalent to the quantitative concept of reliability. It refers to the attempt to show that the conclusions of the study represent a stable construction of events rather than one that is idiosyncratic. This is usually achieved by the use of more than one interviewer, but the presentation of a selection of the raw transcribed data also allows checks to be made.

In-depth studies with small groups of respondents may be criticised for lack of transferability. The focus of these studies however is on what has happened to the particular group of people, rather than whether or not the patterns and processes of that group are representative of all members of that class of individual. This makes it essential to define and describe the characteristics of those providing the data, and the population from which they were drawn, so
that cross-comparisons with other groups may be made to examine transferability. In this way, variables that may result in different findings can be identified. Transferability can not be automatically assumed on the basis of sampling, but has to be established according to theory and the use of devices that are somewhat similar to quantitative research. These devices may involve extension of the study by increasing the number of the respondents, searches for contrasting cases (negative or extreme instances) and the subdivision of respondents into rational and/or theoretically identified sets. These devices are used at a different stage in qualitative than in quantitative research; in qualitative methodology they are part of the analysis and therefore occur within the ongoing research, whereas in quantitative methodology they are incorporated at the planning stage.

Research should not be evaluated on the particular type of method employed, but on the basis of the appropriateness of the method to the phenomenon under investigation and to the research aims. The hallmark of qualitative methodology research is that it attempts to go beyond "how much" to report the most meaningful qualities of events; the central interest is the meaning of what is said. Rather than the deductive approach of quantitative methods, general conclusions are drawn from a network of meanings by an inductive process of reasoning.

It was clear that a qualitative approach was the method that would allow a better insight into the meanings of the experiences of women growing up with a chronic disorder. As Strauss and Corbin (1990) have pointed out, although
most research places the emphasis on one method, it is possible that the aims may best be served by a combination of quantitative and qualitative methods dependent upon the questions being asked.

THE RESEARCH DESIGN AND PROCEDURE

The design

The recognition of the need to separate the effects on psychosocial outcome due to the specific disorder from those due to a chronic condition in general implied at least a two-group comparison study, each group with a different chronic condition. Although a longitudinal study would have provided information on the development of outcomes, time-scale constraints for the research prevented this. A cross-sectional, two group comparison design, which investigated the meanings of experiences at one particular time point was the practical alternative.

The criteria for the selection of the comparison group were that there should be similarities with CAH in terms of chronicity, age of the onset, the frequency and spacing of attendance at hospital outpatient clinics, and the need for daily medication. Discussion with medical colleagues, an examination of the existing published literature, and the availability of subjects suggested that, while some differences did occur, early-onset Type 1 diabetes was a chronic condition that substantially fulfilled the required criteria.
The procedure

Ethical approval

The study was submitted to, and approved by all relevant hospital ethics committees.

Selection and identification of the subjects

In the current study the CAH population was defined as those females referred to The Hospital for Sick Children, who were born between 1953 and 1972. The identification of the population of diabetics was restricted to patients in the same age range attending co-operating hospitals. In addition to a number of exclusion criteria both populations were further reduced through difficulties in tracing some of the patients and the refusal of some to participate.

Fifty-six girls born between 1953 and 1972 diagnosed as having CAH were identified from the hospital records. The exclusion criteria were: the presence of learning difficulties and/or other medical conditions, non-Caucasian cultural background, a single attendance at the hospital for a second medical opinion, and known emigration. These criteria were imposed to minimise effects due to cultural and/or physical factors over and above those of the chronic condition. The absence of hospital medical case notes and tracing difficulties for second-opinion referrals made these patients inappropriate for inclusion. Others were excluded because they could not be traced or because the adult consultant in charge of their management did not agree to their being approached. The characteristics of the initial and final samples of both groups are presented in
Table 1.

<table>
<thead>
<tr>
<th>CATEGORIES</th>
<th>CAH N (%)</th>
<th>DIABETIC N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Identified from records</td>
<td>56</td>
<td>33</td>
</tr>
<tr>
<td>Exclusions:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Non-caucasian</td>
<td>0 (0)</td>
<td>0</td>
</tr>
<tr>
<td>Other medical conditions</td>
<td>4 (7)</td>
<td>0</td>
</tr>
<tr>
<td>Learning difficulties</td>
<td>2 (4)</td>
<td>0</td>
</tr>
<tr>
<td>Second opinion only</td>
<td>5 (9)</td>
<td>0</td>
</tr>
<tr>
<td>Emigrated</td>
<td>4 (7)</td>
<td>0</td>
</tr>
<tr>
<td>Untraceable</td>
<td>4 (7)</td>
<td>0</td>
</tr>
<tr>
<td><strong>SAMPLE AVAILABLE</strong></td>
<td>37 (66)</td>
<td>33 (100)</td>
</tr>
<tr>
<td>Adult consultant not agree</td>
<td>1 (3)</td>
<td>0</td>
</tr>
<tr>
<td>No reply to invitation</td>
<td>17 (46)</td>
<td>16 (48)</td>
</tr>
<tr>
<td><strong>FINAL SAMPLE</strong></td>
<td>19</td>
<td>17</td>
</tr>
<tr>
<td>Sample Ages</td>
<td></td>
<td></td>
</tr>
<tr>
<td>mean</td>
<td>26</td>
<td>27</td>
</tr>
<tr>
<td>s.d.</td>
<td>6</td>
<td>5</td>
</tr>
<tr>
<td>range</td>
<td>18-37</td>
<td>19-35</td>
</tr>
</tbody>
</table>

*Procedure for the identification and invitation to participate for women with CAH*

The CAH population was identified in one of two ways. The hospital to which the children were referred as adults was identified from the paediatric hospital case-notes. Letters explaining the study and asking for agreement for contact to be made with the patient were addressed to the endocrine consultant at the adult hospital. On receipt of a positive reply, a letter of invitation to be part of the study was sent to the patient, together with a consent form and a pre-paid reply envelope (see Appendix I for copies of these letters). Where the medical notes did not record the continuing management hospital, information was sought through the National Health Service Central Register, and their procedures for
contact were followed. On return of the acceptance form agreeing to participate, arrangements were made by telephone for interview. This gave an opportunity for further explanation of the research to be given if required. Written confirmation of the date and time of the interview was sent one week prior to the appointment date. Where the patient returned the form indicating they did not wish to participate in the study, no further contact was made.

**Procedure for the identification and invitation to participate for women with diabetes**

The comparison group of women with early-diagnosed diabetes was obtained from two London teaching hospitals which held regular diabetic clinics. Although diagnosis at a very young age was the ideal requirement, discussion with the endocrinology consultants at both hospitals indicated that diagnosis before eight years was the youngest age of onset that would provide an adequate sample size.

For both hospitals the inclusion criteria were identical to those for the CAH group, however the method of identification and recruitment differed. One provided a list of female diabetic patients who fulfilled the criteria and were judged suitable by the responsible consultant; and a letter of explanation and invitation, with the consent form was sent to these women. The second hospital required the researcher to attend the relevant hospital diabetic outpatient clinics to identify from the hospital notes women who fulfilled the criteria. The identified women were approached by the researcher during their routine clinic visit and given a verbal description of the study, the letter of invitation, and the consent form to be returned by post when completed. For both methods, on
receipt of agreement to participate, arrangements for interview were made by telephone with written confirmation sent one week prior to the appointment date. No further contact was made when a returned form indicated a refusal to be part of the study.

**Procedure for non-respondents**

All women with CAH and those with diabetes who did not return the acceptance form within three weeks were sent one further invitation to participate (see Appendix I).

**Data collection and analysis**

**The collection of data from medical records**

Ideally the medical records would have provided a basis for addressing two interacting issues: whether there was a significant difference between the historical and current characteristics of the conditions, and whether there were significant differences between characteristics (historic and/or current) of those women who were interviewed and those not. To address both of these issues it was necessary to review the historic (paediatric) and the current (adult) medical records for each group.

Attempts to conduct this review were frustrated by two areas of difficulty. Firstly there was a lack of accessibility of the relevant records. In respect of the women with CAH some of the adult records were not available as adult consultants or general practitioners failed to respond to enquiries. For women
with diabetes the paediatric medical records could not (in almost all cases) be accessed because their current records did not include details of the relevant paediatric hospital. Secondly the contents of the adult records were such that they provided virtually no information relevant to the issues of the present study and no basis for comparability with the paediatric records.

As a result of these difficulties it was not practical to consider the issue of the comparison between adult and paediatric characteristics. Furthermore the comparison between those interviewed and those not interviewed could be made only on the basis of the paediatric medical records of the CAH women. The procedure for this comparison is outlined below and the results are reported in a later section.

The required information on the paediatric status of the CAH group was recorded on data sheets (see Appendix II). The information covered the following areas:

Diagnosis: age at, level of genital abnormality, and presence of salt loss;

Operations: age at and type

Related physical Characteristics: presence of cushingoid features, muscular habitus, hirsuteness;

Secondary sexual characteristics: age and development of menstrual function, breast and pubic hair;

Growth and Development: height and weight, comments made about psychosocial problems including referral to psychological services.
The paediatric information on women with CAH who agreed to participate and those who did not were statistically compared for age at diagnosis, age at and number of surgical operations, level of genital abnormality at diagnosis (Prader, 1954), and referral to psychological services. As the groups did not significantly differ on any of these, the interviewed group of women with CAH appeared to be representative of the paediatric sample. Details of the above data and analysis are given in Appendix III.

Prader’s (1954) classification is based on the level of genital abnormality at birth and has five categories that range from 1 (normal female genitalia) to 5 (fully masculinised penis and fused labia without testes). Table 2 shows the distribution of the levels of masculinisation at diagnosis and the types of surgical repair for the CAH women who were interviewed. On this criterion, the sample represents a group of severely masculinised women, with clitoridectomy (removal of the clitoris) and vaginoplasty (opening of the vagina) the most frequently performed surgery.

TABLE 2: Levels of severity of masculinisation at diagnosis and types of surgery for interviewed CAH women

<table>
<thead>
<tr>
<th>Prader category</th>
<th>Clitoral recession</th>
<th>Clitoridectomy</th>
<th>Vulvoplasty</th>
<th>Vaginoplasty</th>
<th>Vaginal dilation</th>
</tr>
</thead>
<tbody>
<tr>
<td>2/3 (mild)</td>
<td>1</td>
<td>3</td>
<td>1</td>
<td>4</td>
<td>1</td>
</tr>
<tr>
<td>N = 5 (28%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4/5 (severe)</td>
<td>1</td>
<td>12</td>
<td>4</td>
<td>10</td>
<td>1</td>
</tr>
<tr>
<td>N = 13 (72%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>TOTALS</td>
<td>2</td>
<td>15</td>
<td>5</td>
<td>14</td>
<td>2</td>
</tr>
<tr>
<td>N = 18*</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*Data not available in medical records for one woman.
The interviews

The role of the interviewer in qualitative research

Research, the aim of which is the understanding of underlying assumptions and meanings placed by individuals on their experiences, necessitates the provision of an atmosphere at interview that enables the exploration of experiences in an insightful, open-ended way. The objective of the interviews was therefore, two-fold: to allow the interviewees to explore and describe areas considered by them to be important, and to cover homogeneous topic areas with all interviewees so as to allow comparison of similarities and differences between the women, and between the groups. It has been suggested that such an atmosphere is best provided by an interviewer who shows the qualities of warmth, empathy, genuineness and understanding (Truax and Carkhuff, 1967) and this requirement in the current study was particularly important as some of the topic areas were both sensitive and intimate. The role and status of the interviewer is complex in a situation with these requirements. The traditional paradigm of the scientifically respectable interview appeals to such values as objectivity, detachment, and status hierarchy as the essential components. Disregarding these requirements has been considered to result in biased data - biased because of the possible communication of the views, attitudes and values of the interviewer to the interviewee.

This bias becomes particularly acute when the interviewer is perceived as an 'expert'- someone who is perceived to have special knowledge and status. In the study reported here, it was extremely likely that the interviewer would be seen
as a member of the hospital consultants' team and thus as a status figure. Hollway (1989) and Oakley (1990) have both argued that this problem is not best resolved by remaining silent; lack of information in such a situation serves to increase the possibility that the assumptions of the interviewee will be consolidated. These authors propose that in qualitative research, where the target data are subjective aspects of experience, hierarchical power relationships should be avoided so as to reduce the probability of such bias, and that a democratic position should be adopted. This position entails the interviewer becoming part of the research rather than retaining the detached role advocated by traditional interviewing theory. Certainly it became clear at the first pilot interview that a position of detachment was unlikely to produce the type of information required. Information presented as 'factual' ('Yes, I went to the hospital three times a year, and it was alright, the doctors were kind.') was given rather than that which allowed an understanding of the perceived meaning of the visits. It must be recognised that in the case of many of these women, it was misleading to believe that answers to questions or clear descriptions of feelings or experiences were readily available to them. Frequently the women with CAH had never discussed their experiences with anyone before the interview, and as Weedon (1987) argues the verbal description of experiences is an important factor in the interpretation of those experiences.

The interviewer has also to avoid biasing the discussion by providing verbal or non-verbal cues that may be perceived as an indication of the importance of any particular content of the discussion. The selection by the interviewee of what was appropriate information is likely to be affected by the perceived demands of
the interview situation (Cannell and Kahn, 1968). The necessity for the interviewer to catch not only the words but also their meaning for the speaker - what was said and what was meant- all indicated that a participating role for the interviewer was most appropriate. The techniques which were used to encourage both of these requirements - the selection of important topic areas, and the valid interpretation of the meaning of the women’s statements will be further discussed in a later section of the report.

The selection of interview topics

The point has already been made that it was considered essential to both allow the interviewees to discuss areas of importance to them, and to establish some consistency of topics across individuals and groups. Although the biological causes of the two endocrine disorders - diabetes and CAH - are understood, the factors affecting psychosocial outcomes are ill defined. The starting point for topic selection arose from a theoretical approach related to the recognition that a complex interaction exists between the individual, the immediate social context and the cultural environment.

This approach was based upon an ecological model. The ecological integrational model of Belsky (1984) was proposed in relation to child abuse, but can be seen to have wider relevance. In this model, outcome is seen as multi-determined by factors within the individual, the family, and the culture, with each of these nested within each other. The central factor was labelled by
Belsky as 'ontogenic development' and described the skills and characteristics which the parents brought with them into the family setting. This factor was seen as nested within the 'micro-system' - the immediate context - and included the characteristics of the child, together with the marital relationships, problem solving strategies and role performances of the parents. These in turn were nested in the exo-system, construed as the social structures, including friendships and social class, that impinged upon the immediate setting. The final system was the macro-system and this referred to cultural values and beliefs. This type of model provided a framework to identify and relate interview topic areas.

From the broad model discussed above and the research literature on the demands of chronic conditions, an outline of possible interview topics was constructed. The main topic headings proposed are presented below and full listings are given in Appendix IV.

1 Education
2 Employment
3 Chronic condition
4 Psychological/behavioural problems
5 Social activities
6 Friendships
7 Dating experiences
8 Sexual experiences and behaviours
9 Other social relationships
10 Attitudes towards parenthood
These topics represented a starting point for the interviews. Topics were amended, dropped or developed when their level of importance for the women emerged during the early stages of the study.

**The interview procedures**

*Location of interviews*

The difficulties discussed above that related to the interviewees' possible perception of the interviewer as a member of the medical care team resulted in the decision to conduct the interviews in the women's homes. It was hoped that greater freedom of discussion and exploration would be achieved in the women's own environment than in the medical atmosphere of the hospital. Consequently home interviews were arranged for all participants with the exception of one in each group. One of the women with CAH was at the time of interview, living in her van having been evicted by her mother from the family home. The 'live-in' partner of a woman with diabetes was unemployed and she did not feel he would be co-operative about leaving their home for the length of the interview. These two women were seen at the hospital.

*Setting the interview ground rules*

Very few of the women in the study were likely to be aware of what might be called 'the rules of the research study game'. They therefore would have very little idea of what would be required of them, what kind of information would be appropriate, or at what depth it was to be offered; the rules of social conversational skills would not be appropriate in this situation (Bernstein *et al*, 62)
Following the suggestions of Hollway (1989) and Oakley (1990) that it was functional for the interviewer to become part of the research rather than a detached recorder, the interviewees were given information on the rationale of the study and its possible implications for future management of the condition. Some information about the training and background of the interviewer was also given and for those with diabetes, this included that the interviewer was diabetic. The independence of the interview from the medical clinic management team was stressed and the confidentiality of all information was emphasised. Permission was requested to tape-record the interview, and in all cases this was willingly given: it was the impression of the interviewer that the recording did not affect the discussion.

Oakley (1990) discussed the high incidence of questions asked by her respondents during the interviews for her Transition to Motherhood project (1979); the largest proportion of these were requests for information about medical and organisational procedures and aspects of baby-care. In the current study informational questions were also frequently asked, particularly from those women with CAH although it had been made clear to the women that the interviewer had no medical training. Members of the public do not see medical experts as the only legitimate source of advice about health issues (Scambler and Scambler, 1984). As will be discussed later, the women with CAH did not have easily available sources of information about their condition so it was not surprising that they took the opportunity to ask questions. Conventional interviewing manuals advise that such questions should not be directly answered, and include appropriate techniques for dealing with them (Goode and
Hatt, 1952). However, the promotion of rapport, especially important in relation to the intimacy of some topic areas, seemed not to be best served by prevarication or denial of these concerns of the women – indeed, the concerns underlying many of the questions were an important areas of interest of the study. Questions were therefore acknowledged, explored and, if appropriate, the suggestion made that they would be discussed later. Care was taken to ensure a return to any such raised issues at the end of the interview.

**The interview format**

The interview sessions lasted between two and a half- and three and a half-hours. The necessity of allowing the interviewee freedom to identify their own important experiences and issues dictated a flexible format. The introduction of topic areas were not formally phrased but were presented in a language and style appropriate for that of the respondent (Watts, 1983). The specific sequence of topics as given above was planned to allow a natural progression from neutral to intimate, and past to current areas; however, natural changes of topic within the interview were explored as they occurred. In the nature of qualitative research information that related to topics other than that being discussed frequently occurred. These leads were followed, and the topic list was mainly used as a check to ensure that interview direction and consistency of topic areas had been achieved.

**Interviewing techniques**

Sensitivity to the verbal and non-verbal cues indicating a need for clarification and elaboration was maintained throughout. The interviewing techniques of probes and reflection were used to ensure the validity and accuracy of the
interviewer's understanding. The use of open-ended questions was helpful in producing reports of feelings (Hopkinson, Cox and Rutter, 1981).

At the end of the interview, the women were asked two further questions. Firstly, they were asked if there was anything else that they would like to add, any experiences or thoughts that had not been covered. Frequently women repeated views and comments made during the discussion, but no new areas were raised. Secondly, they were asked if any part of the interview had been distressing or confusing and a few women in each group mentioned some embarrassment during the discussion of sexual topics. All the women with CAH commented that they had enjoyed talking about their experiences relating to their condition. It had frequently been their first opportunity for such discussion. Although the women with diabetes expressed considerable interest in the interview and the research these comments did not convey the same depth of impact as those made by the CAH group. It is likely that for those with diabetes, many more opportunities have been provided for them to share their condition related experiences. There were some requests for information about the results of the study and these were noted.

The analysis of the data

There were a small number of areas where it was appropriate to collect quantitative data. Any nominal or ordinal level data collected was analysed by the appropriate non-parametric statistic (Siegal, 1956).
The principle of the hypothetico-deductive method upon which quantitative methodology is based, requires that a hypothesis is formulated on the basis of theory and then, quite independently, data is collected which will test this hypothesis (Silverman, 1972). Qualitative research inductively derives models from the study of the phenomenon under investigation (Glaser and Strauss, 1967; Strauss and Corbin, 1990). The qualitative methodology of grounded theory provides an alternative approach (Glaser and Strauss, 1967) and here, the models and theories are built from within the collected data by the use of a systematic set of procedures.

The analysis of any data should be an open, public process. This is perhaps particularly important in grounded qualitative research where the study can only be evaluated accurately if the reader can assess the appropriateness of procedures used for the data organisation and interpretation (Strauss and Corbin, 1990; Yin, 1994). In addition, it has been suggested (Hollway, 1989; Stevenson and Cooper, 1997) that qualitative research necessitates the use of 'researcher reflexivity'. This process is concerned with making public the contextual factors which have influenced the researcher's arrival at particular constructions. As Hollway (1989) has discussed, there are an infinite number of possible accounts of experience; which account is chosen will largely depend on the contextual influences operating on the researcher.

In order to analyse the women's accounts of their experiences, each of the thirty-six taped interviews was transcribed, the analysis being based on these full transcripts (see Appendix V for examples of transcribed CAH and diabetic...
Building a theory from a collection of data involves processes of breaking down, examining, conceptualising and relating and then re-assembling the items of data so as to develop an understanding of the women's experiences that may contribute to more effective management of the medical conditions under consideration.

Figure 1 lists the components of the analysis carried out on the data obtained from the interview transcripts.

As Figure 1 shows, there were a number of analytical components applied to the interview data. The foundation of the analysis is the detail of the interview data. From this detail a hierarchical structure has been built. At each level of this structure collections of related phenomena are combined to form broader components that move from elements through dimensions and concepts to the broadest level which has been labelled categories. In the initial stages of the analysis any categorisation was considered provisional as changes in perceived relationships may occur at any point during the process. The final component
represented in Figure 1 refers to the identification of relationships between categories which allows the construction of a unified theory by enabling a move to be made from description to theory building. This move constitutes a process that allows the researcher to 're-integrate the whole person from the jig-saw of parts created by modern scientific medicine.' (Koeske, 1980.p6). An extract from the work of this analysis of the data is given in Appendix VI.
RESULTS AND DISCUSSION

The separation of method and theory, and the resulting sections of Results and Discussion usually used in quantitative research reports, is not so appropriate in qualitative research. The three activities identified by Miles and Huberman (1985) as characterising the analysis of qualitative data - summarising and identifying themes, organising data and drawing conclusions - occur concurrently throughout the research project. It is therefore more appropriate to combine the results and discussion in reporting qualitative studies, and this convention is followed in this thesis.

THE PROPOSED STRUCTURE OF CHRONIC CONDITION EFFECTS

The overall aim of the research project was to gain an understanding of the experiences for two groups of women, of living from childhood with a chronic condition. Such a broad aim may be construed as comprising a number of components. Two components, the identification of any such experiential effects and some of the mechanisms through which effects might exert their influence, were considered central to the research. Such aims needed a dynamic structure, one that could incorporate new information and accommodate relationships between parts of the structure as they emerge.

Such a structure would not provide a hypothetical model to be tested, but an internally consistent and viable understanding developed from the women's
descriptions of their experiences. This structure must be comprehensive, contain within it the areas and topics discussed during the interviews, be grounded in the data and systematically constructed.

**FIGURE 2: The categories and their inter-relationships**

Figure 2, outlined as a process in Figure 1, represents both the categories identified from the data, and relationships between them. It is proposed that the category 'Demands of chronic conditions', through the learning of 'Strategies', may result in the development of a 'Skeleton-in-the-cupboard'- a belief that some characteristics and experiences must be kept hidden - and that this may have far-reaching 'Psycho-social effects'. An individual's cognitive constructions about their experiences are likely to be affected by the contextual events and beliefs in which they are set, and therefore the overall effects of culturally existing 'discourses' must be considered.

The construction of such a model relied upon the identification in the data of the concepts, dimensions and elements that clarified, added depth to, and supported the suggested relationships between the categories. These components will be examined in the following sections.
The category 'Demands of the chronic conditions': concepts and dimensions

The published literature suggests that chronic conditions may make both social and psychological demands on the individual. For example Strauss et al (1984) identified a number of aspects of chronic illnesses which were construed as making severe demands on the individual. These aspects were mainly concerned with medical regimens, social interactions and practical issues of attending for medical consultations.

Research has explored the nature of encounters between health professionals and their patients, particularly in relation to the explanation of medical terminology and treatments. Issues which have emerged from such research include the effects on health care of the failure of health professionals to take into account the views of the patient, be they about the causes or treatments of the condition (MacIntyre and Oldman, 1985). These issues were identified in the data and form concepts of the category 'Demands of the chronic conditions' (see Table 3).

TABLE 3: Concepts within the category ‘Demands of chronic conditions’

<p>| | |</p>
<table>
<thead>
<tr>
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<tbody>
<tr>
<td>Attending clinics</td>
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<tr>
<td>Understanding the condition</td>
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<tr>
<td>Long term regimens</td>
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<td>Value of the consultations</td>
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</table>
The concept of 'Attending clinics'

The necessity of life-long clinic visits for medical consultations was an important and demanding experience for both groups of women. This concept was supported by a number of dimensions relating to their recall of attending paediatric clinics, and to their feelings about current visits. These dimensions are presented in Table 4.

TABLE 4: The concept 'Attending clinics': dimensions and elements

<table>
<thead>
<tr>
<th>Dimension 1 - emotional response (to paediatric clinics)</th>
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<tbody>
<tr>
<td>Dimension 2 - the doctor knowing me</td>
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<tr>
<td>Dimension 3 - what's it all about?</td>
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<tr>
<td>element of feeling uninformed</td>
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<tr>
<td>element of poor communication</td>
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<tr>
<td>element of doctor's approachability</td>
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<tr>
<td>Dimension 4 - doctor's knowledge</td>
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<tr>
<td>Dimension 5 - user-friendly hospital</td>
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<tr>
<td>element of medical procedures</td>
</tr>
<tr>
<td>element of privacy</td>
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<tr>
<td>element of hospital organisation</td>
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</table>

The dimension of 'Emotional responses'

The first dimension, the 'emotional response', emerged only in the recall of visits to paediatric clinics, and was comprised of both positive and negative aspects. Although the negative aspect, fear responses for children visiting hospital is documented in the literature (Robertson and Robertson 1971, Hodapp, 1982, Brewster, 1982, Koomen and Hoeksma, 1993), in the present study the proportion of those recalling this emotion was dissimilar for the two groups of women. Of those with CAH forty one per cent recalled their visits as a negative emotional experience:

*It scared me very much [CAH patient No.01- (C01)]*
"I used to hate it (C09)"

and the same proportion, as a positive experience:

"I liked going, because they helped you- it was better than going to school (C10)"

In contrast, the majority of the diabetic group reported only negative emotional memories (71 per cent):

"(I was) frightened [Diabetic patient no. 21. (D21) ]"

"(It was) terrible (D02)"

with twenty per cent recalling their visits only as positive experiences:

"(I used to) enjoy them (D01)"

"(It was) very friendly, very relaxed, it was like a play-day (D14)."

A small number of women in each group recalled both positive and negative memories (18 per cent of those with CAH, 9 per cent of those with diabetes):

"It was terrible, but he was a nice guy (D02)"

"Somedays I hated it and some days I loved it 'cos I used to get a present from my mum as well. (C41)."

The group difference then, occurred mainly in the higher recall of negative emotional responses by those women with diabetes. At this point in the analysis it was not possible to identify factors of influence to account for this difference.

Memories of visits to outpatient clinics during childhood were complex and covered a number of other dimensions, however these dimensions were also present in the discussions about current clinic experiences. The extent to which
the recall of childhood memories are influenced by attitudes towards current experiences is not known, however the aim of the present research was not to establish the 'truth' of the women's recall, but to develop an understanding of their existing cognitive constructions about their experiences. The point of interest is what meanings these women have made of the need to attend hospital outpatient clinics at approximately three monthly intervals as children, and twice a year as adults.

The similarity of the remaining aspects of the recall of childhood memories, and the feelings about adult clinic visits, suggests that it is meaningful to combine the discussion of these. The concept of 'Attending clinic' also comprised four other dimensions that of the 'doctor knowing me', 'what's it all about?', the perception of the 'doctor's knowledge', and of attending a 'user-friendly hospital'.

**The dimension of 'Knowing me'**

An important dimension of the concept about visiting paediatric hospital clinics was the clear and similar description by both groups of feeling they were not the focus of communication, of not feeling relevant to the event:

- *The doc spoke to Mum- there was no point in being there* (C09)
- *They talked to others, not you, behind your back* (D21)

These statements were accompanied by those which suggested they did not feel confident about their doctors' interest in them as individuals, a feeling that applied to both paediatric and to adult clinic experiences:

- *There wasn't a doctor that really cared about you* (C03)
I feel they can't be bothered (C41)

No one knew my history or me as a person (C01)

They don't know you (D18)

I don't feel the doctor understands me, they have washed their hands of me (D24)

These two positions, that of not being addressed directly by the doctors, and the doctors' perceived lack of knowledge about them as individuals, resulted in the expression of strong feelings of not being a valuable patient.

The dimension of 'What's it all about?'
The third dimension, 'What's it all about?' perhaps reflected the most important reason for attending outpatient clinic. It was concerned with obtaining advice, help and guidance from an expert in the relevant medical condition who holds medical responsibility for the management and treatment of the condition. Three related elements emerged here, with very similar positions taken by individual women on all three. The elements related to the effectiveness of doctor/patient communication. Poor communication resulted in: feeling uninformed, having the impression that the doctor was unable to explain things in a way that could be understood by the patient, and the perception of the doctor being unapproachable.

The element of feeling uninformed
Lack of information from the doctors was reported by both groups, and was particularly strong for those with CAH:

They should explain better, take more time and say what they are going to do and what it's all about (C29)
If they had told me what the problem was! I wondered what they were going to do and why. (C09)

(They) didn't tell me what they were looking at. (C07)

No one told me why X-rays, or explained the reasons for things. (C52)

The doctors didn't tell me very much. (D04)

I wasn't told anything (D29)

The element of understandable communication

The lack of explanation was associated with the women's impression that the doctors did not have the necessary communication skills. This failure of the doctors to convey understanding may be attributed to communication being formulated at an inappropriate conceptual level and using inappropriate language. The outcome of these communication problems was a deficiency of understanding by both patients, and as reported by those interviewed, their parents, of the medical concepts and reasons for treatments and tests:

I didn't have any feedback from the blood tests- they don't tell me why, just say "We're changing it". (C07)

I've asked the doctors, but they don't seem to explain in a way that you can understand. (C47)

The job is about communication and very few are able to communicate- no interpersonal skills. (D24)

Although both groups remembered such a lack of communication skill in the doctors at the paediatric clinics, the groups were dissimilar in their views of the current doctors. The diabetic women reported that their current doctors were able to:

'explain things a lot more to you- they treat you like an adult'. (D21).
The CAH women however, retained their perception of being faced with explanations, if there were any, at a level that they could not understand.

The need for improved communication between the providers of care and those who receive it is well documented, but the research suggests that there is still some way to go before satisfactory levels are attained. The Audit Commission (1993) found that patients and relatives frequently held the view that they did not get enough information and found asking for information difficult. Strong (1979) analysed four types of communication patterns and in National Health Service clinic consultations, the most frequent was the type designated as bureaucratic. In this type, both the doctor and the patient are polite, courteous and avoid conflict; interaction appeared to be based on the presumption of the doctor as technically authoritative, and although the patients frequently disagreed with the formulations of the doctors, open disagreement was rare. Such a style of interaction during a consultation would seem to perpetuate communication difficulties and to increase dissatisfaction with the visit.

These doctor-patient communication difficulties were identified in the study reported here. The issue of the tendency to agree with the doctor was related to the perceived level of the doctors’ approachability.

The element of the doctor’s approachability

The final element in this section is that of the perceived approachability of the doctor. The women with CAH were clear in expressing their belief that it was difficult to ask their medical advisers for information about their condition, or
their treatment, and therefore to obtain answers to their queries and concerns. Whilst children may be expected to find it difficult to ask questions during clinic visits, it remains less understandable that parents who accompany the children to the hospital should find such tasks difficult. The women with CAH, in connection with the adult clinic visits, felt the issue of approachability very strongly:

"You just can't go in and talk to your doctor like that- it's 'next please'." (C01)

"You feel you can't ask." (C08)

"I couldn't sit down and talk to Dr.X. (C04)

"I don't like to ask. (C36)

Comments such as these were very frequently made by the CAH group, however the diabetic women were more comfortable in approaching their current medical advisers. This difference between the groups could be partly due to a specific condition effect; the sensitive and socially embarrassing nature of CAH may have added to the general reticence of patients to ask their doctors for test results and information about their medical status. This issue will be further discussed below as it re-occurs in relation to other categories.

The dimension of the doctor's level of knowledge

When speaking about their experiences during visits to both paediatric and adult clinics, the CAH women expressed doubts about their doctor's level of knowledge about their condition. About one third of the group felt the level to be very low, and this belief was particularly strongly expressed by the women who also believed the doctors did not see them as valuable patients:
He never seems to know the answers. My G.P. doesn't know anything either. (C33)

They didn't know what was wrong with me. (C47)

I used to blame the doctor cos I didn't get better. (C15)

In summary, the women with CAH were likely to hold negative positions on the three dimensions relating to adult clinic visits. They did not believe they were known by their doctors, they did not know what the visits were all about, and perceived their doctor's level of knowledge to be low.

Women who held these negative beliefs, also expressed dissatisfaction about organisational and procedural elements of the outpatient clinic visits- visits which to them were made to a hospital that they did not feel was 'user-friendly'. The elements identified in the women's statements that contributed to this dimension will now be discussed.

The dimension of ‘User-friendly hospitals’

The element of medical procedures

For the CAH women, two procedures, vaginal examinations and blood tests, were vividly recalled as being distressing during their visits to paediatric clinics:

(the doctor) was so brutal- this is the closest I'm going to get to rape. (C09)

the visits I didn't enjoy 'cos they had to inspect me. (C07)

I didn't like them meddling about with me (C04)

[They] put a wooden thing in your vagina, painful, like being physically abused (C41)
The medical procedures referred to here were vaginal examinations. These comments must be considered in the context of the two preceding dimensions. Most young women during vaginal examinations undergo some level of distress. However the level for those with CAH was likely to have been increased by their perceptions of not being known or valued, being treated by a doctor whom they felt was uninformed about their condition and their failure to understand the reasons for medical procedures.

For those with CAH there was also a dislike of blood tests:

'Every time he saw me, he had a needle in his hand'
(C03).

It is of note that although the women with diabetes regularly have blood tests during their visits to clinics, they did not raise this as an issue. This comparative lack of significance could be due to a number of factors; those with diabetes frequently perform blood tests at home, and therefore such tests, although disliked, have become everyday events for them, and therefore not worthy of comment at interview. It was also reported above that the diabetic group felt more informed about the reasons for tests, and was more comfortable about approaching the doctors for information. Understanding, the possession of information and the perceived ability to acquire these are factors known to decrease anxiety and increase tolerance for pain and discomfort (Marlatt and Gordon 1980).
The element of privacy

Many negative statements of those with CAH were focussed on the lack of privacy during medical procedures:

*Big pile of doctors all around I hated it (C28)*

*You go to see your doctor, not students looking at you (C03)*

*There you are on the bed, they are all looking at you - they stare at you (C04)*

*I hated the examinations, lying there on the bed with no clothes on (C33)*

Again, this was not a strong issue for the women with diabetes, although there were occasional critical comments about the public disclosure of their weight during clinic visits. The medical examinations and procedural requirements for children with diabetes are very different to those for CAH patients; undressing and the examination of intimate body areas are seldom required for diabetics. These different group experiences may account for the differential importance of privacy, and the absence of issues of privacy in connection with adult clinic visits supports this explanation. By adulthood, procedures at CAH clinics rarely have a genital focus, and therefore it is not surprising that privacy was not an issue raised. The position of the women on the dimension of privacy appears to be 'specific' to the condition, with the requirement of intimate physical examinations during paediatric clinic consultations a central factor for those with CAH.

The elements of medical procedures and of privacy appear to be important issues in the attitudes of the women to the hospital visits. Other elements of
their visits connected with the hospital organisation were mentioned as unsatisfactory and as sources of distress.

The element of hospital organisation

For both groups the unsatisfactory aspect of hospital organisation most frequently mentioned was the long waiting times to see the doctor. About a third of each group recalled this as a distressing aspect of paediatric clinic visits and this increased to half of the groups for their attendance at adult clinics:

Sometimes I feel I've been there years, waiting (C52).

A further organisation problem, particularly for the CAH group, was the frequent changes of doctors. The sensitive nature of the condition, the necessary re-capping of medical history, treatment, and general family and social details required when consulting a new doctor perhaps accounted for this emphasis. The following quotes clearly describe this problem and illustrate how this adds to the women's feelings of not being a valued patient:

[a new doctor] every five to six months. No one knew my history or me as a person. It was difficult to establish trust. (C01)

I get attached to a doctor and "Bang!" they've cleared off. (C03)

We always saw the same doctor, this gave you confidence. (C34)

Research into issues of communication in medicine has produced two recurrent findings: the dissatisfaction of the patients with their doctor's communication skills; and the dissatisfaction of many doctors with their patient's compliance with advice and treatment (Fletcher, 1973; Bennett, 1976; Quine and Rutter,
A trusting relationship with the doctor is held to be crucial for both of these issues and it seems that frequent changes of doctor hinder the development of this. When the chronic condition has intimate aspects, as in CAH, the development of a trusting relationship with the doctor appears to be of extra importance. Frequent changes of doctor will also increase the belief that the doctor does not know the patient as an individual, as would be true, and research findings suggest that this would effect the level of perceived satisfaction with the consultation.

In a major study of eight hundred paediatric consultations Korsch and her colleagues (1968) showed that a positive value, or satisfaction with the encounter, was dependent on three characteristics of the doctor conducting the consultation. These were the doctor's friendliness, ability to understand the patient and their parents' concerns, and communication skills. In a cognitive model of doctor-patient communication, Ley (1988) suggested that satisfaction with a consultation depends upon the level of the patient's understanding about the medical material given to them by the doctors; failure to understand will result in dissatisfaction. In a study conducted in the National Health Service (Hardy et al, 1996) the three identified components of patient satisfaction were: the process of care, improvement in and understanding of health, and psychological well-being. The highest loading variable for all three of these predictor components was the level of clarity of information provided by hospital staff. The findings of the current study, with added emphasis on the effects of sensitive and intimate aspects of CAH, are in line with this research.
and support the important role of good communication in patient satisfaction with medical consultations.

**The concept of ‘Understanding the conditions’**

The category of the ‘Demands of chronic conditions’ comprised four concepts (see Table 3). The first of these – ‘Attending clinics’ has been considered above. The second concept, the ‘Understanding the conditions’ comprised two dimensions; the women's level of knowledge about their condition, and their sources of information (Table 5).

**TABLE 5: The concept of 'Understanding the conditions': dimensions and elements**

<table>
<thead>
<tr>
<th>Dimension 1 - Level of knowledge</th>
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<tr>
<td>Dimension 2 - Sources of information</td>
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<tr>
<td>element of - hospital clinics</td>
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<tr>
<td>element of - parents</td>
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<tr>
<td>element of - literature</td>
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<tr>
<td>element of - self-help groups</td>
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</table>

**The dimension of level of knowledge**

It is difficult to obtain a comprehensive, reliable and valid measure of knowledge about a medical condition. Research has been concerned with devising measuring instruments for patient's knowledge about diabetes (Dunn, 1987) which fulfilled these aims, but there are difficulties in the comparative use of such instruments as published scales vary both in their content and in terms of the depth and breadth of the required knowledge. In addition, changes in technology and/or the advised management regimen result in scale items becoming redundant or ambiguous.
For the research reported here, involving both diabetes and CAH, a number of additional factors increased the difficulties. These included the limited time available for data collection, the acceptability of the demands made on the women who agreed to be part of the study, and the absence of any standardised measurement instrument for knowledge about CAH. Therefore the approach taken was to ask the women to 'please explain in your own words what is the problem that has meant visits to hospital over the years'. The women's statements in reply were scored against the views of three consultant endocrinologists. One paediatric and two adult consultants were asked to give the minimum amount of knowledge they would expect their patients to have about their chronic condition. Items of information were identified and combined across the views of the three consultants, and the resulting collection of statements were scored as 'bits' of information. Each 'bit' of information identified in the women's statements was given a score of one point. The criteria show that a greater number of 'bits' were required for CAH knowledge than for diabetes.

The combined criteria for basic levels of knowledge about CAH and about diabetes were:

**CAH**: A condition where the adrenal glands do not work properly. They produce androgens, which are male hormones. I have to take steroids to suppress the adrenal glands and when I was a child I had to have surgery because the adrenal glands has masculinised my genitalia before I was born. I had to take steroids in childhood so I had normal growth and physical development. (12 'bits' of information.)
Diabetes: A condition where my pancreas does not work. As a result I need treatment with insulin. Because it is not possible to mimic my own insulin production I have to control my diet to balance the insulin. (5 'bits' of information.)

Table 6 gives the obtained knowledge scores for both groups.

<table>
<thead>
<tr>
<th>GROUPS</th>
<th>MEDIANS %</th>
<th>MODES %</th>
<th>RANGE %</th>
</tr>
</thead>
<tbody>
<tr>
<td>CAH</td>
<td>8.8</td>
<td>8.0</td>
<td>0-33</td>
</tr>
<tr>
<td>DIABETIC</td>
<td>60.0</td>
<td>60.0</td>
<td>20-80</td>
</tr>
</tbody>
</table>

The results for those with diabetes suggest that knowledge was closely clustered about the centre of the range. The median and mode for this group was sixty per cent, with eighty percent of the scores falling in the centre and no scores in the lowest percentile. The remaining twenty percent of the scores were in the top percentile. Statements representing two different levels of knowledge are given below:

*It's a condition, I hate the word disease. The pancreas has stopped working, or it doesn't give enough insulin, so we have to give it a boost with injections, or tablets or diet. SCORE=3/5, 60% (D14)*

*Certain cells in the pancreas that produce insulin aren't working so I have to inject it. Diabetics have to work it out to balance the amount of insulin with the food they eat. Insulin digests the food. SCORE=3/5, 60% (D26)*

*It's a problem with the pancreas. Part of it produces insulin and that part doesn't, so you have to give artificial insulin. Insulin is a hormone that helps you to digest food and get energy from fats. You have a set amount of insulin and you have to eat to balance it. SCORE=4/5, 80% (D04)*

The scores for this group of diabetics therefore did not meet the basic level required by the endocrine consultants, the median and modal scores falling in
the second and third quartiles. This result is in line with the published research on knowledge levels for adults with diabetes (Dunn, 1987; Watts, 1983, May, 1994) where the amount known about the condition has been found to be unsatisfactorily low.

Whilst the low level of knowledge of those women with diabetes was of considerable concern, the results of those with CAH, however, were of exceptionally unsatisfactory low levels. Both the median and modal percentages were in the lowest percentile with only one score falling in the second percentile. The following quotes of the CAH group show very clearly this extremely poor understanding:

> An abnormality in the genital area/ caused from early birth /, right from the early stages of development, in the X and Y chromosomes, which meant that I had two Ys and one X (C01) SCORE=2/12,17%

> It's just a freak of nature. In the end, chromosomes give a miss here and there and I was that one in a million. I was born a boy but inside a girl/, I had to have an operation/ which made me the girl I should have been (C09) SCORE=2/12,17%

> My body doesn't make any salt and it's all to do with your glands (C10) SCORE=0/12,0%

> Hyperplasia means too much tissue, so I make too much and I take replacement therapy in order to reduce what my body makes already, some sort of cortisone. It's all to do with the cortex of the adrenal glands/ and not the medulla. (C34) SCORE=1/12,8%

> Basically a mix-up of genes between Mum and Dad - dirty genes. I don't produce any adrenalin hence I have to take hydro-cortisone/. And my Florineff is to do with the adrenalin. It's the chemicals, they are all inter-mingled. It's an adrenalin deficiency. (C37) SCORE=1/12,8%

> Besides the physical abnormalities, the adrenal gland/ pumps out hormones and there are different enzymes and they produce poisons, and another enzyme counter-acts
the poison, and all I produce is the poison. So the steroids I take are to put back in the body what I don’t produce. When you are born with it, you are born with partly the opposite sex and you have surgery. I don’t know why we are born like that, genes - that’s the only thing I know. (C41) SCORE=4/12, 33%

Both groups of women in the study had the condition from very early childhood and by the time of the interview some decades later it might have been expected that they would have a clearer idea about the nature of their chronic condition. The levels of knowledge, while in line with the published research on patient’s information about their medical condition, must be regarded as unacceptable especially as a very liberal scoring criteria adopted. A ‘bit’ was scored if the ‘idea’ of the relevant information was given; accurate or technical language was not required.

It is of course possible that the women possessed other important information about their condition. The aim here was not to establish the precise depth or breadth of knowledge, but to assess the extent of the level considered the basic essential by the medical consultants. A further important aspect of these results is that this measure of the knowledge levels of the CAH women supported their subjective beliefs about their understanding of CAH; they believed that they did not understand the nature, cause or management of their condition – and they certainly did not.

Information held by the women about their chronic condition, and an understanding developed from this information, may be a necessary but not a sufficient factor in following medical advice given to them by their medical
consultants. Such compliance with regimens is discussed later in this section on the demands of chronic conditions.

The dimension of sources of information

Information possessed by the general population about illness is most frequently obtained from sources other than their doctors (Scambler and Scambler 1984). The incidence of quasi-medical articles in the media, together with books and pamphlets on specialised subjects that are now available over the counter, offer easily accessible alternatives to medical sources. In addition, published research indicates that information from friends and relatives is nearly always sought, these opinions being taken as some kind of trigger for seeking advice and information from professionals. For example parents, partners, relations and friends, are perceived as available sources of advice and aids to understanding the episode of illness. Any of these individuals may know others who have the condition, or indeed they may have personal experience of the condition. Consultations with medical experts therefore, form only one component in the complex social network of information seeking activities, Scambler and Scambler (1984) reported a ratio of eleven lay consultations, usually involving a spouse or close friend, to every medical consultation. The interview data were analysed to identify the sources of information for both groups of women. Four source elements were identified: the hospital clinic visits, parents, published literature and self-help groups.

The element of hospital outpatient clinic visits as information sources

It might be presumed that regular visits to a hospital for consultation with a doctor who is an acknowledged expert in the relevant medical condition, would
be a major source of information for the development of understanding, however this was not so. The recall of the women’s experiences at paediatric clinics have already been reported and this included the impression that communication with the doctors was unsatisfactory:

*nobody has actually sat down and spoke to me about what's wrong.......A doctor should tell, say "this is wrong with you", and "This is what's going to happen"* (C33),

Similar perceptions about the adult clinics were reported by those with CAH, with the added impression that when any explanation was given, it was in a language that could not be understood:

*I've asked the doctors but they don't seem to explain in a way that you can understand. .....My G.P. couldn't understand when I asked him about not being feminine enough* (C47)

Although the women with diabetes recalled similar difficulties in information transfer during their paediatric visits, the comments of these women about their visits to adult clinics expressed general satisfaction with the doctors' explanations and answers to their questions:

*... over the years I think I've had the most help and the most explanations of things from the adult hospital. .....but I think it's two sided, I mean if you want to know, yes, over the years they have explained things that I wanted to know.* (D09).

This statement exemplifies another clear difference between the two groups of women: their attitude towards taking an active part in acquiring information about their condition. All the women in the diabetic group wanted to know more, and with the one exception of information relating to associated risks of medical complications, to understand about all aspects of diabetes. The women
with CAH were less assertive about asking, and more than half of the group did not want to know, they wanted to 'leave it to the doctors'. (C01).

Although for most people who become ill, information may be available through attendance at hospital or at their general practitioner surgeries, there are as discussed above, other commonly used sources. In addition to other individuals, many conditions are now in the public awareness and self-help groups may exist. For those with diabetes, it had been possible over the years to access most of these information resources, but for those with CAH this was not so. The issue of the willingness of those with CAH, and/or their parents to discuss the condition with others, will be dealt with in a later section, however the elements of the availability of parental information, literature, and self-help support groups will be examined here.

_The element of parents as information sources_

The following extract from a letter from a father of a young daughter with CAH, dated 1984, is taken from hospital records:

X's mother has never been told exactly what X had to look forward to in the future. X has suffered bouts of depression and long periods of anxiety because of her troubles. The doctors have always told her mother to explain to X what is wrong with her, that is beyond her as her mother has not been filled in either. It distresses me to think this is at least my third letter to various sources pleading to let me know what we can and cannot expect from X. X knows about her hormone imbalance but that's about all. What she must worry about can only be imagined-

1. why hasn't she started periods?
2. can she have children?
3. how much a problem or a possibility is this?
4. what does she tell a fiancée or husband?
I realise some of these aspects may or may not be in your field, but X is close to a breakdown caused by uncertainty.

It is essential therefore that we are given the whole story, including doubts and possibilities, on all aspects of X’s health. Not so much the immediate condition of each, but the ‘whens’, ‘hows’ and ‘ifs’.

Please, please help, thank you."

This is a strong plea, but the ignorance of the parents and the lack of any available source of understandable information were echoed in all the statements from those with CAH. Those women who had asked their parents, mainly their mothers, for information had received unclear and unhelpful information and frequently negative responses to their attempts to understand:

Mum? [she said] 'Don't worry about it, don't ask questions like that.' (C08)

Mum said, 'You've got this and you've got to live with it for the rest of your life.' She never really explained it to me, said, 'They can't do anything to help you, you have to take these tablets and if you are sick, have injections.' (C10)

She said 'You're like a diabetic.' (C15)

When my sibs were ill, I used to ask Mum, 'Have they got the same as me?' It was always, 'No-nobody's got the same as you.' I didn't understand why, the same parents, the same family, they hadn't got the same. (C33)

It is perhaps not surprising that the parents of those with CAH were unable to explain or answer questions about the condition. It was the recall of the interviewed women that understanding the doctor's communication was as problematic for their parents as it was for themselves:

*Mum doesn't feel they explained* (C29).
Those with diabetes however, recalled a different situation. Their parents were their major information source when they were children:

*Mum and Dad learned, they passed it on to me (D14).*

**The element of published literature as information sources**

Written information may be helpful as reference material about medical conditions. There are increasing numbers of books, pamphlets and information sheets available from such outlets as hospitals, chemists and general bookshops. Printed material aimed at all ages is certainly available for those with diabetes, and the British Diabetic Association (BDA) provides a considerable amount of information. This Association produces booklets on particular topics and publishes 'Balance' magazine that provides articles and discussion on a wide selection of topics aimed at all ages. Most of the women with diabetes had used these sources and in general found them to be informative and satisfactory:

'Balance' for the first four to five years. Dad and Mum bought me all the books they could find, loads of leaflets. I read all I could on it. (D29).

Such resources were clearly unavailable for individuals with CAH. Booklets had been supplied by some of the paediatric hospitals, but were recalled by the women as very unsatisfactory:

*She got a little booklet, it explains it, but all those long words! (C17)*

*Mother gave me some leaflets that they had given her when I was born, and I sort of read it, but it didn't seem to be about me. (C07).*

**The element of self-help groups as information sources**

The final resource to be discussed here, self-help groups gave rise to similar findings as those on written materials. The majority of those with diabetes had
been to BDA camps as children, and some of them had continued to attend
during their teens, as helpers. Fewer women were members of local diabetic
self-help groups, but all were aware of their existence. Not only were there no
such camping or self help group opportunities for those women with CAH, but
in addition, none of them were even aware of anyone else with the condition.

It would seem that a number of issues of communication have been identified
that may have considerable outcome effects for those with CAH. The
experience of the failure to understand about their condition or the rationale for
the treatment regimen; the difficulties of parents in explaining and discussing
what was wrong with them; the absence of any understandable available
literature; and the lack of any opportunities for discussion about their condition,
may have led to the cognitive construction by the women that their condition
was unique and unacceptable as a subject of discussion.

In summary, women with diabetes appeared to have very different experiences
to those with CAH in relation to information that may have helped them to
understand what was it was that necessitated medical care. Those with diabetes
had a number of resources: their parents, their doctors, published literature, the
media and television; self-help group opportunities and professionally organised
holidays provided them with multiple information resources. These multiple
resources, in addition to providing information, may also have encouraged the
women to develop an interest in discovering more about the chronic condition.
For those with CAH, the information vacuum not only may have impeded the
development of understanding, but also may have limited the extent to which the
women with CAH believed that their condition was either understood by others, including the medical professionals, or was an acceptable topic for discussion.

Both the conditions in this study are chronic and therefore by definition require life-long management. Management can involve a number of factors and may include taking medication, changes of behaviour, learning new skills and strategies, and the acquisition and understanding of information. Some of these factors have already been discussed; other issues reported by the women as important in their attempts to manage their chronic condition were identified and these will now be introduced under the 'Long term regimen' concept of the 'Demands of chronic conditions' category (see Table 3).

The concept of ‘Long term regimen’

The dimensions identified in the interview data relating to the 'Long term regimen' concept were: compliance with the recommended regimen, and views held about possible future changes of regimen (see Table 7).

<table>
<thead>
<tr>
<th>TABLE 7: The ‘Long term regimen’ concept: dimensions</th>
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<tbody>
<tr>
<td>Dimension 1 - Compliance</td>
</tr>
<tr>
<td>Dimension 2 - Future changes of regimen</td>
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The dimension of compliance

Compliance with the various aspects of a prescribed regimen may require high levels of motivation. Although the measurement of non-compliance is problematic, reports in the literature suggest the incidence of failure to adhere to
proscribed regimens is high (Ley, 1988; Roth, 1987). The underlying causes of non-compliance with a medical regimen are likely to be a complex interaction of factors in the lives of the individual, factors such as financial costs, the interference with social and/or psychological events, and the levels of skill required. The level of compliance may depend on the balance of such costs and the perceived benefits of improved health and prognosis.

The data of the present study suggested that the level of compliance of both groups was in line with the low levels reported in published research (Ley, 1988). The routine regimen for those with CAH consisted of taking tablets usually twice a day. Almost half of the CAH group spoke about times when they omitted taking their daily tablets:

Yeah, I quite often forgot now and then, but it doesn't seem to matter (C07)

I used to keep forgetting fairly often- my God I suffered because of it!! I once gave up taking them for three weeks- it took me two months to get back to normal.(C15)

I hadn't been taking enough 'cos I hadn't bothered seeing anyone, I neglected myself. (C34)

The most frequent reason given by those with CAH for this non-compliance was 'forgetting'. However, in line with the previous finding of low levels of understanding the rationale for treatment, almost three-quarters of the CAH group were unsure as to the efficacy of the prescribed medication. Some of the stated reasons for instances of the omission of medication appeared to be related to this low level of confidence; this was construed by the women as 'testing the tablets':

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I don't know what the side-effects of my tablets are, I wanted to find out how serious, how bad it could be [if I didn't take them]. (C15).

In addition to efficacy of the medication, those with CAH expressed some concerns about medication side effects. One side effect, attributed by the women to 'the steroids', was very distressing for those on medication for CAH. The side effect was the commonly seen increase in body weight, and the belief that this was due to the tablets, together with the low level of perceived efficacy of the treatment may have increased the likelihood of non-compliance. Failure to comply with the prescribed regimen for CAH might take some time to show its effects and might also be an additional factor to those already mentioned.

Management of diabetes, in contrast to CAH, is a task that requires constant vigilance. It involves the acquisition of new knowledge, new skills and strategies commonly aimed at the correction of both high and low blood glucose levels. These strategies comprise a collection of complex cognitive and behavioural skills, and include the understanding of the action of insulin, food composition and the effects of exercise. The necessity of eating at regular times, and the availability of carbohydrate supplies to avoid hypoglaecemic attacks were given as the most frequent regimen problems, particularly when the women were engaged in sports, social outings or were responsible for children. The women also recalled regular meals and emergency carbohydrates as important demanding requirements during childhood.
It might be anticipated that non-compliance with such a complex regimen might be common. Non-compliance with the proscribed regimen for diabetes is commonly held by the medical profession to be the cause of failure to maintain consistently acceptable levels of blood glucose. The women in the study were aware of the advisability of this goal although it appeared that it was rarely attained:

*It's too high [my HBA1C], about thirteen I think. It should be six or seven. It's been unbalanced for so long, and not enough exercise. I know what I should do but I can't be bothered.* (D26)

*I tend to stay on the high side for insurance [against hypos] (D02).*

*My control is not wonderful, very up and down cos I'm lazy and don't want it [diabetes] affecting things. High sugars are better than having hypos all the time.* (D26)

For this group of diabetics, it would seem that the avoidance of hypoglaecemic attacks was considered to be of more importance than the low blood glucose levels advised by the doctors at their diabetic clinics. This avoidance was frequently the major criterion for decisions about eating behaviours rather than the avoidance of long-term complications. It was these considerations which determined blood glucose levels rather than carelessness about, or disagreement with, the goals set by their doctors (Marteau et al 1986).

It is not surprising therefore that experiences of 'hypos' were currently rarely a problem for these women:

*not had one for eight years (D01)*

*last year my hypos were terrible, they're O.K. now (D18)*
sometimes minor ones in the night, I just have a biscuit
(D23)

The implication of the outcome of persistently high blood glucose levels for the
development of diabetic complications was also well understood by those in the
diabetic group:

As I've been so uncontrolled for so long, is it cutting my
life span down? (D28).

In summary, it appeared difficult for the women with diabetes to comply with
the goal of consistent blood glucose at the level recommended by their doctors.
The cost of this in terms of possible hypoglycaemic attacks was seen by the
women as too great, especially when other responsibilities or demands were
involved. In the short term, failure to comply with the demands of the regimen
was less noticeable for the CAH group; for them, compliance would therefore
be more likely to depend upon the understanding of their condition and the role
of medication in the control of the effects of hormonal imbalance. In particular
the self-report of the total omission of the medication by some women with
CAH is of concern.

The dimension of future changes of regimen

Neither group believed their regimen would materially change in the future. The
CAH group believed that there would be little medical progress that might lead
to regimen changes:

'I don't think anyone will be able to do anything' (C33)

Once again this may have been related to their general lack of information about
all aspects of the condition. The lower public profile of CAH than diabetes
would add to the considerable difficulties of the women with this condition in becoming aware of any ongoing and/or proposed research. In contrast, women with diabetes were well informed about research into their condition, and were more positive about future possibilities. They felt that medical research would produce advances in both the prevention and management of the condition, even if they were not optimistic that such advances would be achieved in time to be helpful for them:

*With modern technology it's going to get better (D14).*

They were confident that there would be improved long-term prognosis, and increased availability of alternative techniques for the management of diabetes:

*I heard about the nasal spray - good idea. I would use a pump, and transplants will be more available - I might have one (D30).*

They believed that advancement was most likely to be made in the area of genetic research, and that this would allow the identification of those at risk:

*Don't think there will be a cure - they may be able to prevent it, give relatives a test to see if [they are] going to develop it (D25).*

The concept of long term regimen then, contained two dimensions: the levels and difficulties of compliance with the required regimen, and the perceived possibility of future changes in medical management. Both groups of women appeared to have some difficulty meeting the goals set by their medical experts, but the failures of both groups could be attributed to factors other than carelessness or disagreement with goals set.
The concept of the 'Value of the consultation'

The final concept of the category of 'Demands of chronic conditions' (see Table 3), that of the perceived value of the hospital consultation, is related in a complex way to the concepts already discussed. In order to clarify these relationships a summary of the concepts and dimensions identified as comprising the category are given in Figure 3 below:

FIGURE 3: Summary of the concepts, dimensions and elements of the 'Demands of the chronic condition' category

It is proposed that a relationship existed between the concepts, dimensions and elements of the 'Demands of chronic condition' category and the concept of the value placed by the women on their visits to the hospital clinics. The proposed relationship is that negative emotional responses to clinic visits as children, a
perception of not being known by the doctor results in not knowing what the consultation is about. The failure to understand the doctors’ communications and a belief that the doctor was both unapproachable and did not have a high level of knowledge about the condition results in feeling uninformed. When these were experienced in the context of a hospital perceived as unfriendly because the medical procedures were conducted without consideration for privacy or a patient-centred organisation, they result in visits being devalued. The resulting low level of understanding about the condition, reinforced by the absence of sources of information and support, together with low confidence levels in the efficacy of the treatment regimen add to the low value placed on the visit. It is necessary to ground this proposal by examination of the data for evidence of these relationships.

Although paediatric clinic visits were perceived as having had a low value by both groups of women, different values were held for their current clinic visits. About three-quarters of those women who were diabetic believed that their visits were worthwhile,

_The care is brilliant - you can't fault it (D14)_

However this belief was reported by very few, less than one quarter, of those with CAH. The majority of women with CAH expressed their negative feelings about attending adult clinics in very strong terms:

_[A] complete waste of time (C07)_

_I've no faith in it at all (C33)_

_[They] haven't helped me any (C37)_
They treated me fair enough (C29).

The proposed relationships underlying this negative evaluation were supported by the data. Ninety one per cent of those who expressed negative evaluations of clinic visits also expressed negative positions on the other above dimensions. Holding a view that regular visits to hospital for consultation with medical experts had little value may have important consequences, especially in the case of a chronic condition with serious outcome effects. This issue will be considered in the following section of the category of ‘Strategies’.

THE CATEGORY OF ‘STRATEGIES’

It is reasonable to propose that the women responded to their constructions of experiences in ways that they believed were compatible with their perceptions, although these different response strategies may or may not be adaptive in terms of the management of their medical condition. For example, those placing a low value on the hospital consultations would be likely to engage in different behaviours to those placing a high value but a particular strategy may have negative effects on the management of the condition.

In the research literature, such strategies have been grouped under the heading of ‘Health Behaviours’ and include behaviours that are believed to be involved in both the prevention and the management of disease. Health Belief models (Rosenstock 1966; Becker and Maiman 1975; Leventhal and Cameron 1987) have conceptualised the individual most likely to have a good medical outcome as an active problem-solver; passivity in a patient has been seen as detrimental
to the good management of illness (Dunn 1987). Taylor (1979) suggested mechanisms whereby passive behaviour could be detrimental to outcome and proposed that individuals using this approach would be in a state of 'learned helplessness' (Seligman 1975). The women with CAH showed a clear tendency to be passive in relation to the management of their condition. Their failure to seek information about, or understanding of, their condition, and their belief that their doctors knew very little about them or their condition, together with the low value held about clinic consultations would seem to suggest a considerable degree of learned helplessness. The development of this role, Taylor believed, would increase the likelihood of the patient failing to report new or changing patterns of symptoms, and would increase the likelihood of remaining uninformed in matters related to the relevant medical condition. In summary, the low value placed on visits to outpatient clinics appears to be associated with behaviours and attitudes indicating a passive role and a lack of involvement.

The most commonly reported strategy engaged in by the CAH group in relation to their medical consultations, was to be 'not interested', and those who reacted in this way believed it to be as a result of the perceived poor communication skills of their medical advisers:

*I would think there was no point in my being there. In the end I would just get un-cooperative.* (C09)

*You think,"Well, I don't know why I bothered [coming]"!* C52

*I'm one of those people, if they talk *about* me, I tend to ignore it- I don't want to know.* C04

*I just block it out.* C15
Reported withdrawal during paediatric clinic visits appeared to have continued into the present for the CAH group, although not for the women with diabetes. It has been suggested above that other factors, such as parental understanding of the condition and the availability of other sources of information, may have compensated for the early lack of interest by the diabetic group. As adults, they became more active and more interested participants in the management of their condition. In contrast, those with CAH, in addition to showing a continuing lack of interest and control in the management of their condition, adopted additional negative strategies. For example they failed regularly to attend the endocrine outpatient clinics for routine check-ups. The following are examples of statement made by women with CAH who reported a 'low value' position for clinic visits:

*I'm supposed to go twice a year, but I cut it down to one.*
*C08*

*I wouldn't come if it was more than once a year* C09

*Since I had a baby I haven't been at all* C47

This combination by the CAH group, of the strategies of lack of interest and infrequent attendance, would not lead to improved understanding of the condition, or to an active participation in the management of their condition. The model proposed here suggests that the culmination of the meanings formulated about their experiences, based round understanding and communication difficulties, had important repercussions for those with CAH.
and led to a need to avoid communicating with others about their chronic condition and the development of a 'Skeleton-in-the-cupboard'.

THE CATEGORY OF THE 'SKELETON-IN-THE-CUPBOARD'

A skeleton-in-the-cupboard is defined by Collins English Dictionary as 'a scandalous fact or event in the past that is kept hidden'. This definition describes the central core category of the present study, the one to which other categories were related. Related categories may be conceptualised as either contributing to the 'Skeleton-in-the-cupboard', for example the 'Demands of chronic condition' and 'Strategies', or arising as a result of the category as in the case of 'Psychosocial effects' (see Figure 2). This central core category, the 'Skeleton-in-the-cupboard', was comprised of two concepts; 'Health images' and 'A shameful secret' (see Table 8).

<table>
<thead>
<tr>
<th>TABLE 8: Concepts, dimensions and elements of the category of the 'Skeleton-in-the-cupboard'</th>
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<tbody>
<tr>
<td>Concept 1 - Health images</td>
</tr>
<tr>
<td>Dimension 1 - health status</td>
</tr>
<tr>
<td>Dimension 2 - health related difficulties</td>
</tr>
<tr>
<td>Dimension 3 - physical/psychological problems</td>
</tr>
<tr>
<td>Dimension 4 - condition related attributions</td>
</tr>
<tr>
<td>Concept 2 - A shameful secret</td>
</tr>
<tr>
<td>Dimension 1 - parent-child communication</td>
</tr>
<tr>
<td>element of attitudes to condition</td>
</tr>
<tr>
<td>element of willingness to discuss condition</td>
</tr>
<tr>
<td>Dimension 2 - child-other communication</td>
</tr>
<tr>
<td>Dimension 3 - adult-other communication</td>
</tr>
</tbody>
</table>
The concept of 'Health images'

Both the attitudes towards and the perceived effects of a chronic condition would seem to be important issues for the way the self-image is developed; one aspect of the self-image will relate to the individual's health. For the women in this study, images about their health contributed to the development of the 'Skeleton-in-the-cupboard'.

The dimension of health status

The influential work of Parsons (1951) placed emphasis on two aspects of 'being ill', a distinction being made between the biological and the social basis of illness. Both of these aspects may be related to the individual's capacity to fulfil their work and social obligations, and in modern society the release from such obligations due to illness, requires medical sanction. The adoption of a sick-role has been shown to have negative consequences on independence, the maintenance of adequate self-esteem and on social interaction and activities (Freidson 1970). In the present study the women's perceived health status ranged from 'Physically I feel quite well' (D09) to 'Poor, I'm not feeling too good, very knackered-like' (C41). However the majority of women in both groups felt that they were in good health; the sick-role did not appear to have been adopted by either the diabetic or the CAH women.

Wright (1987) suggested that an individual's self-rating of their health status would be influenced by the frequency of health related problems as well as the functional limitations attributed by the individual to the illness. Both during
childhood and currently, there were possible health-related problems for both the diabetic and the CAH group.

*The dimension of health related difficulties*

Both groups denied that their chronic condition had imposed noticeable limits on their activities, either during school days or currently, some restrictions were mentioned although these were perceived as being of little consequence. The women with diabetes felt they might have had more flexibility in their career choices; for example being able to drive for the Post Office, or joining the Diplomatic Service. Their dietary needs had some effect on social activities, and in particular they found it difficult, but not impossible, to enjoy unplanned or extra meals. The CAH group felt strongly that the effects of the condition were on social areas of their lives and more detailed discussion of these will be reported below.

*The dimension of physical/psychological problems*

Women who defined themselves as being in good health and those who did not reported similar types of problems. These problems could be categorised as predominantly physical (for example, aches and pains), or psychological (for example, depression) in character. Comparing the groups on these categories however, there were some differences. The diabetic group reported similar frequencies of physical and psychological problems, whereas those with CAH reported predominantly problems of a psychological type. Eight (42 per cent) CAH group women, and eleven (65 per cent) of the diabetic group reported episodes of depression. These episodes appeared to be severe and three-quarters of the women with CAH and two-thirds of women with diabetes who
reported feeling depressed also reported suicidal thoughts and/or attempts. It is notable that only one woman in each group had received any professional help for their severe depression:

*I get very, very depressed and think about taking a bottle of pain killers, or have a crash in the van. I did once [crash], -I wasn't concentrating - did I want it? Been to the GP, but no help, nobody wants to help (C09).

Always been depressed and miserable and invented problems in my head. I've been getting awful feelings of despair - to go through the tablets. I have in the past, my parents took me to A.& E. I have put my foot down on the throttle and wanted to go into brick walls. I get irrational and it scares me (C15).

I've attempted to kill myself three times... I went to a counsellor after the second and third. I had big mood swings, they said I would grow out of it, I thought I was abnormal (D23).

The dimension of condition related attributions

The pattern of attributions made by the women to account for their problems was complex. Very few of those who were diabetic saw either their physical or psychological problems as being related to their condition. They believed that a loss of weight and more exercise would improve their general health, and were not able to identify causes for their depression except in general terms of 'stress'. The women with CAH also spoke of the causes of their depression in similarly general terms. However, they were more specific in that they saw a connection between some of the physical characteristics of their condition, for example their small stature, and their psychological problems. The women with CAH attributed to their condition a psychological characteristic that was perceived by them as negative. They remembered being shy at school, and as adults perceived themselves as 'introverted'. If it had not been for the CAH:
I don't think I would rely on my family so much; I would be more outgoing. I am reserved, contained within myself. I think there's nobody else I've ever met that's got the same as I've got. I rely on myself for everything. (C33)

I may be more confident in myself. (C47)

Bury (1988) distinguished between the effects of, and the connotations associated with, a chronic condition, and concluded that it was the latter that had the prime effect on the individual's sense of self. Sense of self, or self-image is constructed from meanings and interpretations of a variety of experiences. It is of note that both groups experienced regular visits to hospital clinics and the need for daily medication and these must have been in conflict with their belief that they were in good health. In this area of experience the difficulties of those with CAH in understanding the condition and therefore what was wrong with them, must have been of importance in the development of a somewhat confused self-image:

Ordinary time I'd think, 'it's your fault because if you'd found out what's wrong with me, I wouldn't need to keep coming.' I always feel O.K., do I need to keep coming? (C15)

I even had this fear in my mind that there was something wrong with me, I was unnatural – I was born with something no one else had got. (C07)

The actually stopped me growing because they said if I'd grown any more the dose of steroids wouldn't match with my body weight and my body height, so they stunted my growth. I would have liked to be taller. (C37)

I used to say to Mum, 'Why do I have to go?' and she'd say, 'You just do, that's all.' (C09)
In addition to the confusion relating to the good health/illness dichotomy, they also expressed confusion in relation to whether they were the same or different to other women:

> *I look at women and think 'Well, I'm not like them' you know. The doctor said, 'There's not anybody else quite like you', so I walked out of the door and climbed into my space ship.* (C03)

> *I became introverted because I knew I had this abnormality that no one else has got.* (C01)

It would seem therefore, that the women with CAH attributed to their chronic condition a number of negative outcomes relating to their self-perception. These attributions, although having some validity, again showed the effects of poor understanding about the condition and its treatment.

Bury (1988) considered the reactions and transmitted beliefs of important others in the life of the individual to be important influencing factors in the development of images of the self. The second concept of the development of the category of the ‘Skeleton-in-the-cupboard’ is the presence of a ‘Shameful secret’ (see Table 8). It is proposed that the development of this phenomenon was largely dependent upon the tendency of those in the lives of the women with CAH to fail to communicate to them a clear understanding about CAH. Where no clear understanding is acquired, there is a strong possibility that distorted, and frequently maladaptive meaning and beliefs will be formulated by the individual. This distortion may enhance the perceived need for secrecy.
The concept of a ‘Shameful secret’

There were very clear differences between the CAH and diabetic groups in their need for secrecy. The women with CAH were unanimous in their perception of a general requirement to keep ‘what was wrong with them’ a secret. The women with diabetes were very clearly willing to talk about their condition. Willingness to share information about oneself, particularly intimate information, has been considered an important contributor to the development of relationships (Duck et al, 1984; Miell and Duck, 1985), and in general terms, the closeness of the relationship correlates with greater willingness to share. It is therefore necessary to consider the appropriateness of secrecy in relation to different relationships. Table 8 identifies three dimensions contributing to the concept of a ‘Shameful secret’, the first of which is ‘parent-child communication’.

The dimension of parent-child communication

During the developmental years parents provide the most important, most powerful and closest relationships, and through their attitudes about, and reactions to, the presence in the family of someone with a chronic condition, may have considerable influence on the child's beliefs, self-image and development of family secrets. Two interacting elements connected with parent-child communication were identified in the data: the parents' willingness to discuss the condition with their children, and the parents' attitudes to the chronic condition.
The element of parents' willingness to discuss the condition

As discussed above, the parents of those with diabetes had adopted a teaching role in relation to their daughters' condition. It is not surprising therefore that this group of women mainly recalled their parents as open in communication about the condition, although a few did remember some difficulties in childhood:

*me not being able to say how I felt* (D01)

In contrast, the women with CAH strongly described their parents as wishing to avoid discussing the medical problems:

*They didn't talk about it* (C01)

*Mum wouldn't talk about it. I only spoke to Dad once, and he said 'Your mother doesn't like talking about it 'cos it upsets her'* (C09)

*I don't know how my mother coped, she didn't talk to me about it, I knew there was something wrong when I found out I'd had an operation.* (C41)

These statements clearly support memories of a need during childhood for secrecy about the condition of CAH.

The element of parents' attitudes to the condition

The women of both groups also identified two conflicting issues relating to the element of parents' attitudes towards their condition. The women described their perception of a strong emotional trauma for their parents at the time their condition was diagnosed, and it is not easy to reconcile this with their reports of their parents' insistence that their daughter was 'normal'.

*They were upset by it and found it difficult to take* (C01)
I think it was hard for them to accept it at first. She told me that they took me away and she thought it was 'cos I was abnormal - a vision of me with no legs, arms, ... or two heads (C37)

It was a very big strain on them. She was told I may not live to three - I'd be a midget - she always describes being told 'she's got diabetes, you're stuck with it for life' The doctor was very brutal (D04)

I feel sorry for Mum 'cos she broke down and became hysterical (D28)

In addition, the most frequently expressed belief by women with diabetes about how their parents responded to the condition was that their parents, particularly their mothers, felt guilty or in some way responsible for the development of the condition. In some cases this was because they had not recognised the symptoms of the diabetes quickly enough so as to avoid what these parents considered to be a serious health risk:

'she always felt guilty, hadn't noticed it sooner - I was very ill - they said I was close to death'(D26).

..[They] blame themselves for it, 'what did we do?'(D23)

In contrast to the parents of those with diabetes, the difficulties in understanding and communication by the parents of those with CAH appeared to have made it very difficult for the women with this condition to construct a clear picture of their parents’ attitudes and feelings.

However, both groups were similar in their recall of the efforts of their parents to consider their daughter as 'normal':

She was ....determined not to make me feel an alien (C37)

They treated it as normal (C07)
My parents never made a big thing of it, I was just the same as everybody else (C28)

They never treated me as any different (D14)

This impression of their parents' insistence on normality was supported by their recall that neither group of parents prevented their daughters from engaging in the usual activities of childhood because of their condition:

they never stopped me doing anything(C01)

The women in the diabetic group did remember their mothers expressing anxiety about the availability of food and timing of insulin injections when their daughters engaged in activities away from home. Overcoming these difficulties appeared to have been an important developmental task as they concluded that their diabetes had actually made them more motivated to participate in order to prevent themselves being seen as 'different'. This attitude is similar to the active role in seeking out information about diabetes already reported, and contrasts with the passive responses of those with CAH.

The two groups therefore, have a strong impression of their parents' insistence on 'normality' during their childhood. However, for both groups, this insistence operated against a background of stressful events: the regular visits to outpatient clinics, daily medication, and at some time-point an awareness of the trauma of the diagnosis of their condition for their parents. It is possible that the successful resolution of these conflicting positions- the 'illness experiences' which, as will be described more fully in a later section, included the belief that they were abnormal in some way, and the 'normality of childhood' - depended
upon the degree of openness in communication and the understanding about the condition which this openness encouraged. Parents' openness in discussing their child's diabetes may have encouraged the belief that diabetes was acceptable, whereas for the CAH women the insistence on normality together with the absence of open discussion endorsed the need for secrecy and kept the 'Skeleton-in-the-cupboard' in place.

*When I went back to school they all said to me, “Where have you been?” Mum always said, “Don’t tell them anything”. I couldn’t tell them anything. I didn’t know anything!* (C09)

It must also be stressed that the sexual focus of the CAH was an essential factor in the issue of secrecy versus openness. The combination of this sensitive focus and the difficulty of adequate explanation resulted in the need for the CAH 'Skeleton-in-the-cupboard', whereas the increasing public awareness about diabetes both endorsed its social acceptability and affirmed it as a topic for discussion.

**The dimension of child-other communication**

*That's what happens - it [non-communication] gets passed down, it's a vicious circle (C01)*

For the CAH group, the difficulties in communicating about their condition were not confined to interaction between themselves, the doctors, and their parents, but were apparent across a wide range of relationships- CAH was a well-kept secret:

*You didn’t talk to anyone [about] what was wrong with you.* (C41)
I might need to explain it. None of my friends knew about it, it was a secret. (C01)

They [friends] knew I got ill now and then, but not what it was. I've never told anyone about the ops. I was ashamed of it. It was hidden. (C33)

The difficulties in openness were of particular importance in the development of heterosexual relationships. Although such difficulties will be discussed in the section on adult-other communication, it is of note that the issue of both open communication and normality/abnormality affected early interaction with boy friends:

I hid it from developing relationships cos I knew I'd got that abnormality and I've got to trust that person. You can't be open. C01

**The dimension of adult-other communication**

For both groups, little appeared to have changed over the years; the differences reported by the two groups in open communication during childhood continued into adult life, with the women with CAH retaining their problems in discussing their chronic condition. For some of the women with diabetes, telling their current friends was an event about which they felt essentially positive:

All of them. Sometimes I'm quite proud I'm different, that they think I am brave (D02).

All of them, it's part of my life I've shared with them (D34).

The majority of those with diabetes, however, took a more reticent approach and felt that although all their friends knew about their condition and found it interesting, it was necessary for the friendship to reach a position of trust before this information was shared:
All of them It's a question of it just coming up over a period of time as you get to know people, rather than a definite plan to tell. (D09)

They all knew, but I don't go out of my way to tell people. (D14)

The majority - I'm only open with people I'm going to see on a regular basis - it's a question of trust. (D33)

In contrast to this, only one woman with CAH, felt able to explain to her friends about her condition and the similarity between her attitude and those of the more reticent women with diabetes is noticeable:

Yes, when they reached the right point in the relationship, I explained it to them and asked them what they thought. They looked at me and said, "well, you look alright to me!" (C13).

The remainder of the CAH group spoke about their continuing difficulties:

I hide it. (C07)

You shield yourself. You don't want people to know. (C37)

Not a lot of people know what's wrong with me. If I do, I just mention the tablets and the enzymes - I won't ever, ever, mention the physical side. (C41)

For those with CAH these difficulties had effects at all levels of social relationships and were very strongly evident in relationships that might have been, or were, sexual. The women put forward the belief that when sexual relationships became long term and secure, they would find the condition easier to explain:

When I settle down in a steady relationship, I could talk to that person about it, but at the moment I'm not settled. C08
This confidence in ease of future openness received only partial support from those women in the CAH group who had a steady relationship; some had achieved openness and some had not:

Yes, he used to take me to hospital. He was there when I had the baby, he knows all about it. (C10)

He knows as much as I do, he's been told everything, how it came about, he knows about the operations. (C28)

I never told him, he did ask but I said "I've coped with it for the last 30 years what's it to you to worry about?". (C41)

In line with the other aspects of explaining the condition to others, the partners of those with diabetes were reported as being well informed and, importantly, seemed to be supportive and willing to share the responsibility for the management of the condition. The conditional willingness of the diabetic women to discuss their condition and of their partners to share responsibilities, were clearly expressed in the following statements:

Yes, we told everyone. I didn't go around telling everyone, it became apparent it just came up in conversation. Most of them were interested and wanted to know more. D14

When I first met D, I didn't know what to tell him. I thought I'm not going to bother him, he might drop me like a hot brick. He's brilliant, just bit by bit. He doesn't nag me but it works. D04

With C, he had to learn how to do it (injections), do it infront of my mum, if I'm ill with flu and I haven't got the energy, he does it for me. I can't pick fault. D08

I've never made a point about it [telling my partner, or] it being someone else's responsibility, but to me it's a great bonus, it takes the pressure off a bit. D09

In addition to the possible embarrassment caused by the need to communicate about their condition, explanations would be made more difficult for the CAH
women because of their own lack of understanding—they did not know what to tell. Although some of those with CAH who had steady partners reported they were able to explain to them about their condition, the clarity of their explanations must be questioned. The woman (C28) quoted above, who claimed she had explained 'everything' to her husband, believed that her condition was the result of her mother taking medication to prevent a miscarriage. Both she and her husband learned a more accurate explanation, including the genetic aspects, only when they attended a fertility clinic. The new information created considerable marital difficulties for this couple. For the diabetic women there were some advantages in telling others; many of them spoke about increased feelings of security when others were aware of their condition and knew what to do if they became hypoglycaemic.

In summary, the interview data suggested that for those with CAH a number of experiences culminated in the development of a 'Skeleton-in-the-cupboard' (see Figure 1). Further analysis identified this category as central to the understanding of how these women construed their experiences. Clear support for these proposals was provided in the interview statements, and it is helpful at this point to clarify the ways in which the phenomena came into existence.

There certainly was a 'Skeleton-in-the-cupboard' for the women with CAH; their condition was consistently hidden both as children and as adults. The contributing experiences that led to the perceived need to keep the secret involved many areas of life, with the underlying issue of communication appearing to be of prime importance. Two related characteristics of CAH are
pertinent here: the complexity of the condition together with its socially sensitive nature. The combination of these two created a situation where the parents did not understand the information available to them during medical consultations and were too confused, anxious and embarrassed to ask for clarification. This difficulty extended to communications between parents and their children, and between children and their friends. It persisted into adulthood, to communications between same and opposite sexed friends and to sexual partners. The lack of understanding that arose from the communication difficulties with the doctors could not be rectified by information from other sources, as these were not available. These difficulties also resulted in a devaluation of the hospital outpatient consultations, and a failure to develop the establishment of an active participation in the management of the condition, management shared between the women and their doctors.

The demands of the chronic condition were in opposition to the attitudes of the women's parents. There was a general parental insistence that their daughters were 'normal', and this whilst possibly being adaptive, could be seen as being in opposition to the need for regular hospital visits, surgery and daily medication. These conflicting factors, together with a growing awareness of their parents' reactions to the diagnosis of CAH, underlined the communication difficulties. The women with CAH perceived these issues as contributory factors to some undesired aspects of their self-image, aspects such as childhood shyness and 'introversion' in adult life.
It is experiences such as these and the constructions placed on them that have created the conditions for ensuring that the facts, the skeleton, have been hidden, both from others and, by the failure to gain an understanding of the CAH, from the women themselves. Although some of the early paediatric hospital experiences were similar for both groups of women, two differences emerged: the openness of communication between parents and children and between the children and their friends, and the greater understanding of their condition through information sources other than directly from the clinic doctors. The need for secrecy was inconsequent for those with diabetes because of these factors and the absence of any sexual aspects of the condition. The implications of the need to maintain a 'Skeleton-in-the-cupboard' were pervasive, and it is these outcome issues which will be examined in the following sections.

THE CATEGORY OF PSYCHO-SOCIAL EFFECTS OF THE 'SKELETON-IN-THE-CUPBOARD'

The presence of a 'Skeleton-in-the-cupboard' from an individual's early age may be expected to have far-reaching effects. As Table 9 identifies, the data supported the existence of effects from school to adult life.
TABLE 9: The category of 'Psycho-social effects' of the 'Skeleton-in-the-cupboard': concepts, dimensions and elements

<table>
<thead>
<tr>
<th>Concept 1 - Effects on Education</th>
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<td>Dimension 1 - academic attainment</td>
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<td>Dimension 2 - school attendance</td>
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<td>Dimension 3 - academic problems</td>
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<td>Dimension 5 - attitudes towards learning</td>
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<th>Concept 2 - Effects on social relationships</th>
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<td>Dimension 1 - school-days experiences</td>
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<tr>
<td>element of friendship formation</td>
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<td>element of friendship structure</td>
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<td>Dimension 2 - current experiences</td>
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<td>element of satisfaction with friendships</td>
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<tr>
<td>element of concepts of friendship</td>
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<td>element of fulfilment of needs</td>
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<th>Concept 3 - Effects on sexual/partnership experiences</th>
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<tr>
<td>Dimension 1 - sexual interest/partners</td>
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<td>Dimension 2 - marriage</td>
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<td>Dimension 3 - parenthood</td>
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The concept of the effects of the 'Skeleton-in-the-cupboard' on education

As shown in Table 9, three concepts were identified from the interview data of the psychosocial effects of the 'Skeleton-in-the-cupboard'. The first was concerned with effects on education and was comprised of five dimensions: academic attainment, school attendance, academic problems, family problems and attitudes towards learning.

The dimension of academic attainment

The majority (59 per cent) of the diabetic group was successful in passing at least one national examination at A level. This was only achieved by 16 per cent (N=3) of those with CAH, with 63 per cent (N=12) of this group leaving school without any O or A level examination passes. The two groups were similar in
the types of schools attended, with a high proportion of both groups attending non-selective secondary schools (see Table 10).

TABLE 10: Types of schools attended by both groups

<table>
<thead>
<tr>
<th>GROUP</th>
<th>SELECTED SCHOOLS</th>
<th>NON-SELECTED SCHOOLS</th>
<th>TOTALS</th>
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<tr>
<td></td>
<td>N (%)</td>
<td>N (%)</td>
<td>N (%)</td>
</tr>
<tr>
<td>CAH</td>
<td>3 (16)</td>
<td>16 (84)</td>
<td>19 (100)</td>
</tr>
<tr>
<td>DIABETIC</td>
<td>6 (35)</td>
<td>11 (65)</td>
<td>17 (100)</td>
</tr>
<tr>
<td>TOTALS</td>
<td>9 (25)</td>
<td>27 (75)</td>
<td>36 (100)</td>
</tr>
</tbody>
</table>

Chi square = 0.9288, d.f. = 1  N.S.

The type of school attended was not therefore a significant factor in the considerable differences between the groups in the levels of final educational attainment. The differences are unlikely to be directly related to the regimen demands of the chronic conditions. The demands for those with diabetes would seem to be greater during school days, and as already reported they recalled that the demand of the need for regular meals at school was felt to be considerable. The interview data suggested a number of other factors that may have contributed to school achievement.

The dimension of school attendance

The presence of a chronic condition may necessitate absences from school due to ill health or attendance at hospital for inpatient and/or outpatient treatment. About a third of the women in each group recalled frequent absences during their school years, with the reason commonly given as their chronic condition:

*I wish I could start school again, I've missed out on quite a lot of things. I was in and out of hospital, so I didn't go often, and not being well.* (C10)
I refused school. I kept getting colds, I was always ill. (C15)

I went to hospital a lot due to the fact that I was going through adolescence - I wasn’t too well. I worked it out, it’s about seven years of my life I’ve spent in hospital. (C41)

I missed so much schooling. In the sixth, I had less than one third attendance. I used to say (it was) cos of the diabetes - it was awful. (D24)

By [the] end of the fifth form, it was the equivalent of one year off school with sickness. At the time I never realised it. (D26)

For the diabetic group, absences from school did not appear to be related to achievement; those reporting frequent absences did not also report educational difficulties and perceived themselves as achieving well in their schoolwork. It would seem that the diabetic group achieved their higher examination successes in spite of their frequent absences. In contrast, for those with CAH their frequent absences were associated with both self-perceptions of poor academic achievement and considerable academic problems.

**The dimension of academic problems**

A further difference in the reports of school progress was the recall of experiences of academic learning problems. Although two (12 per cent) of the women with diabetes remembered ‘examination nerves’, no member of this group recalled any specific academic problems. All seven of those women in the CAH group who were frequently absent recalled learning difficulties with five of them having had difficulties in both reading and numerical skills, the remaining two in numerical skills:

*I don't know any maths. Reading is a problem, I get bored 'cos I can't read it quickly enough. People were*
getting on and I wasn't. I thought, what's the point? (C04)

I was in and out of hospital, so I didn't go [to school] very often, and not being well. I found it very hard to cope at school, it was all too much. (C10)

I was ill so much when I was young. It was alright 'til I was about fourteen, then I didn't like it at all, I never used to go. I wasn't very bright, I didn't get on too well. I had a special tutor, I can add up and take away, but anything like 'yards' and things I can't do. I regret not getting on at school very much. (C33).

I missed a lot of school. [I got on] very well actually. I didn't take any exams, but that was my own choice. I would say I was middle to top [in class position]. (D28)

The dimension of family problems

The women raised a number of issues connected with their families that they felt had contributed to school difficulties. These were variable both in the type of problem and the characteristics of those recalling them. The problems ranged from constant house moves, deaths of family members, parental marital problems, and either a lack of or too much parental pressure to achieve at school. In both groups, such attributions were made by those who found it difficult at school, had specific learning problems, and did not obtain any nationally recognised examinations (low achievers) and also by those who obtained either ordinary or advanced level certificates of education (high achievers). Therefore little support was provided for a perception of family problems as major contributors to academic school achievement.

The dimension of attitudes towards learning

The final dimension to be considered was the recall by the women of general attitudes towards academic learning. Rutter (1983) proposed that the
experience of attending school should promote more than academic achievement; school attendance should also develop positive attitudes towards learning and social skills. Both of these areas were discussed during the interviews and the data suggested differences in attitudes between the women with CAH and those with diabetes.

Women with CAH recalled holding negative attitudes towards learning, attitudes of a low level of commitment and co-operation:

\[ \text{I was very argumentative- I didn't want to learn. (C04. low achiever)} \]

\[ \text{I didn't do a lot of work,......I had a 'laid back' attitude to school. (C08.high achiever)} \]

\[ \text{I wasn't happy there, I was used to getting by on my brains alone- I didn't work. (C07.high achiever)} \]

\[ \text{I couldn't be bothered. (C09.low achiever)} \]

In comparison, although women with diabetes expressed regret that they had not made more of their opportunities, they recalled a commitment to learning:

\[ \text{I wouldn't say the school was highly academic, but it was nice- I wish I'd concentrated more, but I did my best. (D09.high achiever)} \]

\[ \text{I didn't enjoy my time there at all. I regret I wasn't wise enough to make the most of the opportunities, I kept on with it. (D24.high achiever)} \]

\[ \text{I had a few problems with maths, but nothing I couldn't sort out, I found it difficult to understand, but I got my GCSE. (D18.low achiever)} \]

\[ \text{I was always near the bottom of the class- I'm not a brilliant person, but I think I'm quick at picking things up. I didn't let school work get me down. (D29.low achiever)} \]
In general, the women with diabetes, both good and poor attenders at all levels of academic achievement, perceived themselves as having coped adequately. The CAH group women did not recall school as easy and appeared to have lacked the determination to overcome any academic difficulties. Explanations for such differences in educational achievement might be through two classes of factors. One would be either the direct effects of the medical condition or the proposed effects of hormone levels both on general intelligence and on cognitive profiles, and the second the psychological effects of an early diagnosed chronic condition.

The first set of attributions needs to be examined on two issues: on the question of the reliability of the reported findings and on the direct attribution of the results of hormone levels.

The evidence from post-mortem studies has implicated hypoglaecemic attacks as a major cause of cerebral lesions even when the attacks themselves are not fatal (Brierley, 1981). It is therefore possible that chronic diabetes could be associated with impairment on a variety of cognitive tasks although the results of studies attempting to directly test this association by comparison of those with diabetes and those without have been ambiguous (Franceschi et al, 1984; Holmes, 1986; Ryan et al, 1984). Prescott et al (1990) compared poorly controlled to well controlled adult diabetics on a memory test, but failed to find any effect of the degree of glycaemic control upon cognitive performance. A highly significant correlation between memory performance and duration of illness was found, although the magnitude was very small and was considered
unlikely to give rise to any material handicap in daily life. The authors agreed with Ryan (1988) who suggested that the demands of diabetes were likely to result in a response style of cautiousness, and that this was the best overall explanation for their findings.

The results of the studies examining the effects of CAH on general intelligence (Sinforiani et al, 1994; Resnick et al, 1986; McGuire and Omenn, 1975) are also ambiguous. The review of cognitive characteristics of CAH patients by Nass and Baker (1991) gave support for four findings: that CAH patients have a cognitive advantage; that this advantage is reversed to a disadvantage in CAH salt losers; that there is a possible increased risk of learning difficulties especially in connection with calculation abilities; and finally that post-pubertal CAH women may show an advantage in spatial ability. The conclusion that those with CAH may have a cognitive advantage has not been consistently replicated (Helleday et al, 1994), and Nass and Baker (1991) reported increased frequencies of learning difficulties for those with CAH. The results of studies comparing salt losers to simple virilisers show lower scores for salt losers (Helleday et al, 1994; Ehrhardt and Baker, 1974; Money and Lewis, 1966; Wentzel et al, 1978). It was not possible in the present study to compare salt losers with simple virilisers due to the small sample sizes. Given the high proportion of salt losers in the sample and the self-report of both general and specific educational difficulties, together with the lower final achievement levels compared to those of the women with diabetes, it might be considered that support for a cognitive disadvantage for those with severe levels of CAH has been supported. However, the educational outcome for this sample of women
with CAH may be a function of psychological factors. The difficulties reported by them in establishing and maintaining social relationships at school may have been an important factor.

The second issue to be considered concerns explanations for any cognitive profile differences. Cognitive characteristic have been attributed to the effects of hormone levels especially pre-natal levels, on the developing brain (Hampson and Kimura, 1992), but such attributions based upon biological essentialism have not received universal support (Vines, 1993; Kelly, 1991), and alternative factors may account for finding of any cognitive differences. It is proposed here that experiences contributing to the development of a 'Skeleton-in-the-cupboard' and its maintenance may be indirectly responsible for the patterns of low school achievement.

The relationship between frequent absences, family problems, attitudes towards learning and educational achievement were complex. Frequent absences and family problems were experienced by high and low school achievers of both groups; however attitudes towards learning suggested group differences with the diabetic group recalling a coping and the CAH group a 'can't be bothered' approach. It would be expected that those who held this latter attitude would not achieve highly at school, however both CAH achievers and non-achievers recalled a non-committal approach. It is proposed that a complex interaction of these factors may have been important in the levels of achievement attained in school. Frequent school absences resulted in poor progress with academic subjects and this, combined with the need to keep hidden the experiences of
CAH and therefore the reasons for their absences, may have had effects on school social relationships. Failure to establish school friendships has been found to result in low self-esteem, and low self-esteem has, in turn, been shown to be associated with poor achievement levels (Rutter, 1983; Asher and Coie, 1990). These proposed relationships will be further considered after the effect of the 'Skeleton-in-the-cupboard' on social relationships, has been examined.

The concept of the effects of the 'Skeleton-in-the-cupboard' on social relationships

Effects of the 'Skeleton-in-the-cupboard' on social relationships were reported for schooldays and adulthood, with the former of these being comprised of two elements – friendship formation and structure (see Table 9).

Research on social relationships through all age-ranges, suggests that the benefit of friendships must be offset against its obligations (Wiseman 1986). One implication of this balance for women with CAH is that the demands of disclosure and openness in friendship formation must be balanced against their perceived need for privacy and right to retain secrets (Duck, 1995). Likewise, friendship also involves a balance between interdependence and independence in the relationship. Such tensions and balances are not conclusively resolved for each relationship on a single occasion but require constant re-negotiation and adjustment over time as relationships develop.

In the development of childhood friendships, parents may be of influence in a variety of ways: for example by directly choosing playmates for their offspring,
and by direct teaching about relationship rules. For the women with CAH it is proposed that the development of the 'Skeleton-in-the-cupboard' during childhood would create considerable difficulties in the balance between relationship demands. The interview data was examined to identify evidence for such effects on school friendships.

*The dimension of school-day experiences*

*The element of friendship formation*

As Rutter (1983) concluded, one function of attendance at school is the acquisition of interaction skills and the acceptance into a 'peer group'. Lewis and Rosenblum (1979) defined a 'peer' as a social equal, someone who is operating at a similar level of both cognitive and behavioural complexity. This similarity plays an important role in the distribution of power in relationships, and the perceived equal balance of power in peer relationships influences opportunities to learn social mores, to try out new behaviours and express attitudes. Such experiences, and the ensuing feedback from others, are believed to influence the development of both an individual's self-concept and the learning of the rules of social relationships.

Peer contacts develop at, and are important from, an early age (Ellis *et al* 1981). Longitudinal studies (Roff 1961; Parker and Asher, 1987) indicate that early poor relationships predict later emotional disturbance during adolescence and adulthood. Certain characteristics have been identified that increase the chances of a child establishing good relationships and acceptance by the peer group.
Those children with well developed role-taking skills (Pellegrini, 1985), and good academic self-concepts are likely to be popular and have high status in the group. The interview data indicated that in addition to the low academic attainment and the incidence of learning problems, the women with CAH generally recalled poor academic self-concepts:

- *I wasn't very good.* (C03)
- *People were getting on and I wasn't.* (C04)
- *I found it very hard to cope at school, it was all too much.* (C10)
- *Not very well, I wasn't very bright. It was one of those schools where you got on, and I didn't.* (C33)

The self-concepts of the women with diabetes were more positive, and appeared to be based upon attributes other than academic achievement, in particular, on their own estimates of their efforts:

- *I did get on well through the course, I got to go down a bit in the exams.* (D21)
- *Very well, even though I was in the bottom group at the end of Junior School.* (D25)
- *Very well actually. I would say middle in class. I think I did quite well.....I was one who had to work fairly steady to pass exams.* (D34)

A number of behavioural characteristics have also been identified as important in peer acceptance. Children who are calm, who are co-operative and supportive of others and ask for help and information, are likely to be accepted, whereas those who are perceived as withdrawn are not. The recall of the women with CAH about their schooldays suggested difficulties that would put them at risk for low peer acceptance during this period of their development. They recalled
difficulties in asking other children for help and general reticence in communicating openly. The issue of open communication in relation to the chronic condition has already been discussed; whereas diabetic women were eager to discuss their condition with their friends, those with CAH were not. It is possible that this perceived need to keep the 'Skeleton-in-the-cupboard' resulting in failure to be open in communication with their peers, together with a poor academic self-concept, may have put the CAH group at risk for social relationship difficulties during childhood.

Although both groups recalled some difficulties in friendships during their schooldays there were some differences in the attributions made by the two groups. The majority of the diabetic group mainly saw the cause of their difficulties in their own behaviours:

- I was pretty quiet at school, hid at the back, I didn't make an effort. (D02)
- I refused to be put up a year when I was eight cos I had difficulties in making friends. (D24)
- When I got to senior school, I was a bit overwhelmed by the other children. (D25)

The CAH women, while identifying their own social goals as contributing to their difficulties, also described an important component of their difficulties as their need to protect themselves from intimacy with their peers:

- I tried not to have any friends, tried not to bring them home because they'd always be talking about school and I didn't like that. (C37)
- As far as I was concerned it [social life] didn't exist- I went to school and I came home. I never really had friends that I would confide in, I was sort of closed in - nobody came inside... [I was] terrified of being hurt, very,
very sensitive. So I didn't have any friends. Maybe I felt I was different, maybe I didn't want to tell people. If you have friends you begin to open up more as time goes on. It was a bit of a worry of what I would actually say to somebody. (C52)

For those women with CAH social experience at school was one of considerable isolation. The proposed effects of the 'Skeleton-in-the-cupboard' on social relationships at school appeared to be supported by the interview data.

The element of friendship structure

The structure of social relationships during school years may take either a group or a 'best-friend' format. Within the literature on children's peer relations, there has been an increasing recognition that friendship and overall peer group acceptance may constitute distinct independent aspects of the child's social world (Budzynski and Hoza, 1989; Parker, 1986). Budzynski and Hoza (1989) argued that having a close friend, a close mutual relationship with a peer, and being popular, being liked by members of the peer group, have independent effects on the child's feelings of self-worth. It might be expected that the perceived necessity for intimacy by school children with CAH, would create particular difficulties in close friendship relations and therefore pre-dispose them towards a group rather than that of a 'best-friend' structure. The level of intimacy has been found to increase as this type of friendship becomes further established (Berndt and Hoyle, 1985).

For those with diabetes, the majority (70 per cent) recalled the development of a 'close special friend' rather than membership of a group.
I had a friend that I always went around with, I met her first thing in the morning. (D25).

This type of friendship structure is not surprising for those with diabetes. It has already been reported that as girls they were comfortable in talking about their medical problems, felt others were interested, and were aware of a possible advantage of having someone who 'knew what to do' in the event of a period of low blood glucose. For the CAH girls, this open communication was difficult and two thirds of the group recalled feeling a 'loner'. The remainder described a loose association with others rather than a well-established group membership:

I didn't really enjoy it [social life at school], sort of being with girls I wasn't used to and didn't really like. (C07)

I had friends, but I didn't really mix to that extent, I lacked confidence..........I didn't really make the effort really. (C47).

I'm not a groupie, too suspicious I think. (C01)

A commonly reported pattern was to engage in social activities with school friends during the weekends and evenings; 57 per cent of the diabetic and 41 per cent of the CAH group recalled this pattern. A similar proportion of the CAH group recalled having friends at school but no one they could see out of school. Only four women with diabetes (24 per cent) failed to have out of school friends, and these were women who lived in a geographically isolated area. It would seem then, that the women in the diabetic group had both friends they could see at school and the same or others with whom they could socialise out of school. Less than half of those in the CAH group had the opportunity to engage in out of school social activities with those of their own age group. Once again evidence of a socially isolated life-style.
It was suggested above that friendship difficulties during schooldays might be associated with a low self-concept of academic ability. There was support for this association in the interview data; in both groups the majority of those who recalled difficulties with childhood friendships also held a poor educational self-concept: those who held a good self-concept were also successful in establishing firm friendships, especially 'best-friend' relationships. Poor self-concepts about academic abilities would also be likely to predict low academic success, and the poor levels of national examinations obtained by those holding such a self-concept was also supported by the data.

In summary, the women with CAH appeared to have experienced considerable difficulties in friendship formation, especially in establishing 'best-friends' as children. It is this type of friendship, especially during the teenage years, that provides rule-learning opportunities for intimate information exchange. The suggestion that children with CAH preferred a 'loose association' with a group of peers, rather than a close relationship was supported by the data. It would seem likely that this preference was due to reticence in disclosing personal information. Low peer acceptance especially when associated with withdrawal from relationships, appeared to be related to low academic attainment. Effects of the 'Skeleton-in-the-cupboard' on both the social relationships of childhood and educational achievement are supported by the interview data.

One outcome from the avoidance of close relationships is the failure to acquire the rules of intimacy (Sprecher and Duck, 1994). Difficulty in establishing early
close attachments, with the inevitable outcome of either 'aloneness' or 'loose associations', may be difficult to rectify making adult friendships also problematic. The data provided the opportunity to consider a number of elements associated with aspects of the women's current friendship experiences: their satisfaction with friendship networks, concepts of friendship, and the fulfilment of needs in friendship relationships (see Table 9).

**The dimension of current relationship experiences**

*The element of satisfaction with friendship networks*

Peplau and Perlman (1982) described loneliness as a discrepancy between an individual's desired and achieved levels of social relations. It is of course possible that some individuals desire a small network of social relationships and prefer them to be at a non-intimate level. Alternatively, there may be a considerable discrepancy between desired and achieved levels of these factors and in this case dissatisfaction with friendships and isolation would occur. The women in the study were asked to rate their level of satisfaction with their friendships on a scale from one to five, where five represented high satisfaction. These ratings were divided into two classes: rating greater than three were classified as 'satisfied', all other ratings as 'dissatisfied'. Table 11 shows the distribution of these two classes for both groups:
TABLE 11: Rating classes for satisfaction with current friendships

<table>
<thead>
<tr>
<th>GROUP</th>
<th>SATISFIED N (%)</th>
<th>DISSATISFIED N (%)</th>
<th>TOTALS N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>CAH</td>
<td>7 (37)</td>
<td>12 (63)</td>
<td>19 (100)</td>
</tr>
<tr>
<td>DIABETIC</td>
<td>14 (82)</td>
<td>3 (18)</td>
<td>17 (100)</td>
</tr>
<tr>
<td>TOTALS</td>
<td>21 (58)</td>
<td>15 (42)</td>
<td>36 (100)</td>
</tr>
</tbody>
</table>

Chi Squared = 5.888 d.f=1 p< 0.02

The distribution of frequencies in Table 11 suggests that significantly more of those with CAH than those with diabetes were dissatisfied with their current friendships.

The suggestion that early friendship difficulties continued into adulthood also gained support from the data; women in both groups who had recalled childhood difficulties also reported current difficulties. For both groups the proportion of women who perceived current difficulties was twice that reported for childhood. Reis et al (1983) reported increased intimacy in social interactions as women grew older, with this increase becoming particularly salient for women aged between eighteen and twenty-two years. The reported increase in friendship difficulties by both groups of women in the study suggests that both chronic conditions may be a risk factor predisposing the development of unsatisfactory friendships, although this was especially so for those with CAH.

In describing their current friendship experiences, the CAH group characterised themselves as isolated:

*I haven't really got anybody [as a friend]. (C34)*
I'm a bit of a loner. (C47)

I haven't got any friends, I'm a loner, I'm wary. I'm a bit shy to begin with. (C09)

I've never really been a social person. (C15)

I can live without that [friendships] I don't have to be dependent on people - it doesn't worry me. (C41)

Those with diabetes, while expressing some current difficulties in making and continuing friendships, attributed these not to their own traits or behaviours, but to lack of time, other competing demands, or to separations from friends because of vocational or family moves:

I don't see a lot of them now, which is a shame, with me being so busy doing my work and everything as it is at the moment, I've got less time. (D01)

Sometimes I wish I had a really good friend, 'cos most of my friends are not reliable. There isn't anyone that is really close to me, that I've made them my own. But I've been with X since I was sixteen so I've had him like, I'd sooner spend time with him. (D18)

It might be expected that dissatisfaction with achieved levels of current friendships would result in a desire to make new and closer friends. However, only two of the twelve dissatisfied women with CAH were interested or willing to consider this possibility; the three dissatisfied women with diabetes all felt that an increase in their friendship networks would be a positive event. It is possible that low satisfaction and unwillingness to make new friends may be a function of the expectations about, and conceptualisation of, friendship.
The element of the concepts of friendship

Friendship is a multi-factorial concept (for example LaGaipa, 1977; Parker and Asher, 1993). The following factors reported by Parker and Asher, (1993), are representative of the similar factors that have been reported across research studies:

- intimate exchanges; the discussion and sharing of problems, disappointments and uncertainties,
- conflict resolution; the ability to resolve conflict quickly,
- provision of companionship and recreation,
- provision of help and advice,
- validation and caring; the acceptance of the individual and the understanding of their feelings,
- loyalty.

The question in the study that focussed on this topic was 'What is a friend?'

Analysis of the women's replies suggested both groups saw friendship as being based on dependability, acceptance of the other and the provision of a source of companionship, which was described as the sharing of activities:

someone who was always there if I wanted her, and go out together, do things together. C10

Both groups were divided about the importance given to similarity of interests.

Some required similar interests:

People that would like to have a chat about nothing with you, similar interests and attitudes. You don't just have friends who think the same way as you, but people who have compatible interests. (D34)

...also, I'm interested in what they're interested, because there is something you've got to have, that's mutual interests. (C03)

Others preferred friends to have interests which were dissimilar:
[someone] that has different interests than your own- is just something a bit different to you. It's just something that clicks. (D01)

My best friend, our lives are poles apart- our views and things are completely different, but we still get on! (C07)

'Dependability' was centred on loyalty for the diabetic group:

somebody who is very loyal and supportive, and can be with you throughout any crisis in life (D09)

For those with CAH a strongly expressed component of confidentiality was added:

But I know she's my friend, I know she won't go telling everyone things. (C01)

You never know when it [information about self] can be turned against you. (C09)

Confidentiality was not expressed as a requirement by those with diabetes. In addition, those with diabetes did not highlight acceptance by a friend as of importance. The CAH group expressed 'acceptance' as a complex component comprising being respected as an individual, and the feeling that they could 'open up' and share with the friend both positive and negative feelings and experiences:

to take me as they find me- there aren't many that'll take me as they find me- that's the problem. (C03)

someone I feel comfortable with, someone who isn't judging me. (C07)

Loyalty, loyalty- that's all.....I have to know somebody for a long time before I would let my guard down, before I could trust them. C13
In summary, both those with diabetes and those with CAH spoke about friendship in terms of commonly held factors, and conceptualised 'a friend' in similar but not identical ways. For both groups, a friend was a companion, someone to do things with, although common interests may not be an essential component. Dependability in terms of loyalty was essential. Important additional concepts were identified in the data for those with CAH: confidentiality about personal information, and the acceptance of both their good and bad traits and their successes and failures. The women in the CAH group were less satisfied with their current friendships than the diabetic group, but did not express a wish to develop new friends. As open communication was recognised by those with CAH as an essential component of friendship, it was not surprising that the women with CAH were reticent in wishing to develop new, close friends. Three quarters of the CAH group described themselves as 'loners', and three (16 per cent) believed that they had greater success with male friends than female.

Self-disclosure, the sharing of intimate aspects of the self was held by the women with CAH to be an important component of friendship, and research in this area has concluded that a successful friendship is dependent on this skill. Self-disclosure not only becomes progressively more intimate and more essential as friendships develop, it is also seen as a requirement that the level of intimacy is reciprocal, requiring a 'match' in level of disclosure by both actors in the relationship. This match applies to both the intimacies of topic -what it is permissible to talk about- and to the depth at which the information is exchanged (Duck et al 1991). The presence of a 'Skeleton-in-the-cupboard'
would tend to inhibit self-disclosure and thus contribute to an avoidance of the
required progressive intimacy; this, in turn, would result in imbalance in
reciprocity. Such an interruption of the chain of behaviours would be likely to
result in difficulties in the development of stable friendships. The interview data
provided information on the willingness of both groups to share personal
information with friends. The differences between the groups in their
willingness to make disclosures about their chronic condition have already been
presented, and the reticence of those with CAH to share information about their
condition highlighted. The friendship formation difficulties and social isolation
during schooldays were continued into adult life for those with CAH and
discussion about more general personal information and needs as adults were
also described as difficult by these women:

*I prefer to work things out for myself. I keep things to
myself- I don't tell anybody. I don't tell anyone what my
problems are.* (C08)

*I don't feel I give to other people, there is something in
the way.* (C03)

*No, it's not that I feel they can't help me, I just- I don't
know, I just can't seem to get myself to say that I need
help, 'Can someone please talk to me?' I can't do
that.* (C15)

*Difficult. I'm a very sort of 'in' person, I don't tell
anybody anything.* (C33).

The above quotations are representative of almost all the CAH group; only two
(10 per cent) women in this group were able to share their feelings with friends.
This finding was reversed for those with diabetes where over three-quarters
described a willingness to share their feelings and problems although this
willingness to communicate was frequently restricted to carefully selected individuals who were believed to be trustworthy:

Pretty good. I usually consider who I would want to tell what to. (D30)

I'm not the sort of person who would start yapping on about myself until I knew them very well. (D34)

The women with CAH were well aware of the requirement of reciprocity and that friendship carried with it the requirement of disclosure. However they did not believe that trustworthy individuals with whom they could share their feelings and problems were available and this resulted in withdrawal from friendships:

It might start off well, but I usually end up getting rid of them. (C03)

I'll listen to them, but there's no way I'll give anything to them about myself. (C09)

I've given up visiting friends - I'm not one of those people who needs people, I'm concentrating on my job at present. (C15)

They seem to have drifted a lot, my friends, they are not a big part of my life, not any more. (C28)

I can live without that [close friends] I can live with just acquaintances, I don't have to be dependent on people. (C41)

The importance of reciprocity for friendships implies that relationships also require the ability to act as an empathic receiver, as well as a giver, of information. If giving information about the self was perceived by the women with CAH as difficult, was the social skill of receiving others' personal disclosures also difficult? This did not appear to be so, very few women in
either group described difficulties in listening to, or in helping others and a high proportion expressed skill in this areas of social interaction:

- *If they came to me for help, or cried over the 'phone, or they were stuck in a jam, I would move heaven and hell to get to that person if it helped them.* (C03)

- *I can be very perceptive to people that I see regularly. I know when they've got something bugging them. If I can help I will.* (C13)

- *I've always been a person that others have come to with their troubles.* (D24)

- *Nobody's shy to share their feelings with me either.* (D30)

It is likely that this created an imbalance: a lack of reciprocity between the women with CAH and those with whom they interacted as friends. The women with CAH were very willing to listen to others and to help, but could not disclose information about themselves nor ask others for help. Such unevenness might increase difficulties in friendship development, especially in the establishment of long-term relationships. This problem did not appear to apply to those women with diabetes where levels of intimacy were more evenly balanced.

The inclusion of reciprocity as a component of friendship implicates broader issues than that of direct communication; reciprocity of need fulfilment is one such issue. Needs have to be clearly stated before others can consider them, and such a requirement may have been difficult for those who felt they had something to keep secret. During the interviews, the women were asked about their strategies for dealing with situations where their needs were not being met to their satisfaction.
The element of the fulfilment of needs

The reported strategies were classified into those described in active terms that were focussed on the need fulfilment, and those that were described in passive and unfocused terms. Table 12 lists the criteria descriptors for these two classes, and class examples are presented below:

**TABLE 12: Descriptors used in the classification of strategies for the fulfilment of needs**

<table>
<thead>
<tr>
<th>ACTIVE/FOCUSSED</th>
<th>PASSIVE/UNFOCUSED</th>
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<tbody>
<tr>
<td>Persistence</td>
<td>Withdrawal</td>
</tr>
<tr>
<td>Compromise</td>
<td>Surrender</td>
</tr>
<tr>
<td>Reasoning</td>
<td>Display of negative emotions</td>
</tr>
<tr>
<td>Re-state and discuss</td>
<td>Indirect manipulation</td>
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</tbody>
</table>

Although about one third of both groups felt that they frequently failed to 'get their own way', there were group differences in the strategies used. For those with CAH the most frequently used strategies were those classified as 'passive/unfocused'.

*I just walk in and say I've had enough, that's it.* (C09) ['withdrawal']

*I generally give in.* (C07) ['surrender']

*I tend to scream a bit.* (D02) ['negative emotions']

*I'm very good at showing my displeasure, say a few cutting remarks.... You need to find the hitting point, I'm very cunning.* (D09) ['indirect manipulation']

Within this passive/unfocussed classification, the CAH group were considerably more likely to use the strategies of 'withdrawal' and 'give in'. Together these strategies accounted for 90 percent of this group's passive/unfocused statements and their use resulted in needs remaining unfulfilled. For the diabetic group the majority reported the repeated use of active/focused strategies:
By stating how I feel about things, giving the pros and cons of why I think it should be, [that I should] have my own way. (D01) [re-state and discuss needs/reasoning]

I keep coming at it different ways, til I get my own way. (D09) [persistence]

I would be prepared to compromise if I was convinced by the others' arguments, discover what the positives and negatives were. (C37) [compromise/reasoning]

The very frequent use of 'withdrawal' and 'give in' strategies by those with CAH, again supports the conclusion that they were reluctant to engage in interactions which concerned their feelings and needs- they preferred to maintain a distance, even if this resulted in their needs remaining unmet.

In summary, more women in both groups reported current friendship difficulties than reported difficulties in childhood. Almost three-quarters of those women with CAH reported dissatisfaction with their current friendships compared with less than one quarter of those with diabetes. However, unlike those women with diabetes, the dissatisfaction with friendship networks of those with CAH was not accompanied by a willingness to enlarge their circle of friends. The two groups were similar in their construction of the meaning of friendship, and considered a friend as someone who offered companionship, shared activities, and was loyal and trustworthy. For the women with CAH, confidentiality, especially that concerned with personal information, was also an important essential criteria, as was the acceptance of their good and bad traits by friends. In relation to the giving of information', the CAH group described this as a very difficult task, and themselves as 'keeping themselves to themselves'. In contrast, they believed that they were good helpers and listeners. It was proposed that
such imbalances in the reciprocity between the friends might increase difficulties in friendship formation. The diabetic group's self perceptions were of themselves as open communicators about feelings and problems with those they trusted. Those with CAH unsuccessfully used 'passive/unfocused' strategies, particularly those of 'withdrawal' and 'surrender', to gain fulfilment of their perceived needs in contrast to those with diabetes who more frequently used strategies that were 'active/focused'. These findings support the proposal that the presence of a 'Skeleton-in-the-cupboard', necessitating as it does behaviours directed to keeping the secret, resulted in continuing difficulties in friendship formation and an unwillingness to develop new friends.

In this section, the discussion so far has centred on concepts of friendship formation and has considered the attributes that may be important for these relationships. With the onset of puberty, the experiences and skills learnt during childhood may be used to develop friendships that include a sexual component. These, in turn, may develop into more permanent partnerships. It is the women's views of their experiences in these areas that will now be discussed.

**The concept of the effects of the 'Skeleton-in-the-cupboard' on sexual partnership experiences**

The dimensions and elements of this third concept identified in the data are shown in Table 13.
### TABLE 13: Dimensions and elements relating to the effects of the 'Skeleton-in-the-cupboard' on sexual partnership experiences

<table>
<thead>
<tr>
<th>Dimension 1 - Sexual interest</th>
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<tbody>
<tr>
<td>element of dating</td>
<td></td>
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<tr>
<td>element of the perception of sexual interest</td>
<td></td>
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<tr>
<td>element of masturbation</td>
<td></td>
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<tr>
<td>element of the extent of sexual experience</td>
<td></td>
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<tr>
<td>element of reasons for engaging in sexual behaviour</td>
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<tr>
<td>element of stimuli eliciting sexual responses</td>
<td></td>
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<tr>
<td>element of satisfaction with sexual activities</td>
<td></td>
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<tr>
<td>element of physical problems due to condition</td>
<td></td>
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<tr>
<td>element of the effects of anxiety on sexual activities</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Dimension 2 - Attitudes towards marriage</th>
<th></th>
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<tbody>
<tr>
<td>element of the value of marriage</td>
<td></td>
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<tr>
<td>element of the requirements of a good marriage</td>
<td></td>
</tr>
<tr>
<td>element of housewife/career choice</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Dimension 3 - Attitudes towards parenthood</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>element of the value of parenthood</td>
<td></td>
</tr>
<tr>
<td>element of knowledge about condition effects on pregnancy</td>
<td></td>
</tr>
<tr>
<td>element of knowledge about genetic issues</td>
<td></td>
</tr>
</tbody>
</table>

The discussion in this section will cover the dimensions and elements identified in the interview data that related to sexual interest and attitudes towards marriage and parenthood. A sexual interest experience, commonly beginning during the adolescent years, is 'dating'.

**The element of 'dating'**

'Dating' is a complex collection of activities that include the development of increasingly intimate relationships and the experiences of sexual behaviours (Swensen 1972). Although dating does occur in the teenage years between same-sexed pairs, and this issue is discussed in a later section, the initial discussion as an introduction to the sensitive areas of sexual experience was based on heterosexual relationships. Dating - defined here as arranged meetings with one person of the opposite sex with the understanding that sexual contact,
however limited, might occur-usually begins in the early teen years. For example, in their in-depth qualitative study of 65 young newly married couples, Mansfield and Collard (1988) reported that the recalled mean age for start of dating was 14 years. In the present study, the recall by the women of their age when beginning dating was not significantly different for the two groups (t=1.629, df = 34 p >0.05, 2-tailed) with the mean age for the combined groups being 16.4 years. This is older than that of Mansfield and Collard's sample, and compares with the results of Money and Schwartz (1977) who also found their group of CAH girls to be late in starting to date. The similarity between the diabetic and the CAH groups suggests that having a chronic medical condition rather than specifically having CAH, may have an influence on the age at which dating begins.

*The element of current perception of levels of sexual interest*

Dividing the sample at the mean recalled age of the start of dating for the groups combined, resulted in differences in the women's statements about their current interest in sexual activities. 'Late-daters' (recall of dating later than the total sample mean) in both groups, and 'early-daters' (recall of dating earlier than the total sample mean) in the diabetic group, saw their current interest in sexual activities as 'average' compared to their friends and acquaintances without a chronic condition. By contrast, early-dating CAH women described themselves as currently less interested in sexual activities than their normal peer group. This reported lack of interest is reflected in the fact that none of the eight (42 per cent) early-dating CAH women had boyfriends or partners at the time of interview. In contrast, eight (47 per cent) of women with diabetes had dated
early and six of the eight (75 per cent) had steady partners and the remaining two, boy friends. This represented a very different pattern for the two groups. Almost half of late-daters in both groups reported having partners at the time of interview.

These data suggest that the CAH women who had dated early may have a particular pattern of difficulties in this area, and it is of importance to understand why this might be so. Dating is an anxiety provoking activity for all women (Kelly 1984). The early physical maturity and development of the secondary sexual characteristics frequently seen in CAH girls may have been interpreted by themselves and by the young males in their environment as signals of a readiness for sexual experience. However, the genital abnormalities, the experience of surgery and the general difficulties in open communication may have made the young women less able to deal with the intimate behaviours required during dating. These early-dating CAH women described themselves as 'loners' and 'shy', and the anxieties arising from the perceived need to be intimate may have resulted in the curtailment and avoidance of intimate heterosexual relationships. Although late dating women with CAH may have experienced similar physical development patterns, the later start in dating may have allowed development of skills that enabled greater control in sexual situations and therefore less anxiety. Early dating women with diabetes would not have the same anxieties about genital normality and therefore would have different experiences.
The data provided many examples from those who were early-daters of later reticence to engage in intimate relationships. The following quotes from early-dating women with CAH clearly exemplify this point:

*I did [end the relationship], it was getting too involved. I wasn't interested in sex.* (C15)

*I got cold feet I think, I got out.* (C08)

*After the first, I never really had a date for years. I liked talking but they were only interested in sex.* (C09)

*None [boy friends] at present. Sometimes I feel lucky not to have anyone who's moaning, but I see people and I think it would be nice to have that experience, but most of the time it doesn't bother me. I get frightened getting too involved, I get worried and back off.* (C33)

The women with diabetes remembered such curtailment decisions as being less one-sided with some having been taken by themselves, some by the boys, and some jointly:

*He did, he found someone else!* (D04)

*I did, it got too serious, ...... he wanted it to be sexual and I wasn't ready.* (D08)

*We drifted apart, it was mutual.* (D14)

The ability to establish and maintain relationships with boy friends is clearly important in providing opportunities for engaging in sexual behaviours. Further elements comprising the dimension of sexual interest are given in Table 13 and are discussed below.

*The element of masturbation*

Engaging in masturbatory behaviour may also be an indication of sexual interest, and the data suggested that the women with CAH were inexperienced in this area. Less than half of the CAH group had engaged in this activity; those who
had started in their early teens. By comparison, just over three quarters of
the diabetic women had masturbated since their early teens and this
approximates to normative levels as reported by Hite (1976) and Kinsey,
Pomeroy, Martin and Bebhard (1953). An important finding was that the CAH
women commonly spoke about masturbation as a necessary medical procedure,
rather than primarily a sexual activity:

[masturbation was necessary] to keep it [the vagina]
open. I used to be nervous of it closing up. (C04)

I have done so, everybody does, I used to have to, I would
explore- what had [the surgeons] done? (C52)

Even now, I've never thought about it for enjoyment. (C41)

The element of sexual experiences with a partner

Sexual experience was further examined by asking the women which of a
number of specific sexual behaviours with partners they had ever experienced.
The behaviours were of three related classes: those occurring in the foreplay
stage of sexual encounters and which are believed to increase arousal (Masters
and Johnson, 1966: Beck and Barlow, 1984), those concerned with vaginal
penetration and lastly, orgasm itself. Table 14 shows the grouping of these
classes of sexual experiences. The experiences are listed in descending order of
the ratio of the number reporting experience in the diabetic group over the CAH
group.
TABLE 14: Sexual experiences reported by women in both groups

<table>
<thead>
<tr>
<th>Q. Order</th>
<th>SEXUAL ACTIVITY</th>
<th>CAH (N=17)</th>
<th>DIABETIC (N=17)</th>
<th>Ratio to 1 Diabetic/CAH</th>
</tr>
</thead>
<tbody>
<tr>
<td>25</td>
<td>You dressed up in a particular way</td>
<td>2</td>
<td>5</td>
<td>2.5</td>
</tr>
<tr>
<td>24</td>
<td>Your partner dressed up in a particular way</td>
<td>1</td>
<td>2</td>
<td>2.0</td>
</tr>
<tr>
<td>23</td>
<td>You used sex toys</td>
<td>1</td>
<td>2</td>
<td>2.0</td>
</tr>
<tr>
<td>9</td>
<td>Deep kissing with your tongues touching</td>
<td>10</td>
<td>16</td>
<td>1.6</td>
</tr>
<tr>
<td>3</td>
<td>You kissed your partner's face, arms, legs, back or other non-sexual areas when dressed</td>
<td>11</td>
<td>17</td>
<td>1.5</td>
</tr>
<tr>
<td>12</td>
<td>Your partner caressed your face, arms, legs, back or other non sexual areas with clothes on</td>
<td>11</td>
<td>15</td>
<td>1.4</td>
</tr>
<tr>
<td>8</td>
<td>Hugging each other when naked</td>
<td>12</td>
<td>16</td>
<td>1.4</td>
</tr>
<tr>
<td>11</td>
<td>Your partner caressed your genitals without your clothes on</td>
<td>12</td>
<td>16</td>
<td>1.4</td>
</tr>
<tr>
<td>7</td>
<td>Your partner caressed your breasts with your clothes on</td>
<td>10</td>
<td>13</td>
<td>1.3</td>
</tr>
<tr>
<td>13</td>
<td>Your partner caressed your face arms, legs, back or other non sexual areas with you naked</td>
<td>12</td>
<td>15</td>
<td>1.2</td>
</tr>
<tr>
<td>6</td>
<td>You caress with your hands your partner's genitals (penis and balls) with clothes on</td>
<td>10</td>
<td>12</td>
<td>1.2</td>
</tr>
<tr>
<td>10</td>
<td>Your partner caressed your genitals (clitoris vagina pussy) with clothes on</td>
<td>9</td>
<td>11</td>
<td>1.2</td>
</tr>
<tr>
<td>15</td>
<td>You used your tongue or lips to stimulate your partner's genitals</td>
<td>10</td>
<td>12</td>
<td>1.2</td>
</tr>
<tr>
<td>1</td>
<td>You caressed with your hands your partner's face, arms,legs,back or other non-sexual areas when you naked</td>
<td>15</td>
<td>16</td>
<td>1.1</td>
</tr>
<tr>
<td>2</td>
<td>Hugging each other with your clothes on</td>
<td>13</td>
<td>14</td>
<td>1.1</td>
</tr>
<tr>
<td>4</td>
<td>Kissed with mouth closed</td>
<td>12</td>
<td>13</td>
<td>1.1</td>
</tr>
<tr>
<td>16</td>
<td>Your partner used his tongue or lips to stimulate your genitals</td>
<td>11</td>
<td>12</td>
<td>1.1</td>
</tr>
<tr>
<td>21</td>
<td>Your partner put his penis inside your anus (back passage)</td>
<td>0</td>
<td>3</td>
<td></td>
</tr>
<tr>
<td>14</td>
<td>Your partner put a finger into your vagina</td>
<td>7</td>
<td>15</td>
<td>2.1</td>
</tr>
<tr>
<td>17</td>
<td>Your partner put his penis inside your vagina</td>
<td>12</td>
<td>17</td>
<td>1.4</td>
</tr>
<tr>
<td>22</td>
<td>Your partner ejaculated (came) in your mouth</td>
<td>1</td>
<td>7</td>
<td>7.0</td>
</tr>
<tr>
<td>20</td>
<td>Your partner ejaculated (came) outside your vagina</td>
<td>4</td>
<td>10</td>
<td>2.5</td>
</tr>
<tr>
<td>19</td>
<td>Your partner came in your vagina</td>
<td>4</td>
<td>14</td>
<td>2.3</td>
</tr>
<tr>
<td>18</td>
<td>You had an orgasm (came)</td>
<td>7</td>
<td>15</td>
<td>2.1</td>
</tr>
</tbody>
</table>

* Missing data for two woman.

Overall comparison of CAH and Diabetic women reporting listed sexual activities by Wilcoxon Matched-Pairs Signed-Rank Test, T=0, N=25, p<0.000009 (2-tailed).

The analysis of reported experiences in Table 14, clearly supports the greater experience of those women with diabetes for all behaviours. Although more of the diabetic women had partners at the time of interview, this did not explain these reported differences as the women were asked about total and not current experience. The group differences are illustrated by the following quotes:

*I'm not too keen on kissing him down there. He doesn't kiss me there either. (C04)*
Deep kissing makes me feel sick. (C15)

I enjoy a broad range of things. (D28)

I enjoy sex with more than one person at a time. I enjoy most things. (D30)

The lower levels of the sexual experiences of women with CAH included all of the three classes of behaviours, those that increase arousal, those of penetration and of orgasm. It has been proposed (Beck and Barlow, 1984) that anxiety may be a major inhibitor of sexual arousal. In addition to a generally high anxiety level for all sexual activities of those with CAH, low arousal may also be experienced because of the reluctance of these women to engage in intimate interactions where their partners may become aware of their genital abnormalities. This anxiety may also partially account for the lower level of experience for both finger and penile penetration, and the issue of the effects of anxiety on sexual activities will be taken up later in the report. It is also possible that successful penetration may be a function of the type of surgery undergone by women with CAH; this issue will be further examined under the element of physical problems due to the condition.

There is very little normative data on the range of sexual activities and it is difficult to make direct comparisons of sexual experience as the age ranges and the descriptions of the activities are rarely entirely comparable. A general population study by Breakwell and Fife-Schaw (1992) however reported on three similar behaviours: deep kissing, vaginal penetration and anal penetration (see Table 15).
TABLE 15: Comparison of percentages of those reporting sexual experiences in Breakwell and Fife-Schaw (1992), the diabetic and the CAH groups

<table>
<thead>
<tr>
<th>SEXUAL EXPERIENCE</th>
<th>BREAKWELL and FIFE-SCHAW N=1315</th>
<th>DIABETIC GROUP N=17</th>
<th>CAH GROUP N=17</th>
</tr>
</thead>
<tbody>
<tr>
<td>Deep kissing</td>
<td>92</td>
<td>94</td>
<td>59</td>
</tr>
<tr>
<td>Penile penetration</td>
<td>70</td>
<td>100</td>
<td>71</td>
</tr>
<tr>
<td>Anal penetration</td>
<td>9</td>
<td>18</td>
<td>0</td>
</tr>
</tbody>
</table>

The samples of women in the present study are older than those in Breakwell and Fife-Schaw's female sample and, in line with these researchers' findings of increased sexual experience with age, the diabetic women are somewhat more sexually experienced in the behaviours examined in both studies. By contrast, the women with CAH, who are also older than Breakwell and Fife-Schaw's sample, are not.

Table 14 also indicates lower incidence of orgasmic attainment by those with CAH. The issue of the experience of orgasm is of particular interest in relation to women with CAH. The finding of this area and the information obtained from the discussion about masturbation both suggested that the women with CAH were less likely than those with diabetes to have experienced orgasm. The women with diabetes who had masturbated (over three-quarters) and the smaller percentage (less than half) of those with CAH having engaged in this activity, were asked if they had masturbated to orgasm. About half in each group reported that they had. These women, and those who had not but reported orgasm during sexual activity with a partner, were asked to give a description of 'what it felt like'. These descriptions were classified into either high arousal or orgasm (Hite 1976) by two independent, experienced
psychosexual therapists who were blind as to group membership of those providing the descriptions (see Appendix VII).

Seventeen of the nineteen women in the CAH group reported they had had the opportunity to experience orgasm either through masturbation or sex with a partner. Of these, thirteen (76 percent) reported experiencing orgasm. The independent classifiers failed to agree on one of these descriptions which was dropped from the analysis, and of the remaining twelve descriptions from the women in the CAH group, seven (58 per cent) were classified as descriptions of orgasm. By contrast, the entire diabetic group had had the opportunity to experience orgasm through masturbation or with a partner, and fifteen (88 per cent) of these women reported that experience. All of these were classified as descriptions of orgasm. Although in this study the percentage of women with CAH who reported orgasm was considerably greater than the proportion of the 1977 study by Money and Schwartz (22 per cent), also based on self-reports, it is still considerably less than for the diabetic group. There are two possible explanations for this lower level of orgasm: that it is due to the medical aspects of the condition, the genital abnormalities and the surgical repair, or to anxiety arising from engaging in this intimate sexual behaviour.

The relationships between the effects on the ability to attain orgasm of the extent of genital abnormality or corrective surgery are difficult to examine, as more severe levels of masculinisation require more extensive surgery. It is therefore not possible to separate out these two effects. Additionally, the sample of women with CAH in the present study, being diagnosed early in the
understanding of the condition, had received similar levels of corrective surgery across the Prader classification. Although it was therefore not possible to assess the relationship between orgasmic experience and surgery or level of abnormality, examination of the data identified a number of areas that might further contribute to the understanding of non-achievement of orgasm by these women. These further areas will be reported under the remaining five elements of this concept: reasons for engaging in sexual behaviour, stimuli that elicited a sexual response, satisfaction with sexual behaviour, physical problems due to the condition and the effects of anxiety of sexual activities.

Seventeen of the nineteen women in the CAH group reported that they had had the opportunity to experience orgasm either through masturbation or sex with a partner. Of these, thirteen (76 percent) reported experiencing orgasm. The independent classifiers failed to agree on one of these descriptions which was dropped from the analysis; of the remaining twelve descriptions from the women in the CAH group, seven (58 per cent) were classified as descriptions of orgasm. By contrast, the entire diabetic group had had the opportunity to experience orgasm through masturbation or with a partner, and fifteen (88 per cent) of these women reported that experience. All of these were classified as descriptions of orgasm. Although in this study the percentage of women with CAH who reported orgasm was double the proportion of the 1977 study by Money and Schwartz (22 per cent), also based on women's self-reports, it is still considerably less than for the diabetic group. There are two possible explanations for this lower level of orgasm: that it is due to the medical aspect
The element of reasons for engaging in sexual behaviour

Although about one quarter of each group stated that they occasionally 'had sex' because their partners expected it, both groups felt the most common motivation was their own sexual need. For all the women this view was accompanied by a very strong statement that sexual activity was dependent on the existence of a close emotional relationship:

- cos I felt I loved him enough to let him into my body, your body has to be shared with somebody that you love. (C37)
- I have to know them, trust them, for a long time. (C52)
- I've never been able to distinguish between sex and love. (D30)
- Women have sex for love. (D23)

The element of stimuli eliciting sexual responses

Such reasons for making love are in line with normative female gender stereotypes (Hite 1976), with both groups of women reporting the most important sexual stimulus to be that of a 'special person'. Interestingly, only the CAH women mentioned male characteristics such as 'tight bums', 'muscular shoulders' and 'hairy chests' as stimuli that might elicit sexual interest or behaviour. This focus by the CAH women on male physical characteristics may reflect a more general conceptualisation of 'sexuality' as external physical attributes. Two factors may have contributed to this: first, the frequent
experiences of the CAH women at the hospital clinics where there was a strong focus on the external genitalia. Second, although these women subscribed to the view that sex should be part of a close relationship, the difficulties they had with such relationships may have led them to less threatening stimuli such as external physical characteristics.

All of the women with diabetes were very clear about their lack of sexual interest in other women:

- "I can't say that any woman has ever done anything for me." (D29)
- "The whole idea kind of turns me off." (D23)
- "No, never." (D34)

Two (11 percent) of the women with CAH had experienced lesbian relationships, and another three (16 percent) expressed a strong sense of sexual appreciation of other females. They described considering what it would be like to have lesbian relationships, although they were very hesitant about acting on these thoughts. This hesitancy was attributed to an awareness of pressure against such behaviour from their families and friends:

- "If I see someone and I think 'she's pretty', I don't know- I know it's sort of wrong, you just don't do it do you? In our family that sort of thing is frowned upon, when you have these sort of thoughts you don't share them with anybody 'cos they'll think you're strange." (C33)

- "You look at a woman's body and think 'she's lovely', but I'm not sure about wanting to touch." (C01)

- "Some other female once said, and I agree with her, that another female would be able to please a woman." (C13)
It is difficult to directly compare these results to available normative data due to differences in age and marital status and to variations in definitions of homosexuality (Hedblom 1973; Hite 1976; Kinsey et al, 1953). Overall, the incidence of homosexual behaviour among women with CAH in the present study is likely to be similar to that expected in the general population. It is lower than the 29 per cent in Money and Schwartz's (1977) CAH sample; it is also lower than the 22 per cent reported by Dittmann et al (1992) but their definition of homosexual orientation included both behaviour and fantasy. The figure obtained was, however, higher than the 5 per cent reported for women with CAH by Mulaikalk et al (1987), and those in the study by Zucker et al (1996). Zucker et al did not find significant differences in sexual experiences with other women for those with CAH compared to their unaffected sisters or cousins. Hite (1976) reported that women in her sample frequently mentioned sexual appreciation of other women, but that this did not predict engagement with lesbian relationships. The fact that women with CAH may be particularly anxious about penetrative intercourse could make it more likely that they would welcome homosexual relationships. Alternatively, their general anxiety about sex might inhibit both heterosexual and homosexual relationships. These social and psychological issues that surround CAH make it difficult to interpret sexual preference in terms of any single factor.

The element of satisfaction with sexual activities

There is obviously no absolute appropriate level of sexual activity, and there are likely to be wide individual differences in the levels that are held to be satisfactory. Both groups of women studied here might be satisfied with their
current levels even if these seemed low, particularly as the women with CAH reported low interest in the opposite sex. To gain more information on this issue, the women were asked to rate their level of satisfaction on a 5-point scale where low ratings indicated dissatisfaction. The median satisfaction rating for the CAH and the diabetic groups were 3 and 4 respectively; an overall comparison of the distribution of scores for the two groups showed that the satisfaction ratings of the women with CAH were significantly lower than those of the women with diabetes (Wilcoxon's rank-sum test, W=205, N=16, p=.05, 2-tailed). The two groups of women also reported different kinds of necessary changes to increase their level of satisfaction. The different responses reflected the restricted opportunities for having sex with a male partner and the preoccupation with having 'successful' intercourse of those with CAH, whereas the women with diabetes would have liked an extension of their current sexual activity:

If I could have sexual intercourse with a man inside my vagina. (C01)

As soon as I manage to have sex. Oh my God, I'm 19 and still a virgin! (C07)

I'd like a more active sex life, more of it. (C33)

More time to enjoy it. (D09)

We don't have enough time for it, in the evenings we are too tired and we settle for a cuddle. (D14)

Element of physical problems due to the condition

It is clearly possible that for women with CAH the genital abnormalities and their surgical correction may have resulted in physical difficulties during sexual
intercourse, problems connected for example, with penetration, lubrication, residual pain from tissue repair and reduced sensitivity of the clitoris.

Just over half (56 per cent) of the CAH women who had experienced sexual intercourse reported problems with penile penetration. Once again the small number of simple virilisers in the sample and the similarity of surgical procedures did not allow an examination of the relationship between surgical procedures and penetration difficulties. However the problems did not appear to be related to Prader levels of masculinisation which is in line with the results of the study by Mulaikal et al (1987). In the current study, the penetration problems decreased over time for those having regular intercourse. The women themselves attributed their difficulties to tightness or shortness of the vagina or to their operations. For two women in the CAH group failure of penetration, or anxiety that this would occur, had resulted in the complete avoidance of heterosexual activities.

[I've not had sex]...because like......the fear, the fear that the man would find out, he'd not get it in.(C01)

Penetration was not a reported as a problem by the diabetic group with only two (12 per cent) mentioning the occurrence of this difficulty in the early days of their sexual experience; this they attributed to being a virgin.

The first few times of sexual intercourse were recalled as painful by three quarters of the CAH and half of the diabetic group:

It wasn't a very happy experience, it was very painful, although he was very nice. I felt such a failure- it was
hurting me, I wasn't enjoying it- he seemed to be having a whale of a time!(C09)

Pain was persistent, however, only for those women with CAH; those with this condition who reported early experience of pain also reported current pain:

Yes, even now when he puts his penis inside. I've nearly cried with it sometimes.(C10)

As with penetration difficulties, current pain did not appear to be related to level of masculinisation. Equal proportions of those women who reported current pain had been classified in the 'mild' and in the 'severe' Prader (1954) categories. As might be expected, those women with CAH who reported current pain also reported current penetration problems. The women with diabetes reported a different pattern of pain with no suggestion of a relationship between early and current pain.

The attributed cause of the pain also differed for the two groups. While the women with CAH perceived it as due to the size of the vagina or 'something there, blocking him getting in', the women with diabetes attributed their pain to dryness of the vagina:

It seems to vary with the position, so I don't know what's in the way.(C03)

I feel my womb has dropped- it seems to be hitting something, where you get more penetration, you get more pain.(C41)

Only when it's dry.(D08)

When I'm tense and I don't get wet, it is painful.(D25)
The sensitivity of the clitoris may be reduced or eliminated by the surgical procedures of early childhood in females with CAH. Whereas all the diabetics perceived their clitoris to be sensitive, seven (41 per cent) of the sixteen CAH women who had masturbated or had sexual intercourse reported a lack of sensitivity. The report of sensitivity or insensitivity was not related to the report of pain, nor to the level of masculinisation. This latter finding may again be due to the radical surgery (clitoridectomy) experienced by the women in this study across levels of masculinisation. Both orgasmic and non-orgasmic women reported clitoral insensitivity. Other research, for example Lightfoot-Klein (1989), also suggests that climax is not dependent on perceived clitoral sensitivity and that fully circumcised infibulated females are capable of attaining orgasm. In the present study however, those women reporting clitoral insensitivity were twice as likely to have penetration problems; it is possible that this difficulty may be partly due to decreased arousal leading to decreased vaginal lubrication and therefore increased risk of penetration difficulties.

Lubrication of the vagina is an essential factor in ease of penetration during intercourse, and is also believed to be an indicator of arousal. Arousal and, therefore, lubrication are inhibited by anxiety (Masters and Johnson 1966; Beck and Barlow 1984). As the data already reported here suggest that sexual anxiety may be common in CAH women, it might be expected that these women would experience lubrication difficulties. Nine (60 percent) of the fifteen women with CAH and six (35 percent) of the seventeen with diabetes who had reported sexual intercourse also reported lubrication problems. The women
with diabetes, however, believed that 'dryness was common in diabetics', and all of them had used artificial lubricants to alleviate the problem:

Yes, I use jelly sometimes, I know diabetics have problems. (D04)

Yes (don't lubricate). It upsets me. Its only to be expected. At first I didn't know what had happened, and then I thought it is because I've only had two blokes, and D said to me, in the 'You and Me' magazine there was an article on diabetics, and he said they don't produce the lubricant, they are very dry. He'd read it and he told me! I thought it great, fantastic, at least I know there's nothing wrong with me. And he said, 'If at first we don't succeed, we'll try and try again'. ....I think I should (use jelly) but I'm embarrassed to, but it....I'm waiting for a free offer. (D08)

This practical approach to a medically attributed problem was not apparent in the women with CAH who had neither a medical nor a psychological explanation for their lack of lubrication and did not appear to have considered that there might be a remedy:

I think I do (get wet), but I can never tell if it's him or me doing it. I'm often dry, but I've never used jelly. (C10)

I do get wet, and then I dry up quickly. It takes a bit of time...... You just have to work at it. (C41)

As expected, those with lubrication problems were twice as likely to have penetration difficulties compared to those for whom lubrication was not a problem.

In considering the relationship between the physical problems experienced by the CAH women in this study, and their levels of masculinisation, it is necessary to take a number of factors into account. The sample is small and includes a number of women in the CAH group who have not had sexual experience. The
fact that some aspects of surgery were similar for women across levels of masculinisation makes any relationship between this and sexual problems difficult to examine. Data from those women who had had a clitoridectomy and relevant sexual experience (see Table 16) show that the numbers reporting the presence of current pain during intercourse are about the same as those reporting its absence; similarly, almost equal numbers reported experiencing orgasm as not experiencing it. More women, however, reported problems with penetration, lubrication and clitoral sensitivity than reported the lack of such problems. As can be seen from Table 16, however, the numbers in each category are so small that it is difficult to draw clear conclusions. Further investigation is needed before outcome relationships between levels of masculinisation, type of surgery, and sexual problems can be clarified.

TABLE 16: Physical problems for sexually experienced women with CAH who had a clitoridectomy

<table>
<thead>
<tr>
<th>Current Pain</th>
<th>Penetration problems</th>
<th>Lubrication Problems</th>
<th>Orgasm Problems</th>
<th>Clitoral Insensitivity</th>
</tr>
</thead>
<tbody>
<tr>
<td>N.(%)</td>
<td>N.(%)</td>
<td>N.(%)</td>
<td>N.(%)</td>
<td>N.(%)</td>
</tr>
<tr>
<td>+ Problem</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Reported</td>
<td>6(55)</td>
<td>7(64)</td>
<td>8(73)</td>
<td>5(56)</td>
</tr>
<tr>
<td>+ Problem</td>
<td>5(46)</td>
<td>4(36)</td>
<td>3(27)</td>
<td>4(44)</td>
</tr>
<tr>
<td>not reported</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>* No Experience</td>
<td>4(27)</td>
<td>4(27)</td>
<td>4(27)</td>
<td>6(40)</td>
</tr>
<tr>
<td>Totals</td>
<td>15</td>
<td>15</td>
<td>15</td>
<td>15</td>
</tr>
</tbody>
</table>

+ Expressed as percentages of those having relevant sexual experience
* Expressed as percentages of total number

The element of the effects of anxiety on sexual activities

It was common for the women with CAH to conceptualise sexual intercourse as 'a hurdle to be got over', and the majority perceived their surgery to be implicated in the outcome of sexual activity. Women remembered extreme worries that they would not be able to achieve penetration:

Well, I've tried- he couldn't even begin to get in. (C07)
At the beginning, I was so tensed up anyway- there seems to be a block even now, as if it's not long enough. (C41)

It wasn't at the beginning, but it's not deep because of scar tissue, but he's getting further in each time. (C04)

I had the feeling that I couldn't have sex, cos of the ops, I wouldn't be able to get him in. (C03)

The first time, I don't think he could get it in at all. (C09)

Although it is likely that most women experience anxiety about the beginnings of sexual experience, the strong expressions of anxiety by the women in the CAH group were not echoed by those in the diabetic group. The anxiety of the women with CAH appeared to be focussed on their genital abnormalities, actual or perceived, and there was considerable hesitancy about the success of the surgery. These two factors may have resulted in increased awareness of possible difficulties:

I'm very worried about scarring from the operation- it's not normal, it looks knocked about, I've always got the feeling that it's going to close up, and is he going to freak or something? (C33)

My boy friend says I'm small, but he said it's not surprising cos I'm a virgin. I wanted to be able to do it [penetration] off my own bat without using dilators. They said you had to do it gradually rather than go 'bash', or you would cause a great deal of damage. (C13)

The women with CAH additionally expressed anxiety about the acceptability of their genitalia to their partners. They recalled their considerable relief when no comment was made about their 'normality':

......the fear that the man would find out, would he say 'My God, what's the matter with you'? (C01)

I must be alright cos he didn't say anything. (C33)
He said I was doing it properly, but I don't know if I was.
I was reassured when he didn't make any comments about
it being 'normal'.(C13)

I was worried 'cos I've had the ops. The first time I didn't
know if it was going right - I didn't want to be a failure.
You are not normal down there, and men are worldly, and
oh! is he going to freak out sort of thing? (C33)

It was not surprising that this issue of normality was central in these women's
lives, and it is not difficult to understand their uncertainty about the acceptable
state of their genitalia:

How do you know what you are supposed to look like and
how it is supposed to BE down there, you never see
anybody else do you?(C52)

I used to put my fingers inside my vagina and say 'Is this
supposed to be this low? and look in the mirror and
think, 'Is this normal?'(C41)

Neither those with CAH nor those with diabetes felt that they could talk to their
hospital doctors about any sexual problems. Nor did they see it as appropriate
to discuss their anxieties about genital abnormalities with either the hospital
doctors or their general practitioners. There was a general attitude in the CAH
group that nothing could be done and there was no one to go to for help. A
popular solution for any sexual difficulties or anxieties was to wait for 'the right
man' to come along; it was believed by them that this event would alleviate any
problems, and everything would then be achievable:

I'm not actually going out with anybody, I'm still too tight
to have sex. When I settle down in a steady relationship,
it will be alright.(C08)

In summary, the findings of the dimension of 'sexual interest' raise a number of
concerns that require further consideration. For the women with CAH,
difficulties during childhood in establishing and maintaining friendships with both girls and boys, may have provided fewer opportunities to learn appropriate social rules for intimacy and partnership formation (Duck, 1977; Argyle and Henderson, 1985). These difficulties may have been even greater and have led to disengagement of relationships for those who began heterosexual dating early when they were perhaps less socially experienced and able to deal with the intimacy that is central to sexual relationships.

Compared to the group of women with diabetes those with CAH were less sexually experienced, were more reluctant to establish relationships with the opposite sex and were less satisfied with their sexual activities. Remedies for increased satisfaction reported by the two groups also differed; those with CAH required increased opportunities and those with diabetes more time to enjoy their sexual experiences. The women with CAH also reported higher levels of difficulties with penile penetration and a pattern of persistent pain during intercourse. Fewer women with CAH than with diabetes masturbated or attained orgasm. Adequate lubrication did seem to be a difficulty for both groups; for the diabetic group this may suggest some parallel with the neuropathy seen in diabetic men.

It was of particular concern that anxiety about a number of areas involving sexual activities was a problem for the women with CAH. While the pain and the difficulties they experienced with penetration may have led them to avoid sexual activities, such experiences would also reinforce concerns about the normality of their genitalia and be construed as confirmation of 'not being like
other women'. Such concerns are, in fact, common among women (Hite, 1976), but are likely to be given added force among those with CAH by their medical history, even if their partners do not comment on any abnormality. In addition, although both the CAH and the diabetic groups expressed the ideal of sexual intercourse as being part of a loving relationship, those with CAH frequently construed intercourse as a negative event. Some spoke of it as 'a hurdle to be got over', others as 'invasions', 'violations of my body' and 'rape':

\begin{quote}
It wasn't a happy experience, I felt such a failure, it was hurting me and I wasn't enjoying it. At the back of my mind was the experiences at hospital, it was all like rape. (C09)
\end{quote}

\begin{quote}
I said 'I'm too scared'. I'm terrified of being touched - I just up and cry. I feel very scared to have sex. It's probably very nice but I've got bad memories so I don't want to share my body - sex represents something bad, I feel it's an invasion of my body. (C15)
\end{quote}

It is not clear to what extent these constructions reflect the behaviour of the male partners of the women with CAH. Nevertheless it is not surprising that the women with CAH should associate their genitalia with unwanted procedures rather than primarily with sexual need or satisfaction. The early surgery, vaginoplasty at adolescence and repeated vaginal examinations during visits to outpatient clinics make this very understandable. Indeed it was notable that the women with CAH also used the terms 'rape' 'invasion' and 'violation' when talking about the vaginal examinations and other procedures carried out during the visits to paediatric clinics. They frequently drew parallels between their response to hospital experiences and to sexual activities. Money and Schwartz (1977) reported similar findings.
The extensiveness and invasiveness of the medical and surgical procedures are, in addition, likely to have contributed to the generally held view of the CAH group that their sexual problems were entirely a result of CAH. In addition, in contrast to the diabetic group, these procedures were likely to be implicated in the striking failure of those with CAH to consider non-medical influences or methods of alleviation. This all-or-none approach was echoed in the belief, expressed only by those with CAH, that 'the right man' would somehow make everything achievable. Such a belief is of course strongly encouraged by women's romantic fiction (Coward, 1984), and in this case it appeared to be related to the women's sense of helplessness about their problems.

While women with CAH do appear to have some sexual difficulties which are particular to their condition, it is difficult to interpret the distribution of reported sexual problems across initial levels of masculinisation because similar surgical techniques were applied across levels of abnormality in the early days of treatment. Further investigation using larger numbers of mildly masculinised women who have received less radical surgery is essential before the direct effects of physical abnormality and surgery on sexual functioning can be clarified.

These data from the women support the proposal that the 'Skeleton-in-the-cupboard' had an effect on sexual activities of those with CAH. The effects appeared to be related to two components: the belief that sexual intercourse, particularly penetration would be difficult or even impossible because of the vaginal abnormalities and surgery, and the response of
partners to the physical appearances of their external genitalia. The medical examinations and focus on vaginal and reproductive functions during the teenage years, may also have resulted in a perception of sexual behaviours as violations rather than pleasures. Although both groups of women expressed reluctance to ask for professional advice on sexual activities and problems, for those with CAH this reluctance was augmented by the more general feelings of isolation from all sources of information and understanding about their condition and its effects (May, 1994). This isolation and the belief that their condition was not acceptable as a topic for discussion, that the skeleton must be kept hidden, culminated in the feeling that there was no practical remedy for problems connected with sexual intercourse. Addressing these psychological aspects of CAH will necessitate the provision of opportunities for open discussion, and this issue will be addressed more fully later in the thesis.

Sexual activities between partners are commonly part of relationships that are construed as permanent or semi-permanent. Such relationships may or may not be recognised by marriage ceremony, but their essential characteristic is that of a long-term commitment. In the light of the difficulties reported in this study for those with CAH, attitudes towards such long-term relationships were further examined. It is the findings on these dimensions (see Table 9) that will be discussed in the following section on marriage and careers.
The dimension of attitudes towards marriage and careers

Table 13 lists the four elements identified in the data that were related to the effects of the 'Skeleton-in-the-cupboard' on the second dimension of attitudes towards marriage. The elements were those of the value and requirements of marriage, and the choice between the role of housewife or career.

The element of the value of marriage

It is becoming increasingly common for couples to 'live together' in a committed relationship without undergoing a formal marriage ceremony; the General Household Survey of 1990 found that 17 per cent of women chose to co-habit rather than get married. In the present study therefore, the term 'marriage' was defined as co-habiting in a stable relationship with or without a marriage ceremony.

Attitudes towards marriage, particularly those of women between the ages of 18 and 37 years of age, are changing. A 1955 survey of such attitudes by Gorer concluded that both sexes regarded the most important element of marriage to be the efficient fulfilment of culturally stereotyped gender roles, namely 'breadwinner' for males and 'homemaker' for females. Nearly twenty years later, these attitudes had changed; a second British survey (Gorer 1971) found the most important element to be the need for husbands and wives to like each other. The criteria for a successful long-term relationship had moved from a social role to an intimate personal scenario, with the couple now requiring a reciprocity of respect and nurturance, and sustained sexual satisfaction, a move which Giddens (1992) has considered to be a generic restructuring of intimacy.
Both the inevitability and the duration of marriage have also changed. With four out of ten marriages ending in divorce Britain now has the second highest number of divorces in Europe (Jowell and Airey, 1994). There is also an increase in those who marry late or never marry. The number of British women under thirty years of age who have never married, rose by 12 per cent in the ten years between 1981 and 1991 (Faludi, 1992). Although this trend is reported as particularly evident for women earning higher incomes, in the present study this finding was reversed with the group with the lower modal income, the CAH group, having the lower frequency of marriage.

The CAH and diabetic groups differed in the proportions of those who saw the establishment of a stable relationship as a positive goal. With the exception of one woman who was divorced, all of those with diabetes were either already married or wished to be in a stable relationship. Four (21 per cent) of the women with CAH did not wish to establish a relationship of that kind, and only five of the remaining fifteen women were married at the time of the interview. These proportions suggest that the reported difficulties in both same- and opposite-sexed friendship formations, together with the anxieties about sexual performance, may have affected attitudes towards, and the establishment of, stable relationships. Frequently the women with CAH, but not those with diabetes, spoke of their lack of confidence about the survival of such partnerships:

_I'd live with a boy friend, but not get married. You can't guarantee that you are going to be with that bloke, madly in love, ten years from now._(C01)
I always have the thought - are we going to be together all our lives, or finish in the divorce court five years from now? (C03)

I wouldn't describe myself as 'living with him'. I have a home, I can opt out any time I want. (C52)

The element of the requirements of a good marriage

Of course, it is possible that the two groups held different criteria for, or had different needs of a relationship that they designated 'stable'. Two requirements of a good marriage were identified in the interview data: open communication and the maintenance of one's own individuality and interests. Although both groups acknowledged these requirements, the overall frequency with which they were mentioned, and the clarity with which they were expressed, were considerably greater in the group with diabetes.

A frequently cited reason for divorce is a failure to communicate (Davis and Murch, 1988), and both groups, believed good communication was a necessary and important strategy for solving problems that arose within stable relationships:

.....getting things out into the open, then they don't build up. It may upset the other person but it takes the burden off you. (D33)

It [my marriage] is quite good. We talk, nothing is sort of - everything is sort of out in the open, no secrets, nothing. (C47)

You have to talk about things, there's no way round it - getting problems out into the open. (D29)
An appreciation of the importance of open communication for a stable relationship may have provided yet another source of anxiety for those with a 'Skeleton-in-the-cupboard'. Those in the CAH group who were negative about their own wish to establish a stable relationship were also those who believed that 'open communication' was a central and important component of such relationships.

The second identified requirement for a good marriage, the maintenance of the woman's own individuality and interests was similar to the intentions reported by the newlywed women in the study by Mansfield and Collard (1988). In the present study, the women with diabetes hoped for an empathic partner who would be active in a close exchange of intimacy, but who would also make them feel valued as an independent individual with their own interests and needs:

...but to have your own life and not feel you have to explain everything. (D01)

...an acceptance of the other person, of their individuality. (D24)

This emphasis on individuality by those with diabetes was accompanied by statements that suggested this requirement was allied with that of the partner as a 'friend', someone with whom there was mutual trust and empathy:

You don't have to worry about them, work as a team, the other person laughs and cries with you. (D09)

The requirements for individuality and friendship in a good marriage were not identified in the data of those with CAH.
In summary, the group of women with CAH seriously questioned both the desirability and the stability of stable relationships. This group did not express a clear concept of 'a good marriage', with few women able to identify important components. Open communication was recognised as important by about one third of those with CAH, mainly those who did not wish to marry. In contrast, the group with diabetes was fairly cohesive in their views that a successful marriage was mainly dependent upon open communication, especially in relation to problem solving within marriage. The women with diabetes, while believing it was important that a partner was a friend also wished to maintain separate interests and their individuality within the relationship. The lack of clarity of those with CAH for the requirements for a successful stable relationship may be partly due to the smaller proportion of married women in the CAH group compared to the group with diabetes. The experience of living in a stable relationship would be likely to result in more clearly defined concepts. The effects of the 'Skeleton-in-the-cupboard' on attitudes towards marriage were likely to have operated through the more general effects on relationships. The lack of experience in close relationships of those with CAH, and the perception of the general advisability of open communication may have added to anxieties about intimacy.

*The element of housewife or career choice*

A low level of confidence in the stability of relationships together with anxiety related to intimacy may result in a high value being placed on a career. The role of women in the workplace and the value given by them to their roles out of the home has changed over the last century. By the first half of the twentieth
century, the housewife had become a symbol of femininity and social success. The husband was designated as the breadwinner, and it was the accepted view, particularly by the growing middle-class, that a married woman having to work outside the home was a misfortune and a disgrace (Oakley, 1974). However, the last decades of the century have seen attitudes changing and by 1992, The National Council of Women of Great Britain reported that 79 per cent of women between the ages of sixteen and thirty five saw 'getting on at work', or finding employment as a major goal. Only 50 per cent of women in the sample were concerned with having any, or more, children; diversity of roles had become important for the contemporary woman. Mansfield and Collard (1988) reported that even women who anticipated career breaks to accommodate pregnancy could not imagine permanently leaving the work force. However while 'the career woman' is a currently acceptable social role for many women who have higher levels of education and training, for those who are in low paid mundane work, staying at home after marriage, particularly if there are children, may be a culturally acceptable decision. In the present study, the salaries of those with CAH were lower than the salaries for women with diabetes. The modal salaries for the two groups were between £10,000-£14,999 and £15,000-£19,999 respectively.

However, both groups conformed to the current general attitudes towards employment discussed above. Over two-thirds of the women in both groups stated firmly that they wished to continue working after marriage and pregnancy:
-both, I don't want to be a mother- staying home with the children all the time, sort of person.(C01)

I would like both- I would like a nanny to look after the children and do all the nasty bits.(C08)

In the long term, I don't think I could stand to be a housewife all the time.(D09)

My career is very important. I wouldn't give it up, but I would have to find some compromise. I would fight hard [to stay working]. (D23)

In the end I'm certainly not going to exclude marriage cos of my career, but I can't imagine not having a career.(D34)

Although there appeared to be similar levels of commitment to a career in the groups, there were some differences that may be related to the effects of the 'Skeleton-in-the-cupboard'. The group differences in attitudes towards learning and academic progress at school that were discussed earlier were paralleled by differences in vocational training (see Table 17).

The women with diabetes stayed in full time education longer than those with CAH; the mean ages at leaving was 19.2 years (s.d. 2.3) for those with diabetes and 16.7 years (s.d.1.7) for those with CAH (t=3.8 d.f. 34. p< 0.001 2-tailed).

**TABLE 17: Post-school training for both groups**

<table>
<thead>
<tr>
<th>GROUP</th>
<th>DEGREE N. (%)</th>
<th>NON-DEGREE N. (%)</th>
<th>NONE N. (%)</th>
<th>TOTAL N. (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>CAH</td>
<td>2(11)</td>
<td>5(26)</td>
<td>12(63)</td>
<td>19(100)</td>
</tr>
<tr>
<td>DIABETIC</td>
<td>8(47)</td>
<td>4(24)</td>
<td>5(29)</td>
<td>17(100)</td>
</tr>
<tr>
<td>TOTAL</td>
<td>10(28)</td>
<td>9(25)</td>
<td>17(47)</td>
<td>36(100)</td>
</tr>
</tbody>
</table>

Chi square= 6.5, d.f. =2. p<0.05
Table 16 suggests that the difference between the groups in post-school training is mainly due to a reversal at the extremes. The proportion of those in the diabetic group who followed degree courses is four times as great as in the CAH group. In contrast over twice as many women with CAH as women with diabetes had no training. Only two women with CAH were satisfied with their career progress and the most frequently expressed reason for dissatisfaction was the effects of their poor academic record on the employment opportunities open to them:

- *I'm not trained for anything.* (C33)
- *I find it hard to get a job- if I could do maths I could work in a shop. I get in so much of a hassle I can't cope with it.* (C04)
- *Lack of schooling- I can't read- I thought about going to night school, but when I come home I'm too tired.* (C10)
- *I never stayed at school long enough.* (C17)

Levels of vocational training and types of employment may, of course, be related to the needs of the individual in terms of what they require from their employment. The data suggested that the women with diabetes saw a 'good job' as one that was challenging, allowed them to use their initiative and one where they would meet friendly people. In contrast, opportunities for using initiative were not reported as important for those with CAH: they were looking for employment with colleagues with whom they felt accepted, and the pay was good:

- *[I was] working with my brother. I knew them all, it was like working with the family.* (C10)
- *They were so friendly. I liked that and it was well paid.* (C03)
It would seem that the two groups required different components in their employment. The diabetic group looked for opportunities that contained some element of risk through demands made on them to use their initiative, whereas those with CAH required social acceptance and an underpinning of safety. These requirements by the CAH group appeared to be associated with a lack of active career planning; less than one third had made any future plans. This passive approach of those with CAH was evident across all levels of educational success; as one woman, a medical student expressed it:

_I haven't a clue! I was talking about this last night with a friend- I just don't know._(C07)

The women with diabetes were more active in planning their employment futures, again across all levels of educational progress:

_I thought I would like to get a job in London at a teaching hospital, so that's what I did. I thoroughly enjoyed it, the best job I've had so far._(D25)

_I'm yet to go higher, hopefully. I should hear in the next three weeks, to Supervisor, I've moved up._(D28)

In summary, the findings relating to attitudes towards careers suggest that the groups were similar in their wish to continue working after marriage and parenthood. However, those with CAH were dissatisfied with their career development and clearly attributed this to their poor academic progress at school. The unsatisfactory career progress was also seen as determined by their low levels of vocational training. Differences were also expressed in the components of employment required by the two groups of women; those with diabetes required opportunities that contained some openings for initiative, whereas those with CAH were looking for the security of social acceptance and
good pay. The final difference was in the area of career planning where, across all career levels those with diabetes appear to be active in formulating and following plans whereas those with CAH had no plans to follow.

Once again the data supported the proposal that the 'Skeleton-in-the-cupboard' had effects on attitudes towards marriage, and approaches to careers. A stable, committed relationship may bring with it not only the necessity for making decisions about continuing in employment, but also those about 'starting a family'. The final dimension of the effects of the 'Skeleton-in-the-cupboard' on sexual partnership experiences, that of attitudes towards parenthood (see Table 13) will now be discussed.

The dimension of the effects of the 'Skeleton-in-the-cupboard' on attitudes towards parenthood

The analysis of the data identified three elements that contributed to the dimension of the effects of the 'Skeleton-in-the-cupboard' on attitudes towards parenthood. These are listed in Table 13 and were the value of parenthood, knowledge of condition effects on pregnancy and knowledge of genetic issues.

The element of the value of parenthood

Although feminism has long emphasised the need for women to have a real choice as to whether or not to become mothers, there is still a prevailing emphasis on the inevitability of motherhood (Baber and Monaghan, 1988; Morell, 1994). Pregnancy and motherhood remain a central social experience for women, one that perhaps confers status on young women, especially those who have little prospect of obtaining fulfilling employment (Campbell 1984).
However it is also reported that motherhood is not a positive experience for all women (Ussher, 1989; Phoenix et al, 1991; Oakley, 1986; Nicholson, 1988). For women with CAH, the 'motherhood' cultural stereotype may be difficult to emulate. The focus of the chronic condition, as has been suggested earlier in this thesis, is on the reproductive systems and this together with the likelihood of genetic risks, might increase the chance of the development of negative attitudes towards pregnancy and parenthood. Equally the experience of living with the chronic condition of diabetes, with the availability of information about the disruptive effects of pregnancy on blood glucose levels, the risk factors for the neonate, and the known genetic components, may increase the probability of the development of similar negative attitudes towards motherhood. It could be expected therefore, that both groups of women might question the advisability of parenthood for themselves.

A number of the women in the study were very clear that they did not wish to have children. Of those with CAH, six (32 per cent) expressed this view, with one of these having elected to be sterilised, three (18 per cent) of those with diabetes did not want to have children. The remainder of each group were positive about parenthood although the timing of this event in their lives was unclear. This included three of those with CAH who had been informed by their doctors that they would not be able to become pregnant, and one woman who had undergone a hysterectomy when she was sixteen as treatment for problematic menstruation, an event which for her was very distressing. The literature suggests that there are fertility and delivery difficulties for those with CAH (Mulaikal et al 1987), and although the proportion of pregnant women
with diabetes coming to full term and producing healthy babies is increasing, considerable medical support and self-care is necessary for this outcome. In all, three of the CAH group women had become mothers at the time of interview, one having had three successful unplanned pregnancies and the other women each having two children. A further two of those with CAH were receiving fertility treatment; and one mother had received treatment in the past. Of the fourteen women in the diabetic group who held positive attitudes towards parenthood, two had one child each.

Attitudes towards parenthood were further investigated by asking the women why they wished, or did not wish, to become parents, and what differences becoming a parent would make to their lives. The analysis of their answers produced four positive and three negative values about parenthood (see Table 18):

<table>
<thead>
<tr>
<th>POSITIVE VALUES</th>
<th>CAH</th>
<th>DIABETIC</th>
</tr>
</thead>
<tbody>
<tr>
<td>Joint creation</td>
<td>22</td>
<td>19</td>
</tr>
<tr>
<td>Company/attachment</td>
<td>33</td>
<td>19</td>
</tr>
<tr>
<td>Completion of marriage</td>
<td>22</td>
<td>18</td>
</tr>
<tr>
<td>Helping child's development</td>
<td>22</td>
<td>31</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>NEGATIVE VALUES</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Too great a sacrifice</td>
<td>13</td>
<td>33</td>
</tr>
<tr>
<td>Medical/physical difficulties</td>
<td>31</td>
<td>11</td>
</tr>
</tbody>
</table>

As can be seen from Table 17, the percentages of those expressing the value of the parenthood as being a 'joint creation' and as the 'completion of the marriage', were similar for both groups:
The remaining two positive values appeared to be of different importance for the two groups: the CAH group most frequently expressed the view that a child would be company and provide emotional ties:

- It would be someone to love, someone extra to include in our lives-I get lonely. (C47)
- It would be nice to have someone always there really. (C04)
- I would picture my life as being pretty empty without children. (C47)

In contrast, for those with diabetes the most frequently expressed positive value was that of taking part in the child’s development:

- It would be nice....to have something we'd created, we can watch it grow up and help it grow up. (D23)
- It's the future happening now, you know, it's like an extension of my future, seeing your child grow up and what happens to it. (D01)

The negative elements of parenthood also showed group differences. For those with CAH, the emphasis was on the anticipated physical and/or medical difficulties; these will be discussed under the element of problems during pregnancy below. Those women with diabetes were more concerned with the anticipated sacrifices that would have to be made:
At the moment I'm quite a selfish person and I just don't feel as though I want to give up everything. (D18)

I'm the type of person who, if they want, can get in the car and drive off. I can go abroad, I can go on holiday- I can afford to. It would mean you can't do that- there's somebody else there that needs you and you've got to think about. (D28)

I couldn't cope with it- I think I could see it getting in the way. (D25)

The elements in this study that have been identified as related to becoming a parent, are very similar to those found nearly twenty years ago in the study by Marcus (1979) on eighty pregnant women aged 18-37 years. The use of factorial correspondence analysis produced five distinct configurations of themes, three positive, one negative and one mixed. Of these, the author suggested that for the women in his study, becoming a parent was composed of the following factors; it's normal for a woman (to have a child); it's an extension of the couple; to shape someone, to shape a life; and to create emotional links. The similarities between the two studies suggest that the presence of CAH or of diabetes has not made changes to the value and reasons given for desiring parenthood. However for the women with CAH there were considerable anticipated medical and physiological problems; while being aware of the necessary increased demands for blood glucose level control during pregnancy, unexpectedly fewer of those with diabetes were concerned about this issue. It may be that their greater level of awareness and easier access to information resulted in an assumption that any difficulties were manageable and would be supervised by their medical advisers. This assumption reduced uncertainty and speculation.
The element of knowledge of condition effects on pregnancy

Pregnancy for those with CAH was perceived as somewhat of a problem, and it might be anticipated that the issue would have been raised during discussion with their medical advisers: this, however, did not appear to have been achieved. In line with the findings reported above in the section on doctor-patient communication, less than a quarter of the CAH group had discussed with or received information from their doctors about any aspect of pregnancy. In general such communication that had occurred was reported as poor:

No, they haven't said anything to me. Do people with CAH have children? Are there people out there today with CAH who have got children?(C01)

The doctor has said that I can have children. It was way at the back of my mind. It was one of those doubts that you don't think about, but it's there. The doctor just came out with it for no reason- I'd never asked, there was no reason [for him] to bring it up, to say that, and I thought, 'Well, there you go', and cast it aside.(C09)

The only thing I remember is that they said it would probably be difficult. What that implies- if it is a physical or a fertile thing- I've no idea.(C52)

For just under one half of the group with diabetes, there had been some discussion with their doctors, and it was again very common for information to have been obtained from the BDA. and the media:

No, it hasn't come into question yet. The Balance Magazine had an article about being pregnant and the sort of checks and control you have to have. I feel fairly well informed, but I'd like to know more, but nearer the time.(D01)

My GP said, 'When are you going to have your children, you are getting a bit old'. Actually trying to find out about it was quite difficult......I remember reading that a diabetic's innards atrophied quicker, and so you should have children early.(D24)
As with all groups of women there was concern about becoming pregnant and/or the experience of delivery. The women with diabetes appeared to have realistically founded beliefs that pregnancy would necessitate frequent monitoring and were aware that a strong emphasis would be placed on good blood glucose control:

*I've never discussed getting pregnant with any of my doctors. My Mum's told me she doesn't want me to have any. For a year or six months before I would need really good control, and I'd really have to want the child. And then towards the end, doctors appointments every two weeks - I know that from what I've read.* (D23)

Those with CAH held one of two positions: they were either unaware that there might be problems, or were concerned that conception and/or delivery would be problematic. Neither of these positions appeared to be based on accurate knowledge nor an understanding of the effects of their condition on reproduction:

*Personally, I think there would be [difficulties], cos I've got such a small frame. I think it would have to be a caesarean, but that effects a lot of people, doesn't it? I would hazard a guess that it might be a bit harder to conceive.* (C13)

*They said they were worried to start with, that I might never have a period. When I started, they were chuffed-they knew I was gonna be able to have children. I'm going to be able to have children, bear like a normal person. That's what they said to me. I'm going to have no problems.* (C37)

*No, none at all [discussion]. When I was at [paediatric hospital] I had to have an ultra-sound, but they didn't say anything. When Dr.X. saw the results and saw my ovaries, he said, 'well, there they are- glad to see they exist'. I thought 'why shouldn't they?' Nobody had told me the association with hormones. It never crossed my mind, I never thought that they might not be there. I don't know why even now. Is there a link between CAH and fertility? I'd like to know more.* (C07)
The element of knowledge of genetic issues

Although there is a known genetic factor in the incidence of both diabetes and CAH, it is likely that for those with CAH the low level of understanding of their condition included a lack of information about this issue; only one woman with CAH appeared to have any understanding. It was more surprising that the women with diabetes also appeared to have a low level, with only one woman having any clear understanding of the inherited aspects of diabetes. More of the women in this group had limited information:

I think it's amazing that my parents happened to meet and they both carried the same gene and they had a one in four chance of having a child with it. We went into it when we were planning to have the children, the chances of us having a child with it, and I understand that both the girls carry the gene, but that everything will go alright so long as they don't meet someone with the same thing. Fortunately, neither of the children have it so I presume that my husband isn't a carrier. But we haven't stopped it in its tracks, so we'll always wonder what's going to happen with them. (C34)

I mean I'd like to know what the chances are of - I'd like to know what sort of percentage possibility there would be of my child being diabetic. (D01)

More commonly however, the women in both groups had either no concept of the issue:

I don't know what it is. (C01)

What? (C03)

or an inaccurate, superficial, and confused understanding:

...cos it's only hereditary in girls... Well you see. I'd had quite a few miscarriages... and when I lost that I said to C 'they're girls, they've got to be girls - they've got to be, 'cos God's saying to me 'S, you can't carry girls, there's got to be something wrong with them, I'll get rid of it.' ..... When I had D... I got to three months I
still had no bleeding and C said to me 'That is a boy.'
(D08)

Some women with CAH had recently become aware of the issue in distressing circumstances:

I've always been under the impression that I got the condition through a drug that my Mum took- she was loosing me at five weeks, and they injected her with something or other and she had that for the rest of her pregnancy. She was told that the condition was caused by the drug. Now I'm having fertility problems, it's come out at the clinic that it's hereditary. One doctor said to us 'Do you realise that if you have a child, they could end up with the same condition?' Well, I was flabbergasted!(C28)

Yes, it's a worry. They did investigate her [third child], but apparently she was all right, but I kept asking questions- 'why', 'cos I never knew that it could be passed on from me to my children. They got a bit stroppy and said, 'If you don't bring her in for tests, she could die.'(C47)

The low numbers of women in both groups who had any understanding of the genetic component of their condition makes it difficult to identify the effects such knowledge might have on motivation and commitment to parenthood. Two women in each group (11 percent of the CAH group and 12 percent of those with diabetes) felt that if there was a possibility of the condition being passed on, they would not wish to become pregnant:

I would say [if partner was a carrier] not to have children. I know it's not life threatening, but I wouldn't want to bring something into the world that was sick if it could be avoided...you are thinking of a person who has to be on drugs all its life.(C13)

I just had these worries about the babies on my own. I noticed that the second one has a very small penis, I've taken him to the doctor about that cos I got a bit neurotic about it. The doctor reckons he's fine-it's just that he's fat. I was shitting it, thinking they might have got what
I've got, which I would never have forgiven myself for, if they'd got it. (C41)

But for the majority, consideration of possible hereditary aspects of their condition did not result in a decision to remain childless:

I don't think I'd blame myself - it's just life, even though I know it would be my side of the family that it got it from. (D23)

We decided, as it wasn't exactly a life-threatening illness, it was something you could control to a certain degree, that we would go ahead. (C34)

The chances of meeting a guy with the same genes as what Mum and Dad have got are very very rare. No one's mentioned it, my plan would be to go ahead and have children. (C37)

In summary, with reference to views about parenthood the majority of the women in both groups were similar in their attitudes to those women in the general population. Parenthood was still their accepted expectation, and the reasons given by them for desiring pregnancy contained similar elements to those of women without chronic conditions described in the literature. The theme that has emerged throughout the present study of relationship difficulties due to the presence of a 'Skeleton-in-the-cupboard' for those with CAH, also showed its effect in this area by the strong emphasis on the child as a provider of company and as an emotional tie. The belief that there would be physical and/or medical difficulties which would effect conception, pregnancy and delivery appeared to have been formulated by those with CAH without any substantial information or understanding.
The proposal that the existence of the 'Skeleton-in-the-cupboard' would exert effects across a wide range of 'sexual partnership experiences' (see Table 13) has been supported by the interview data. The information given about attitudes towards partnership formation suggested that both groups of women were positive about the desirability of such relationships, although for those with CAH there was a considerable doubt about the stability of present day heterosexual relationships. For both groups a good partnership depended upon open communication, and for those with diabetes on the maintenance of their independence and individuality. In addition women with diabetes wished their partner to be a 'friend' as well as a sexual partner. Those with CAH, perhaps because of the lower percentage of women in the group who were in stable relationships, were less clear about the required essential elements contributing to the concept of a 'good marriage'.

In relation to the gender role issue of choice between marriage, becoming a full time housewife or retaining a working career the data supported the view that both groups of women mainly desired both marriage and a career. This was so even though women with CAH were generally dissatisfied with their career progress which they attributed to their poor academic achievement at school. They did not take an active role towards their career development plans, and within their employment were looking for social acceptance by colleagues, and good pay. In contrast, for those with diabetes working life was more planned and they required employment where they were challenged and could use their initiative.
The majority of both groups was positive in their attitude towards having children, and this finding was in line with present day attitudes in wishing to continue working after marriage and parenthood. Those with CAH anticipated some difficulties in reproduction that they attributed to their condition; however this view was not based on reliable information. In contrast, but in line with other areas reported in this thesis, those with diabetes were better informed having obtained some information from their doctors and the British Diabetic Association. Women with CAH believed children would provide companionship and an emotional focus to their lives, whereas those with diabetes looked forward to the interest created by their contribution to, and observation of their child's development. Few members of either group had discussed or been counselled about the genetic implications of their condition, with those who had been frequently receiving information as problems occurred in relation to their own reproduction experiences. For those women with CAH, this had often been traumatic. The absence of understanding by the women makes it difficult to assess the impact of the genetic component on their wish for children; the findings of this study do not allow a prediction about the direction of the effects of such understanding on attitudes to parenthood.

THE CATEGORY OF THE CONTRIBUTION OF CULTURAL DISCOURSES TO THE MEANINGS OF EXPERIENCE

The findings of this study have involved information about broad areas of the women's lives and the meanings they have attributed to their experiences. The model outlined proposed that the presence of a 'Skeleton-in-the-cupboard'
would represent a central focus for the experiences of living with the chronic condition of congenital adrenal hyperplasia and this has been supported. In contrast, living with diabetes has been characterised as the absence of such a skeleton. Meanings which individuals impose on their experiences may be construed as a product both of that experience, and the collection of socially constructed 'truths' which are part of every member of that society's repertoire of beliefs, actions and expectations. Such 'truths', the ways in which the members of the society verbalise and give meaning to their experiences, have been described as 'discourses'. A discourse is a complex concept described by Prior (1989) as '....not merely a narrow set of linguistic practices which report on the world, but .. composed of a whole assemblage of activities, events, objects, settings and epistemological precepts.' (p.3). More briefly Ransom (1993) described discourses as structured ways of knowing. Discourses therefore operate across many social structures, situations and institutions, and may act as 'filters' or, to use Bem's 1993 terminology, lenses through which meaning is construed from experience. In his analysis of discourses, Foucault (1981) proposed that discourses actually create the experience to which they refer. For example, discourses of femininity both constitute what it means to be a woman and in so doing also influence the constructions of experiences though the beliefs inherent in the discourse. It follows from this model that as social discourses are developed or changed, the meaning of experiences will be re-construed, and through these changes the view of the self will be re-created.

A society establishes and maintains its discourses by the tacit communication of meta-messages about what is considered to be important and what is to be held
in high esteem for that particular culture. Meta-messages may be transmitted from various sources: for example from educational practices, by the media and advertising, or more directly by direct communication from other individuals. Discourse acquisition then, is perceived as being a multi-faceted activity and is achieved by an active transaction between the individual and the total social and cultural environment.

Any investigation of the meanings of experiences for those with a chronic medical condition needs therefore take into account some of the established cultural discourses that may be pertinent to the experiences of the condition. Table 19 outlines the main and component discourses believed to have relevance to the experiences of those women with CAH.

**TABLE 19: Outline of discourses relating to CAH**

<table>
<thead>
<tr>
<th>Discourses</th>
<th>Discourse components</th>
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<tbody>
<tr>
<td>1. 'The opposite sex'</td>
<td>'Programmed-in'</td>
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<td></td>
<td>'Happiness is having some-body'</td>
</tr>
<tr>
<td>2. 'Under doctor's orders'</td>
<td>'The doctor knows best'</td>
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<td></td>
<td>'Good health is good for you'</td>
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**The ‘opposite sex’ discourse**

It is now usual in academic writing to distinguish between the terms 'sex' and 'gender'. Sex is used to refer to the biological differences in chromosomes, genitalia and reproductive function by which humans are assigned to the male or female group. Gender refers to other behavioural and physical differences in
characteristics such as social roles, dress, hairstyles, ways of walking and sitting. This separation between sex and gender can also be accompanied by the belief that gender, the experience of the self and others as men or women, is socially constructed rather than being based on biological differences. However this separation appears to re-establish an unsatisfactory split between a social and psychological state (gender) and the body (sex). This seems unsatisfactory because it is reasonable that the experience of living in a male or female body is an integral part of the construction of the self as a man or as a woman. For those women with CAH, where there has been some ambiguity at birth about the sex of their body, and perhaps a difficulty for the parents to present unambiguous gender expectations, the discourse of the 'opposite sex', with its clear implication of gender differences, may have strong effects. When the society in which one lives is comprehensively organised around a dichotomy of the sexes into males and females, those whose self-perception includes a belief that they may not belong in either class,

*I was born a boy, but inside you are a girl. Had an operation that made me into the thing I should have been.*

are very likely to feel effects in many areas of their lives.

The dichotomization of the sexes into male versus female not only serves as the basis for the organisation of social life with, for example, gender designated games and hobbies, but it also mis-divides the continuum of many biological processes. For example the hormonal system, where men and women produce both androgens and oestrogen, has been dichotomised into 'male' and 'female'
hormones. The polarisation of gender, as Bem (1993) suggested operates in two ways: firstly, it defines gender scripts which are mutually exclusive, and secondly, it implies that any deviation from these scripts will be held by society to be pathological. For those where behaviour has been designated as deviant, where it is believed that the behaviour does not conform to the social definition of the gender, then it follows that the label ‘pathological’ will be applied to that behaviour. Such labels have far reaching effects especially in the responses of others and the perception of self by those so labelled (Goffman, 1963).

The idea that males and females are 'opposites' exists in virtually all societies (Ortner, 1974). Bem (1993) has discussed the extent of this 'polarization of gender' through a description of a hypothetical society in which it has been eliminated. She suggests that the sexes would no longer be culturally defined in terms of such differences as dress codes, social roles, power, cognitive and manual skills, emotional expression or sexuality. In addition females would not be defined against the masculine standard as more submissive, less independent, less excitable, or more adventurous (author’s emphasis). This would imply that '...being a boy means having a penis and testicles; being a girl means having a vagina, a clitoris and a uterus; and whether you're a boy or a girl, a man or a woman, doesn't need to matter unless and until you want to make a baby.' (p.149).

Until that hypothetical state exists, the existence of the precept that men and women are of 'opposite sex' has a number of consequences. These include both the expectation of, and differential response to, other's behaviour; thus tasks are
still allocated on the basis of sex. Although there is now less gender polarisation in employment (Institute of Employment Research, 1993), there is evidence that the 1970s equal opportunities legislation in Britain has not resulted in significant changes in women's employment positions. Feminine work is still largely defined as domestic and/or the practical and emotional servicing of others (Oakley, 1974; Ussher, 1989). Cassell and Walsh (1993) for example have shown that females still occupy work positions of 'servers to others', and men, those of rationality and instrumentality.

For women with CAH, the cultural understanding that women's role in employment is a caring one, and the basis of their power is a relational one (Gilligan, 1982) may provide a source of threat given their general difficulties in this area. It is possible that this aspect of the widely held 'opposite sex' discourse has, in this way contributed to their low career interest and achievement. Although such a threat might have led the women with CAH to enter employment traditionally regarded as 'male' and accept the risk of being labelled 'deviant', it is perhaps more likely that the strong insistence of their parents and themselves that they were 'normal' would tend to make it more essential for them to seek a 'female' work role. The ensuing anxieties and relationship difficulties in this type of employment might then be seen as resulting in a position of lack of interest and withdrawal. The diabetic group, less likely to be labelled deviant and therefore not have these anxieties, would be more likely to be active, interested and achieving in their careers.
Programmed-in

The 'opposite sex' discourse can be seen to contain within it two related ones; that gender differences are held to be fixed, immutable and 'programmed-in' being commonly attributed to prenatal environmental events, and 'happiness is having some-body'.

The first of these, that gender differences are 'programmed-in' has 'borrowed' components from scientific theories. There is, of course, support for some prenatal effects; one - the known effect of pre-natal hormones on the female external genitalia - is a theme of the present study. However, support for other theories about the effects of hormones on the developing brain (LeVay 1993: Hampson and Kimura, 1992) have been, at best, ambiguous (Vines 1993). The important issue here, is that these and similar ideas have become part of the cultural language, may have acquired important roles and become filters through which the meaning of experience is constructed.

One borrowed component belief, that 'women are ruled by their hormones' has come to be used to explain a variety of women's experiences (Ussher 1989). Experiences such as those of aggression, stress, eating disorders, shoplifting, agoraphobia and lesbianism have all been attributed to hormonal factors. If it is believed that behaviour is attributed to hormonal factors, then it may appear logical to believe the converse- that abnormal hormones, as in CAH, would result in behaviours that are commonly construed as outside the limits of stereotyped female behaviours. Research into the relationship between behaviour, hormones and CAH began in the 1960s and continues to the present
day; the results have been claimed in support of the belief that the direction of causality is from hormones to behaviour. However data which supports a co-relationship, does not directly define a cause, and it is suggested here that psychological, cognitive and social effects are as likely as hormone abnormalities to result in behaviour that is classified as gender atypical. Examples of research that has supported hormones as the cause of atypical behaviour in those with CAH are Hines and Kaufman's (1994) investigation of aggressive behaviour in young children with CAH, and Erdhart et al's (1968) findings suggesting increased lesbian tendencies in women with the same condition. The discourse of 'programmed-in' behaviour, together with the belief that women are ruled by their hormones, may have increased the reliance on conclusions of hormonal causes for behaviours believed to be sex-typed. The alternative view, which looks at the cognitive construction of individuals and the cultural connections of their experiences, provides a more complex model.

In spite of the entry of the concept of 'androgeny' into social language, it would appear that the polarisation of maleness and femaleness, of men and women, still appears entrenched. The two alternative positions, biological reductionism and social constructionism (Gergen, 1985), outlined above and proposed as the underlying basis of this dichotomy, imply different focuses of management for those with chronic conditions. Biological reductionism, with its focus on 'abnormal behaviour caused by hormone imbalance' perpetuates a reliance on medical intervention; the focus of social constructionism on meanings and experiential understanding necessitates a more psychological and cognitive approach. These alternative approaches will be examined in greater depth in a
later section after discussion of the implication of a second discourse related to that of the 'opposite sex' (see Table 19). This related discourse arises from the cultural construction and emphasis on the ideal female body.

**Happiness is having some-body**

Ussher (1989) has proposed that 'An important theme running throughout much of the discourse concerning the adolescent girl is that of the ideal archetypal woman having a perfect body' (p.38). As a highly desired goal, this is believed to become an important issue from early adolescence onwards. This cultural discourse is of course reinforced by the media and fashion industry, and has resulted in common female concerns about body size, weight, shape, and style. The attempts of some feminists to alter the construction of the ideal body away from the 'tall and slim' does not appear to have been successful. For those women with CAH, the associated weight increase at adolescence is likely to increase dissatisfaction with their body image, as it fails to conform to society's current stereotype of feminine beauty. This dissatisfaction may be enhanced as CAH increases the likelihood of short stature and a heavy physique, and can be accompanied by hirsuteness. Under these conditions, the body-appearance of the CAH woman may be more likely to be a source of discontent rather than one of happiness. For those with diabetes, the attainment of the ideal body image is also affected by the condition. The necessary balance of insulin and available blood glucose obtained by regular and frequent meals make it very difficult to lose weight, especially during adolescence when an increase in weight is commonly experienced.
Another aspect of the maturing body, the beginning of menstruation, is considered to be an important status event that indicates the achievement of womanhood. For those with CAH this developmental event, or its disruption, is frequently a focus of medical concern and becomes an added negative body issue in the achievement of the cultural idealised woman, (see also Malson and Ussher, 1996). For those who have some existing concerns about their membership of 'womanhood', concerns about body image, menstruation and genital adequacy may culminate in a strong self-perception as being 'different', as being a 'freak'. This word was used by more than one woman with CAH and others used synonymous descriptions.

'Happiness is having some-body' also refers to the discourse of female fulfilment through relationships, particularly with those with the opposite sex. Issues about close relationships have been a central theme in this study, particularly those relating to the acceptability to the opposite sex of the CAH women's bodies. It has been suggested that for these women, the presence of a 'Skeleton-in-the-cupboard' had resulted in considerable difficulties in establishing close heterosexual relationships. Similar difficulties were found in same sexed relationships where friendship demands of intimacy were a source of anxiety. The cultural discourse which places relationships, particularly heterosexual relationships, at the centre of women's sense of achievement, identity and satisfaction (Gilligan and Murphy 1979. Gilligan 1982) is likely to be an additional source of stress for women with CAH.
In conclusion, the cultural discourse of 'the opposite sex' can be seen to contain a firm stereotyped representation of 'woman not man', and the differences between the two polarised genders are commonly believed to be fixed, 'programmed-in' by either genetic or hormonal pathways. The perceived need for women to conform to the culturally constructed ideal feminine body may lead to increases in dissatisfaction with their self-image. This outcome may be more likely to occur for those whose chronic condition involves physical characteristics that depart from the feminine stereotype. Aspects of these discourses that relate to immutability are likely to result in a belief that change is not within the control of the individual. Examples from the interview data of this lack of controllability by those with CAH have been presented in various sections of the discussion. In particular, group differences were clear in the statements relating to the medical management of the two conditions. In relation to the part played by them in the management of the condition, those with CAH did not take an active role and did not believe things would change, whereas adult women with diabetes played an active role and anticipated considerable future management improvements. Similar positions were apparent in relation to the sexual difficulties of those with CAH, where, in contrast to those with diabetes, there was an absence of strategies to remedy vaginal dryness and penetration difficulties. The data on sexual experience also provided support for the concept of the body as the perceived basis of happiness. The belief extensively expressed in the interviews that happiness could be achieved when and if the genitals of those with CAH were found to be acceptable by their partners, found to be 'normal', exemplifies this position. The final strand in this section was the effect of the discourse relating to the
importance of relationships for women's self image and satisfaction. Again, this discourse may create particular difficulties for those who believe they have something about their bodies that has to be kept secret.

Although the meaning of experiences for both groups of women would be open to the influence of such discourses, they may have greater pertinence where the chronic condition itself has a strong sexual and gender focus. This applies to the chronic condition of CAH and the significance of these discourses for clear, comprehensible communication is considerable. This issue will be taken up in the later section on the implications of the current research findings for the management of CAH.

The discourse of 'Under doctor's orders'
The second discourse which has implication for the women in the study is that entitled, 'Under doctor's orders' (see Table 19). In the life of those who have a chronic condition, a central experience is likely to be the regular hospital visits to consult the doctor. In contemporary western society, the medical profession, particularly hospital consultants, is perceived as scientists and experts who have high status and authority. Public language contains many exemplars of this; 'just what the doctor ordered' refers to an object or event which is highly desired and is likely to make things good; 'you're the doctor' suggests that the person to whom this comment is directed has ultimate responsibility for decisions. The reciprocal but unequal relationship between the doctor and those who consult them is suggested by the double meaning of the word 'patient'. This word according to Collins Dictionary (1986) refers to both a person who is receiving
medical care, and an individual who endures trying circumstances with an even temper! Two component discourses appear to be related to the experiences of the diabetic and CAH groups, these being 'The doctor knows best', and, 'Good health is good for you'.

'The doctor knows best'

A consultation with a medical expert presents a number of conflicts on both sides of the relationship, conflicts which arise out of the assumptions each side holds about the rights and duties of the other. Patients are expected to bring to the doctor 'real' rather than irrelevant or trivial symptoms; for the patient the decision as to which symptoms are of sufficient seriousness to be reported depends upon their subjective judgement. Once the consultation is operating however, the doctor becomes the decision-maker. It is thus the doctor who both defines the area of concern, which symptoms are worthy of investigation, and the strategies of management (Pitts, 1991). This focus of authority in the doctor has been held to be a factor in their perceived failure to take account of the patients' opinions, knowledge and experience about their condition, and in the patient's non-compliance with the doctor's orders.

It would seem that this traditional view of the doctor-patient interaction is now less appropriate, especially where the consultation occurs as a result of a chronic condition. It has been suggested that doctor-patient relationships are moving from a knowledgeable expert/ignorant lay patient model towards one conceptualised as a more equal interaction between experts (Tucket et al, 1985; Arney and Bergen, 1984). As a result, doctors may now be more likely to
encourage their patients to voice their opinions, and to take responsibility for their health by sharing in the decision process about treatment regimens (Hertzlich and Pierret, 1987). The data from the two groups of women which related to their current hospital clinic visits however, indicated a major difference in the position taken on the discourse of the 'doctor knows best'. The experiences of visits for those with diabetes appeared to have moved significantly towards the 'meeting of experts', whereas for those with CAH it remained at the 'expert/ignorant lay patient' position.

The social role of medical practitioners includes the belief that doctors have both the power and the obligation to use their expert knowledge to return their patients to good health. This discourse is not in line with experience for those with a chronic condition as by definition a cure is not possible. The goal of management for them is the adequate control of symptoms, the avoidance of complications, and the possibility of an acceptable life-style. For those with diabetes, the position of a 'meeting of experts' in relation to the doctor is parallel to and endorsed by the responsibility taken by them for the day to day management of their condition. For those with CAH, the responsibility for day to day management is limited to taking medication that appears only tangentially related to their physical condition. This, together with their unsatisfactory levels of understanding, the lack of information about test results and their health status in general, are in opposition to the cultural move towards shared responsibility for health status.
In summary, the experiences of the two groups in relation to the changing cultural discourse of 'the doctor knows best' appear to have been different. For those with diabetes, their responsibilities for daily management, and their knowledge about their condition, even though still at a low level, have frequently resulted in an equalising of roles with themselves as 'experts' and the doctor as an informed support source. For those with CAH, this change of role has not occurred. This outcome may partly be attributed to the women's failure to understand their doctors' communications and the absence of any alternative sources of information. Together these may have resulted in the low value placed on clinic visits and the women's reported failure to adopt an active participation in the management of their condition.

The recognition of patients as having 'expert' knowledge about their own experiences, is mirrored in a further cultural discourse component, that of 'good health is good for you'.

'Good health is good for you'

The move towards patients taking an active role in the management of their chronic condition is paralleled by the more general cultural discourse of the value of 'good health'. The social emphasis on health is a broad one and this is exemplified by the naming of medical services as the 'National Health Service', by 'Health and Fitness centres', and by 'Well Woman Clinics'. The media, be it television, radio, newspapers or magazines present the public with information on every aspect of a 'healthy life-style'. Dietary issues and their relationship to a healthy ideal weight and their implications for diseases, the effects and side-
effects of prescribed and non-prescribed drugs, smoking, exercise and stress are all regularly addressed. The public is repeatedly confronted with dictates and methods whereby they can achieve good health.

The main responsibility for this state-of-health commonly falls not on the medical profession, but on a network of lay personnel: the individual, the members of their family, and friends and colleagues. Stacey (1988) has suggested that almost any member of an individual’s network of acquaintances can be considered a health care worker. Visits to hospital outpatient clinics therefore comprise only a small part of the action taken for the achievement of good health. In this research, the women with diabetes made use of this broad area of health care; their parents, friends, self-help groups and the literature were all utilised as sources of information, guidance and support. However, two factors denied the CAH group access to such resources: the perception of the need for secrecy about their condition, and the virtual absence of any available support or information services both professional and lay. The combination of these two factors would act to increase the perception of the non-acceptability of their condition and thus reinforce the requirement to maintain their 'Skeleton-in-the-cupboard'.

For both groups, a second consequence of society's emphasis on good health was the conflict between the women’s perception of self as 'sick' or as healthy'. In western society the sick role is composed of a number of components. It acts to legitimatise the withdrawal from a number of obligations such as those relating to work and/or relationships. The sick individual is also exempt from
total responsibility for the medical condition; they are seen as requiring some medical help. Those who are sick however, have a social obligation to 'get better' (Parsons, 1975). These components of the discourse relating to good health provide a source of conflict about the self-image of those with chronic conditions. The data made clear that neither the women with diabetes nor those with CAH fulfilled the above criteria for being 'sick', except perhaps that of needing to consult a medical expert. Those with diabetes appear to have accommodated this need into their view of self, whereas those with CAH have not. For this latter group, difficulties in accepting the need for aspects of 'sickness' may have been balanced somewhat by the greater social acceptance of 'sickness' than of 'oddness'. Their preference for 'sickness' may act to maintain some value for their attendance at hospital clinics.

In chronic conditions, with the change of focus from intervention to surveillance, 'health' becomes more a question of the ability to engage in normal social activities than an absence of biological pathology. The implication is that the doctors as experts also face a dilemma and a conflict. Attempts by them to widen their areas of concern from a narrow medical one into one which is concerned with the broader social life of their patients, may be construed as intrusion into areas not traditionally accepted as the domain of the medical practitioner. It seemed however, that both those with CAH and those with diabetes felt that their consultants did not know them as individuals, had no idea about their lives and would have welcomed a broader interest by the doctors. These feelings may reflect a perceived need to discuss aspects of the effect of their condition on their lives. It is being increasingly recognised (Gabe et al,
1994) that a biomedical solution is inappropriate or insufficient for many of the social, psychological and behavioural problems associated with medical conditions. A change of focus in the management of illness could however, place the medical profession in the kind of dilemma described by Blaxter (1983):

\begin{quote}
To meddle with matters which are social, economic and political is medical imperialism. On the other hand, given that health systems are a major institution of advanced societies, it can be argued that this counsel of despair, is a weak refusal to accept responsibility, and an excuse for giving up the most intractable problems as the business of somebody else. (p.143)
\end{quote}

Failure to 'meddle' may also leave the patient with a feeling of dissatisfaction about the consultation. The prevailing discourse relating to health and sickness encourages the recognition that attention to factors other than those biomedical plays a considerable part in the optimum management of chronic conditions. For the women with CAH, many social relationship factors were not addressed during their outpatient clinic visits. These included their anxieties about sexual behaviour, especially the effects of their surgery on sexual intercourse and the acceptability of their condition by themselves, their family and friends. For these women, these important areas remained problematic without any obvious source of professional help.

In considering the implications of the medical expert's failure, or at least perceived failure, to consider social and behavioural factors as part of the long-term on-going management of chronic conditions, it is helpful to remember the point of view of Stainton-Rogers (1991). She pointed out that individuals do not construct realities, but are 'clever weavers of stories, whose supreme
competence is that they can and do create order out of chaos, and moment-to-moment make sense out of their world amid the cacophony' (pp 9-10). In the absence of sufficient information and/or understanding, the stories woven about the self by those with chronic conditions, in particular those with CAH, are likely to be inaccurate, even bizarre. Where social and psychological issues are not discussed, inaccuracy may increase. The outcome may be woven stories of themselves, stories in which they perceive themselves to be unacceptable to others.

As both the content of medical knowledge and the definition of areas of important concern and investigation for those who are ill are socially constructed (Atkinson, 1988; Turner, 1995; Lupton, 1994) it is possible that changes occurring in such constructions will have implications for the delivery of health care. It may no longer be possible, given an increase in the breadth of appropriate concern, for one individual to have sufficient expert knowledge of all the above relevant areas believed to effect any chronic condition. Clinical management that aims to address broader areas of life's experiences would entail a team approach, members of the team contributing expertise in disciplines traditionally concerned with the various areas. The support provided by the data from the research reported here, for such an approach will be considered in the final section.
CRITICAL EVALUATION

In the methodology section of this thesis, questions relating to the critical evaluation of qualitative research methods were discussed. It remains necessary to evaluate the present study in relation to those issues.

Qualitative methods should be as open to critical assessment as are methods which are quantitative (Gabriel, 1990). Parallels have been drawn earlier between the critique criteria for the two methods; whereas in quantitative research the validity, reliability and generalisability are the recognised criteria, in qualitative methods credibility, confirmability, transferability and dependability are seen as more appropriate concepts (Ashworth, 1987; Gabriel, 1990).

In the conduct of the current research, credibility was addressed during the interviews by the technique of reflection, that is the reflection back to the interviewee of the interviewer's perceived meaning of their statements so as to establish correct understanding. Such reflections produced a number of both confirmatory and disconfirmatory responses, suggesting that the technique aided in the maximising of credibility. Discussion, presentations of transcripts and review with another research clinical psychologist of the ongoing analysis served as checks on the confirmability of the findings. In addition, throughout this report a variety of quotes from various women have been presented to enable
similarities and differences to be used to confirm the construction of categories, concepts, dimensions and elements which have made up the proposed model.

Detailed quotes also enable the reader to evaluate the suggested links between the components of the analysis, and to evaluate whether these links are grounded in the data. As the necessary division of the contents of the interviews into arbitrary sections results in a loss of the overall ambience of the interview; two verbatim transcripts, one from each group, are included (see Appendix V) to enable the reader to examine the tenor of the interviews. In addition, an example of the system used to progress from the raw data to the identification of elements, dimensions, concepts and categories can be found in Appendix VI.

Satisfactory fulfilment of assessment criteria in qualitative research ultimately rests on both the sufficiency of detail and the clarity of the induction process in the construction of meaning from identified components. Strauss and Corbin (1990) suggested that the explanatory power of a grounded theory was a function of the 'tightness' of the proposed links between the components; that is, the demonstration of differences and similarities between component relationships and the attributions of such variations. In the current research report, such similarities and differences have been described, and attributed to differential experiences of those women with diabetes and those with CAH.

A grounded theory should also consider the broad contextual conditions and their possible links to the phenomena under study (Strauss and Corbin, 1990; Henriques et al, 1984). The consideration of some cultural discourses and the
connections of these to, and their importance in, understanding the women’s experiences has formed an important strand of the discussion and formulation of the proposed theoretical model.

The essential criteria for acceptable evaluation of a qualitative study have therefore been met. In addition, the method appears to have achieved the goal of the analysis of the data - to increase the awareness of the ways in which the women had come to understand themselves in their social world. There were however, some difficulties that require consideration.

The method allowed the possibility of the collection of data across a number of broad topic areas, but it was inevitable that not all the interview topics proposed at the planning stage of the study were covered in depth. Firstly the procedural objective of the study was both to allow the women to identify and explore areas of importance to themselves while ensuring a reasonable comparability across interviews. Secondly it was necessary to work within acceptable time limits for the women who were participants in the research. The decision not to pursue some areas does not imply that they were of little importance, rather that not all areas could be covered in a single research interview.

Although the final sample of women in the group with CAH appeared to be representative of those registered at the hospital as children, they were biased in a number of ways. The larger part of the group of women with CAH were severely masculinized and were salt-losers. It is of course possible that less severely affected women would have constructed different meaning from similar
experiences, or have had different experiences. Further investigation into the experiences, and the meanings construed from them, by those with less severe variants of CAH is required in order to establish the transferability and dependability of the findings across all ranges of severity of the chronic condition. A second factor related to the sample of women with CAH was their attendance at the same hospital throughout childhood. Differences in the management styles of paediatric clinics may result in different experiences and the comparison of such differences would allow important management effects to be identified.

Difficulties were also experienced in obtaining a sample of women with diabetes who had been diagnosed at an age comparable to the women with CAH. It would not have been practicable to obtain a sufficient sample whose diabetes was diagnosed at birth. The differential age at diagnosis may account for some of the differences between the groups. However, experiential factors other than age at diagnosis seemed to be implicated in the differences that were found; the contributions of experiences suggested in the discussion must remain as proposals until opportunities to examine the effects of age at diagnosis are obtained.

The interviews provided a rich source of data and the identified relationships enabled the proposal of a dynamic model. The aim of going beyond the question of 'How much?' to increase understanding of 'What it was like' to experience a chronic condition was realised. The process of induction allowed the construction of a model that focused on meanings constructed by the two
groups of women out of their experiences. It is for future research to provide support for this model, and to identify variations attributable to particular features of chronic conditions and concomitant experiences.

The issue of transferability of findings raises the problem of historical factors (Cook and Campbell, 1979). The childhood experiences of the women in the study occurred some decades ago, and changes in medical management techniques and clinical skills may have rendered the findings out of date. In order to investigate this issue data was obtained from a group of parents with young children with CAH. The design and the results of this subsidiary study are reported in the following section.
CODA: AN UPDATE OF REPORTED EXPERIENCES

INTRODUCTION

It is of course possible that medical treatment, the nature of the relationships between patients and the medical advisors, and styles of communication within the consultation have changed during the last few decades. Information available to the public through the media and the publication of both national (1995) and local Patient Charters has increased awareness of patient rights. These rights include that of clear explanations about proposed treatment and access to personal medical record. Access to medical records and explanations about treatments would include information about the reason for, and the results of, medical tests. In combination, these service goals suggest that current visits to paediatric clinics by the parents of female children with CAH, may have a very different outcome from those reported in the main study. An examination of the possibility of such changes was provided by data obtained from a supplementary questionnaire study.

METHODOLOGY

The supplementary parental questionnaire study

Parents who were members of a newly formed support group for those with CAH and were attending a conference agreed to the circulation of a
questionnaire and covering letter (see Appendix VIII). One hundred and sixty-nine mothers and fathers of children with CAH representing eighty-five families received the questionnaire.

The questionnaire and procedure

The parental questionnaire was constructed to fulfil two objectives: to obtain feedback relating to the conference, and to obtain information about the parents’ responses to and perceptions of a number of experiences; the responses relating to the conference are, of course, of no interest here. The experiences mirrored those that were of importance to the original sample of women with CAH. The areas were as follows:

- the respondent
- the respondent's family, including members with CAH
- diagnosis, medication and operations relating to CAH
- understanding of CAH
- emotional response at diagnosis
- communication about the condition
- anxieties relating to the condition
- satisfaction with the outpatient hospital visits

Each parent was asked to complete the questionnaire without consultation with the other parent and to return it separately in a pre-paid envelope.

The questionnaire returns and the selection of the data sample

Questionnaires were returned from forty-eight mothers (56 per cent), and thirty-two fathers (38 per cent); this represented forty-nine families (58 per cent). Fifty-two children with CAH, with an age range from less than one year to
RESULTS AND DISCUSSION

The sample characteristics

In order to investigate possible recent changes in some of the issues identified in the main study, those questionnaires from parents of girls with CAH born in the last ten years were identified and it was these that were used in this coda analysis. These questionnaires represented eighteen families comprising nineteen girls with CAH, eighteen mothers and twelve fathers. The characteristics of these girls are given in Table 20.

TABLE 20: Characteristics of the sample of girls with CAH identified from the questionnaires

<table>
<thead>
<tr>
<th>Sample Characteristics (N=19)</th>
<th>Frequencies</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age: mean = 4 years</td>
<td></td>
</tr>
<tr>
<td>range = 3 mos -10 years</td>
<td></td>
</tr>
<tr>
<td>Age at diagnosis:</td>
<td></td>
</tr>
<tr>
<td>-before one week old</td>
<td>17 (90%)</td>
</tr>
<tr>
<td>-before three weeks old</td>
<td>1 (5%)</td>
</tr>
<tr>
<td>-at 3.5 years old</td>
<td>1 (5%)</td>
</tr>
<tr>
<td>Age at surgery:</td>
<td></td>
</tr>
<tr>
<td>-under 2 years of age</td>
<td>4 (50% of this aged children)</td>
</tr>
<tr>
<td>-over 2 years of age</td>
<td>15 (100% of this aged children)</td>
</tr>
</tbody>
</table>

Because data relating to age and type of surgery in the main research and the coda study were obtained from quite different sources, the medical records and current recollection, it was considered that no direct comparison of these
characteristics identified by the questionnaire sub-set and the main research sample could be reliably conducted. By inspection however, the two groups would seem to be reasonably comparable; the identified subset appears to be a group of early diagnosed children who had undergone surgery and were therefore quite severely effected young girls with CAH.

**Parents’ understanding of CAH**

One of the central questions to consider was the extent of the parents’ understanding of their daughters' chronic condition. It was suggested in the main study that the reported low level of understanding and knowledge of the women’s parents’, through the failure to provide an accessible source of information, had been a contributor to the low level of understanding by the women with CAH. The scoring system used in the main study was applied to the responses given in the questionnaire; the 'bits' of information which corresponded to those required by the endocrine consultants were totalled and expressed as a percentage of the required total score. The statements made by the parents to 'Describe in your own words what CAH is', suggested that although the percentage scores were double those of the main study (see Table 21), they still represented, at best, less than 25 percent of the information believed to be the satisfactory minimum. In addition, the top of the score range for both mothers and fathers was lower than that of the main study.
TABLE 21: Comparison of the median, modal and range of percentage knowledge scores for the main and questionnaire studies

<table>
<thead>
<tr>
<th>SAMPLE</th>
<th>MEANS</th>
<th>MODE</th>
<th>RANGE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Main study N=19</td>
<td>8.8</td>
<td>8.0</td>
<td>0-33</td>
</tr>
<tr>
<td>Questionnaire-mothers N=18</td>
<td>16.6</td>
<td>17.0</td>
<td>0-25</td>
</tr>
<tr>
<td>Questionnaire-fathers N=12</td>
<td>16.6</td>
<td>17.0</td>
<td>0-17</td>
</tr>
</tbody>
</table>

The median and modal scores were in the lowest percentile in both studies.

Examples of parents' statements, with percentage scores, show the similarity of level of knowledge to those in the main study:

*I still don't quite understand what CAH is, I know she has hormones missing and it effects her adrenal glands. And she needs the medicines to keep her quite normal. (18%) (Mother of a 1.5 year old).*

*It's because my genes and X's father's genes did not match, and this caused X to keep growing while I was carrying her. (0%) (Mother of a 5 year old).*

*A condition which is caused through the genes of the parents (1%) (Father of a 4 year old).*

The parents as an information source

For the parents to be effective sources of information for their children, they must feel able to discuss the condition with them. Five of the mothers, whose daughters ranged in age from under one to four years, did not complete this section because, they stated, their daughters were too young for discussion. The remaining fourteen mothers indicated on a ten centimetre line how easy they found talking about the chronic condition to their children. The frequencies of ‘easy’ and ‘difficult’ obtained by dividing the scale at the mid-point, suggested that ease of discussion might be related to the current age of
the child, but the distribution of scores between daughters aged 0-4 and 5-8 years, just failed to reach significance (see Table 22). Less than half of the fathers answered this question, and their scores were therefore not analysed. The implications of the fathers' omission to answer the question forms part of a general finding related to their perceived role that will be discussed below.

**TABLE 22: Frequencies of mothers reporting ease/difficulty in talking to children about CAH**

<table>
<thead>
<tr>
<th>AGE OF CHILD</th>
<th>REPORTED 'DIFFICULT'</th>
<th>REPORTED 'EASY'</th>
<th>TOTALS</th>
</tr>
</thead>
<tbody>
<tr>
<td>0-4 years</td>
<td>5</td>
<td>2</td>
<td>7</td>
</tr>
<tr>
<td>5-8 years</td>
<td>2</td>
<td>5</td>
<td>7</td>
</tr>
<tr>
<td>TOTALS*</td>
<td>7</td>
<td>7</td>
<td>14</td>
</tr>
</tbody>
</table>

*N=14, 5 mothers omitted to answer as daughter perceived to be too young.
Fisher's Exact Probability Test. NS

Decisions about 'telling' also involve decisions about who should tell, when explanations should be given, and what should be told. Parents were considered to be the most appropriate informants by 100 percent of the questionnaire respondents. In line with the general responsibility held by mothers in relation to their children's health, only two of the thirty-two fathers who returned the questionnaire (6 per cent), had been involved in explaining about CAH. Such rejection of shared responsibility may partially account for the fathers' failure to complete the question requesting information about how easy or difficult they found this task - they were not engaged in such a task.

In answer to the question, 'What do you think is the best age to tell your child?' the advisability of early explanation was expressed in such terms as 'As soon as she can understand', or 'Whenever she asks'. As the data suggested that mothers
were regarded as responsible for giving information about the condition, and this was reported as a source of some difficulty for them, two further sections of data were analysed and related to this issue. The sections were: the answers to the direct question asking what had been told to their daughters, and data from a question asking for the identification of 'difficult to talk about topics'. What had been told appeared to be related, as would be expected, to the girl's age: for pre-school children it was the need to take the medication; for primary school children the idea that something didn't work properly had been added:

*She has been told that when God made her, one part didn't work properly so He has told the doctors how to help mend her. That it is important that she takes the medicine.* (Mother of a 5 year old)

The data indicated that for secondary school children, parents believed that 'everything' had been told. In the light of the low levels of knowledge and understanding by the mothers in this sample, the clarity and adequacy of the explanations must be questioned. The 'difficult to talk about' topics were those that might be expected, and centred round two main issues: the abnormalities of the genitalia together with the necessary operations, and the anticipated effects of CAH on sexual behaviour and relationships:

*The aspect re the operation and its aftermath is so difficult I lie awake at night worrying about the best way to approach it. I know 'matter of fact' is best but such an approach is difficult when one is so involved.* (Mother of a 10 year old)

*I find it difficult to talk to her about the ops. as I worry she will think of herself as different to her friends.* (Mother of a 7 year old)

*...as she reaches her teens when she will obviously be far more aware of the implications of CAH, she is very likely to have psychological problems and worries relating to future contact with boy friends and hence a sex life.*
These very difficult to talk about topics are identical to the concerns and areas of confusion identified by the women in the main research. It would seem that the decades since the women in the main study attended outpatient clinics with their parents have not changed attitudes towards discussion of intimate sexual details within a medical context. Unless the parents can be helped to address these issues, their daughters are at risk of carrying with them into adulthood similar difficulties to those of the older sample.

**Communication issues**

Issues of communication are the final comparison made between the main and supplementary findings. The main research placed considerable emphasis on the contributions of two areas of communication to the development of the 'Skeleton-in-the-cupboard'. The areas were those of communication between the clinic doctors and their patients, and between the individuals with CAH and their friends and relations. Information about these was obtained in the Coda study from the questionnaire responses.

*Communication with clinic doctors*

The data from the parents on issues of communication about their daughters’ chronic condition is not directly comparable to that of the main study because of two main differences: differences in the time lapse since the experience, and the role position of the providers of data.
The main study provided data on the recalled experiences of being a paediatric patient, the subsidiary study provided data on current experiences by parents who were accompanying their children to hospital clinics. Any variation between the two sets of data therefore may reflect these differences rather than be attributable to differences of experience. However, the review of research by Brewin et al (1993) concluded that the retrospective reports by adults of central features of their specific childhood experiences were likely to be reasonably accurate. If this is accepted, then the similarities between the two sets of reports would be valid. On examination, the current and retrospective clinic communication experiences reported by the women with CAH in the main study appeared to be very similar to those of the parents in the subsidiary study. Although the statements of the parents included positive feelings about supportive relationship with regular doctors, there were strongly expressed desires for more information about both CAH in general, and about the health status of their children:

The hospital is very good. I think they could explain more. And give me results from blood tests. I never even find out about how her blood is doing.

I would like the doctors to be more forthcoming, instead of me having to ask all the time.

We feel uninformed on the practical aspects of CAH and its management.

I have asked for some written information about CAH both for myself and my child, but none seems available.

A main issue for these parents was the need for counselling and support; this it was felt, would provide both information and opportunities for relaxed discussion of feeling; It was seen to be helpful for such opportunities to be provided by professionals and/or other more experienced parents of those with CAH.
Most doctors do not have the conversation or counselling skills and I think it needs careful handling. For preference, I would like the counselling for myself, and then use it to tell my daughter more. .......I think if time could be found, parents should have the opportunity for a private discussion with the medical team involved in the child's care so that questions can be asked and as full a picture as possible given. The only one we've been given was at birth, when the doctor in charge of the delivery gave as much information as he himself was in possession of. Naturally, one forgets, and the memory distorts as time passes. I would now like to see someone with expert knowledge of handling the problems created by our daughter's condition......I would like to talk to these experts without my daughter present. The doctor has often disclosed information in front of her which has upset her and would have been better left 'til later.

...more time to sit and talk to someone about X's illness. Should be more help for mothers, someone outside the family who could explain to the child what she should or should not do. Should also be more local doctors that know about CAH.

_The hospital is good.....however, the question of isolation remains._

These communication difficulties between the parents of young girls with CAH and the doctors at the hospitals, clearly reflected both the childhood memories and the current experiences of those women in the main research. The final area of communication about which the questionnaire provided data was that between parents and their friends and relations.

_**Communication about CAH between parents and their friends and relations**_

The differences in sources of data referred to in the previous section are also relevant to this issue. The questionnaire study did not provide information on communication between the children with CAH and their friends and relations, however there was information on who else had been informed by the parents about their child's condition. All of the families reported that grandparents and
close aunts and uncles had been told. Almost three-quarters of the families had informed their very close friends, and where the child was of school age, class teachers had been given some information. This would seem to be a considerable increase of communication over that reported by those in the main research; there, the impression was of an almost total absence of communication about the chronic condition. Additional questionnaire information however, suggested that communication remained extremely difficult, and information given was expressed in vague terms:

*I find it very embarrassing and difficult, talking to the grandparents and friends.*

*Their knowledge is very limited. We simply say they are salt-losers and require medication and that in the event of an emergency requiring an anaesthetic, the hospital should be contacted so that the necessary injection and medication could be given.*

CONCLUSIONS FROM THE COMPARISONS OF FINDINGS OF THE MAIN AND SUBSIDIARY STUDIES

The contemporary data from the parents of young girls with CAH suggested that little had changed during the decades between the experiences of the two groups of respondents. The current parents' understanding of the condition remained low, and they found it difficult to discuss the condition either with their daughters or with others. Although there was a reported increase in communication with close relatives, friends and teachers, the content of information given was very limited and was restricted to that thought by the parents to be important in the event of any medical emergency. This type of information is perhaps very appropriate for friends and teachers, however it fails
to address the issue of the perceived acceptability of the condition as something that can be shared as a topic of communication with appropriate individuals. The similarity in the experiences of feelings of isolation, and the perceived lack of opportunities for relaxed discussion are clearly seen. These outcomes were proposed as being attributable to communication difficulties and the similarities are likely to repeat the development of a 'Skeleton-in-the-cupboard' for young contemporary girls with CAH through the same mechanisms as those proposed in the main research.

The similarity of findings between the supplementary and main research strongly supports the proposal that consideration be given to identifying improved ways of increasing understanding by patients and parents of CAH and all its wide implications. It is especially important to provide support that allows the acceptance of CAH as a topic of easy discussion between appropriate individuals who are involved in the care and development of those with the condition. Appropriate individuals in this context must not be limited to the medical team.

The implication of the findings for the management of CAH will be further considered in the next section of the report.
OVERVIEW AND MANAGEMENT IMPLICATIONS

OVERVIEW OF THE RESEARCH FINDINGS

The aim of the research reported here was the identification and understanding of the constructed meanings of the experiences of women with a chronic condition: the condition of CAH. In the introduction a number of issues were raised, issues which have been examined through the analysis of the data and which are now re-considered.

An important component of the research was the examination of some of the transactions between the individual and the social and cultural context in which their experiences were embedded (see Figure 1). This component was examined by the consideration of a number of cultural discourses believed to be relevant to the experiences of CAH. Those with a chronic condition, their parents, and the doctors responsible for medical treatment enter into the understanding of experiences with a repertoire of beliefs or discourses that constitute part of their social learning experiences. As Foucault (1991) suggested, there are likely to be complex relationships among a collection of discourses perceived as relevant to any experience; they will structure an experiential event by specifying both relevant areas of knowledge, and the permissible topics for discussion about that event. Although the affect of discourses on the construed meaning of any particular event will be individual and attributable to the position taken by the
individual on that discourse, it might be expected that different individuals would mainly perceive similar discourses as influential in similar events. The following discourses effecting experiential outcome were identified in the interview data.

The discourse of 'the opposite sex', together with the associated belief that gender differences are 'programmed-in', were proposed as likely to structure decisions about which characteristics of those with CAH would be perceived as acceptable or as pathological. Where concern about the gender appropriateness of behaviour is minimal, incidences of behaviour considered by others to be that of the opposite sex, for example 'tomboy' behaviour in girls, may be attributed to 'a passing phase' or believed to be socially acceptable for female children. Where the concerns about the gender appropriateness of behaviour by any of those involved in the care of the children are stronger, this behaviour may be perceived as pathological and thus become a source of anxiety. Such behaviour may be seen as requiring change. When construed through the related discourse of gender differences being 'programmed-in', such pathological behaviour may come to be considered unchangeable. If hormones are believed to be a 'programming-in' mechanism, this outcome is particularly likely when there is a diagnosis of CAH, a chronic condition where imbalances to hormones commonly perceived as male or female related, are a primary element of the known pathology.

The second component of the 'opposite sex' discourse, that concerned with the belief that women must possess particular physical characteristics to be
attractive to the opposite sex ('Happiness is having some-body'), may create or increase dissatisfaction with personal appearance. This will be especially so when personal appearance does not conform to the socially constructed ideal of 'beautiful' or 'sexy'. Personal appearances that do not meet such requirements may add to the self-perception of women with CAH as being 'different' from other women, and to the idea that they do not belong to the group of females held by their culture as desirable people. The discourses surrounding the essential possession of a perfect body, of course refer not only to the current stereotype of 'tall and slim', but also to the appearance and satisfactory functioning of the external genitalia. The acceptability of their genitalia to sexual partners, together with the discourse that places relationships at the centre of women's sense of achievement (Hollway 1989), may add weight to any dissatisfaction with self, any feelings of being 'different' by women with CAH.

The discourse that holds the doctor is an 'expert', a 'specialist', a 'scientist', may also have effects on the meanings constructed from the experiences of visits to hospital outpatient clinics. In addition, the traditionally held view of the authority of the doctor who 'knows best', while tempered by the increasingly popular view of the patient as knowledgeable about their own symptoms and condition, may be influenced by factors related to a specific chronic condition. When the culturally expected authority is placed against a background of incomprehensible communication about a condition which has no cure, then the validity of the doctors' authority, knowledge and power (Turner, 1995) is likely to be questioned. These, then, are some of the 'filters' through which parents, doctors, the women themselves and others concerned with understanding the
chronic condition, view their experiences and attempt to make sense of them; they will colour the stories they weave (Stainton-Rogers, 1991).

An issue raised in the introduction was the degree of possible intrusion made by the characteristics of the chronic condition into the individual's day-to-day activities (Strauss et al., 1984). The findings of the current research in relation to this issue were complex. Although women in the diabetic and the CAH groups did not perceive their condition as preventing them from engaging in either the ordinary activities of childhood or of adulthood, both groups felt their condition had resulted in poor attendance at school. However, the effects of school absences differentially affected the groups. Those with CAH attributed their lowered academic achievement to their frequent absences, and their unsatisfactory careers to poor academic progress at school. In contrast, those with diabetes did not feel absences had hindered their academic attainment, and their interest and commitment to education separated them from the group with CAH. Their higher level of educational achievement had enabled them to make better progress in, and plans for, their careers.

In addition, the women with CAH attributed their level of sexual experience to their condition. They believed that their low level of sexual activity resulted from their concerns about difficulties in penile penetration. These difficulties, either anticipated or experienced, they attributed to the vaginal surgery. Their lack of confidence in the surgical correction, together with the discourses relating to the important need to conform to the socially constructed but mainly unspecified standards of the female body, resulted in considerable anxiety and
embarrassment about the acceptability of their genitalia to their sexual partners. This in turn resulted in considerable hesitation about engaging in sexual relationships.

For those with diabetes the intrusions of their medical condition into their everyday life were different and were related to the practical daily demands of their medical regimen. Although some embarrassment about dietary and injection needs was experienced, this had mostly been minimal and was not perceived as limiting their activities; overcoming these requirements by the women with diabetes, was frequently at the expense of satisfactory control of blood glucose level.

The two groups responded in different ways to other effects of their condition on their lives. Those women with diabetes, although their strategies for minimising intrusions sometimes resulted in sacrificing good diabetic control, appeared to take an active role in the control of effects. In contrast, those with CAH were more passive in their response to condition effects, rarely perceiving any non-medical causes or remedies that might mitigate intrusive effects. It is true that the condition characteristics responsible for intrusions were of different levels of social acceptability. For those with diabetes, although injections are not generally administered in public they are within the experience of most individuals, everyone has had the need at some time of their life for this procedure. The same applies to diets; all individuals and members of their social networks are likely to have been ‘on a diet’ at some time for either personal or medical reasons - the discourse relating to ‘good health’ is ubiquitous in western
society. In contrast, the intrusions for those with CAH were experiences and procedures relating to and focused on 'private parts'. Society has still firm prohibitions on the display of or even discussions about details of personal characteristics of sexuality.

The issue of general or specific to the condition effects of chronic conditions (Nolan and Pless, 1986; Cassileth et al, 1984) cannot then, be simply resolved. The comparisons made in this research between those with CAH and those with diabetes, suggest that the conditions have some general effects, but that some are specific to the condition. In addition to school absences, both groups recalled their attendance at hospital clinics as intrusive, unfriendly and as providing a poor learning resource. Both groups also experienced similar conflicts between their self-perception as 'normal' or 'sick'. These similar experiences, however, resulted in some outcome differences for the two chronic conditions. It is proposed that these different outcomes were attributable to the differential development of the 'Skeleton-in-the-cupboard' and to the on-going need to 'keep the secret'.
A report by Kuhnle et al (1995)\(^1\) of an investigation of the quality of life for 45 women with CAH (mean age = 27 years), compared to those of a matched healthy control group, covered many of the areas of investigation of this thesis, and in those areas showed very similar findings to those reported here. Kuhnle et al assessed the quality of life through two procedures; a semi-structured interview and the completion of ten questionnaires covering the four parameters identified by Furberg (1985)\(^2\). These parameters of the quality of life were: physical state (complaints about symptoms of the disease and the effects of treatment), psychological wellbeing (moods and strategies developed to deal with moods), social relationships (the amounts and quality of relations to others and social activities), and functional capacity (the ability to master everyday life demands).

The outcome of the study included the finding that the quality of life overall (as evaluated by the individuals themselves) was not reduced compared to the healthy controls. The authors suggest that this reflected ability by the women to

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cognitively minimise both the effect and treatment of the condition on their lives. This mirrors statements of the women in the current study emphasising their belief that their condition had not limited their lives. However the detailed analysis in both studies suggests that those with CAH differed from the comparison groups in all aspects of the psychosocial area. Common findings included: more of the women with CAH were sexually inactive, were unsuccessful in establishing close friendships and partnerships, saw themselves as socially shy, were less satisfied with their sexual experience, were described as portraying decreased information seeking behaviours. Furthermore in neither study was an increase in lesbian tendencies of the women with CAH compared with the comparison group reported. Kuhnle et al state, ‘In the interview it became apparent that it was not easy for these women to disclose their thoughts and feelings about their body and its sexual function.’ (p.714).

The proposed concepts that contribute to the ‘Skeleton-in-the-cupboard’ are reviewed in Figure 4.
The main theme that links these concepts is communication. The complexity of the condition, the required acquisition of understanding and new areas of knowledge, the effects of cultural discourses and the socially sensitive nature of CAH, culminate in a difficulty in finding a language with which to communicate and discuss the experiences integral to the condition.

The attributions and outcome aims of parents, doctors, counsellors, those who design and carry out research, as well as individuals with CAH may all be affected by both the discourses of the culture in which they exist, and by the absence of a language with which to explore the meanings of experiences. The achievements of those who professionally manage the well being of those with CAH will depend upon the recognition and implementation of such issues in
management approaches. Such implications for the management of CAH are discussed in the final section.

**IMPLICATIONS OF THE RESEARCH FINDINGS FOR THE MANAGEMENT OF CAH**

The findings of the main research study together with those of the supplementary questionnaire study, suggest that Strauss et al's (1984) final characteristic of chronic conditions, that is the development of new skills and strategies to handle new problems, becomes very pertinent to the maximum adaptation to CAH. Whereas for those with diabetes the necessary new strategies and behaviours are mainly knowledge and skill based, for those with CAH they include major cognitive restructuring. Knowledge about the hormonal control of prenatal development may be important, but is less psychologically demanding than the required shift in ways of construing and discussing events occurring as part of the experience of growing up, and living as an adult, with CAH. The central change requires the questioning of the socially entrenched cultural discourse relating to the 'opposite sex'. The belief that males and females are two independent sexes with fixed behaviour and skills must be replaced by the belief that they have a common beginning and that gender differences are socially constructed. This is a difficult endeavour because, to use Bem's (1993) terminology, 'the internalised lenses...make the societal ways of being and behaviours seem so normal and natural that alternative ways of being and behaving rarely even come to mind' (p.151-152).
The model proposed in this thesis is based on the construction of meanings placed on experiences by those with CAH or with diabetes, and the experiences can be seen to be organised around three levels. The levels are those within the individual including knowledge of the condition and related concepts and attitudes; those arising from interaction with others in the individual's environment; and those existing within the culture, in the discourses and institutions of the society within which the individuals gain their experiences. Interventions leading to change in any or all of the three levels of experience could be suggested; there are established psychological interventions that may be helpful in attempting to institute changes. However the above division must not be taken to imply that each level could be matched with an intervention approach. The specific components of any difficulty would need to be identified and from this, appropriate cognitive, educational, organisational and/or skills based approaches chosen so as to effect any appropriate changes to ameliorate the problem.

The need for a thorough analysis of the problems, including medical problems, implies the need for a multidisciplinary service team (Elander and Midence, 1997) from diagnosis, through childhood and into the adult years (Garwick, 1995). In addition to medical specialists, clinical, developmental, educational and health psychologists would have important roles in the provision of services designed to meet such broad areas of need. The aim of such a service would be to identify individuals at risk, to provide intervention and support where and when required (Dunn, 1995) and to provide an easily accessible informational resource.
Health professionals share with the patient the experiences of life in a two-sexed society, a society that sorts individuals, attributes, behaviours (and objects) on the basis of the polarised social definitions of masculinity and femininity. The task for those who help women with CAH to understand their experiences will be to attempt to deconstruct this dichotomy for themselves and their clients, to overcome the cultural belief that the biological sex of the body should match and dictate an individual’s psychological and behavioural attributes. Such a deconstruction comprises a three-sided task. First, the understanding of the physiological and anatomical similarities of male and female sex — biological sex is not as polarised as might be thought. Secondly, the understanding of the attribution of gender — placing it firmly as being constructed by society rather than ‘biological sex driven’. Finally, the removal of the assumption that there are ‘‘real men” and “real women” rather than “real humans” ’ (Bem, 1993. p.194). The attempt to change the cultural discourse of the ‘opposite sex’ is the attempt to sever the culturally constructed connections between sex and gender.

The identification of the deficiencies or lack of communication as a common thread throughout the development and maintenance of the 'Skeleton-in-the-cupboard' implies an additional task; the acquisition of a language through which the unsayable can be said and shared. Communication difficulties were identified in the main and questionnaire research studies, and the following quote from a questionnaire response emphasises that the need for help is clearly perceived by today’s parents. The questionnaire respondent was a mother of a ten-year-old daughter and a thirteen-year-old son, both of whom had CAH:
I booked a private appointment with the surgeon who will perform my daughter's operation because it worried me so much. I found this immensely helpful and found that my imagination had created problems which didn't exist. I think if time could be found, parents should have the opportunity for a private discussion with the medical team involved in their child's care so that questions can be asked and as full a picture as possible given. The only one we were provided with was in the hospital at the time of birth when the doctor/consultant in charge of the delivery gave as much information as he himself was in possession of. Naturally one forgets and the memory also distorts as time passes. I paid for my appointment because no offer had been made by the hospital, so it seemed the only way of gaining necessary information. Now I would like to see someone with expert knowledge of handling the problems created by our daughter's physical condition - a psychiatric councillor of some sort.

The quote highlights two important aspects of the help the parents feel they require: the amelioration of the general lack of information resources currently provided by the routine outpatient services, and the provision of more than one opportunity to learn and to discuss their concerns. The requirement for repeated opportunities is very relevant for both those with CAH and those with diabetes. During childhood, difficulties appear and re-appear throughout the developmental period even though in a slightly different form and context. The developing abilities of the child necessitate explanations and strategies for dealing with problems that are presented at different cognitive levels (Eiser, 1985).

On-going support for parents, particularly mothers, of children and young people with chronic conditions would be extremely beneficial. The published research findings of this area support two important issues: that mothers carry the day-to-day responsibility for the medical and emotional care of the children,
as also evidenced within the present research, and the importance of the mother’s mental health in the adaptive development of the child. For example Miller (1993), in a review of research on chronic rheumatic diseases in children, concluded that the child’s social support, especially maternal competence, was the most important influence on psychosocial adjustment. Whatever the direction of causality of the reported correlation between maternal mental health and behavioural difficulties among children affected by chronic illnesses (Midence et al 1996) interventions providing continuing support for the carers would surely be of benefit. Equally, a single presentation of information is unlikely to be sufficient for patients; failure to remember complex material presented during consultations with medical advisers, especially when stress and anxiety levels are high, has been well documented (for example Ley, 1988). It follows that opportunities for information acquisition, discussion and support are required throughout the life of any individual with a chronic condition.

In conclusion, the proposed model provides suggested areas of possible change and intervention that would be of benefit to both those experiencing the chronic condition of CAH and those responsible for their care. Adequate management would require a multidisciplinary team to identify those at risk, those already experiencing specific difficulties, and those needing continuing support. The need for a broader style of help, particularly psychological counselling, is felt by the parents of young girls with CAH and by adults with the condition. The recent formation of a national parent-led CAH support group is a hopeful active development, and may be instrumental in encouraging the development of a language with which to share issues and information relating to the management
and experience of CAH. These events may enable doctors, parents and those with the condition to prevent the development of a 'Skeleton-in-the-cupboard'. An equally major task for all, however, is the deconstruction of cultural discourses supporting the polarisation of gender into 'real men' and 'real women' and, to encourage instead, the construct of 'real humans'.

Recent advances in genetics have led to procedures that allow antenatal diagnosis of CAH and intrauterine hormonal treatment. Such procedures are at present limited to a second pregnancy where an affected child has already been born to the parents, where there is an affected sibling of the mother or father, or where the pregnant woman and/or her husband have been diagnosed as having CAH. In such cases, hormone treatment with dexamethasone is initiated as soon as the pregnancy is confirmed and where an affected female fetus is established, continued until the birth, with the aim of preventing fetal exposure to excess androgens and resulting masculinization of the external genitalia.

Mercado et al (1995) have report on the outcome of nine CAH affected female fetuses where dexamethasone was pre-natally administered. Where the administration of the hormone was at or before ten weeks gestation; four were.

---

born with completely feminized genitalia and five were mildly affected (Prader 1 or 2), levels that were less affected than expected in comparison with the other affected member of their families.

These results are encouraging and with respect to females with CAH born without any masculinization would seem to suggest that in these cases, what has been called here the ‘Skeleton-in-the-cupboard’, would be eliminated. However, the prevention of masculinized genitalia may not be sufficient to result in the total elimination of the ‘Skeleton-in-the-cupboard’; there may be remaining experiences which would minimise but not eliminate the need to keep the secret.

It has been proposed in this thesis that a common element contributing to the development of the ‘Skeleton-in-the-cupboard’ is that of communication; in particular, that communication of information leading to a clear understanding of the chronic condition is essential for good outcome. At all success levels of prenatal hormone therapy there will remain a number of issues that will require clear communication. The need for continued administration of hormone replacement after birth requiring as it will regular visits to hospital clinics, and, for those without complete genital feminisation, the surgical repair, will require an understanding of why these experiences are necessary. The difficulties of normalising the physiological developments at adolescence, and possible fertility problems in adulthood, including the issues of genetic transmission of CAH, would be two key occasions of informational and counselling need. Furthermore, even the complete elimination of prenatal genital masculinization
would not eliminate the effects of all the socially sensitive aspects of CAH nor those of the prevailing cultural discourses. The need to develop a socially acceptable language to enable open discussion by all concerned with the management of those with CAH would remain unchanged.

Prenatal diagnosis, even where complete genital feminisation has not been achieved, would allow any ambiguity about the sex of the child to be at best eliminated, and at least considerably reduced. It would seem that the identification of the XX chromosome, together with an understanding of the effects of androgens on the external genitalia, should enable the parents to more readily accept that their baby is a daughter. This acceptance however, would only be achieved where there is education, support and counselling commencing during the prenatal period as soon as the issues about CAH are raised. Improved confidence in the decision about the sex of the child should lead to more consistent styles of parenting behaviours and expectations in keeping with the cultural beliefs. It may remain difficult for the parents of more genitally masculinized neonates to accept a female classification. In these cases, continued education and counselling would perhaps need to be more intensive.

In summary, although the administration of prenatal hormones makes possible encouraging changes in the physical status of some females with CAH, this procedure would necessitate early counselling and would not necessarily result in the removal of all post natal risk factors leading to the development of the ‘Skeleton-in-the-cupboard’. Not only will there be remaining issues which need consideration for those individuals where the intervention has resulted in
complete genital feminization, but there will also remain substantial issues in relation to those females born with masculinized genitalia - at the current rates of success a considerable proportion. The present screening policy will leave unaltered the risks of genital masculinization for females born to families without a previously effected child. In addition, for a considerable time, there will be children and adult women born before the development of prenatal diagnosis and treatment who have been to some degree prenatally affected by excess androgens. Long term follow-up of pre-natally treated CAH females will also be required to identify both physical and psychological outcomes. An awareness of the experiences contributing to, and the effects of, the ‘Skeleton-in-the-cupboard’ remain vital to the optimal management of all females with congenital adrenal hyperplasia.
REFERENCES


Care: Converging Medical and Psychosocial Perspectives. pp.49-56. Chur: Harwood.


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APPENDIX I. EXAMPLES OF LETTERS

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b) Examples of consent forms 264
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   ii) Co-operating adult hospital
c) Second letter of invitation 266
d) Letter to adult endocrinological consultant requesting agreement to invite a patient with CAH to be part of the study 267
Dear

I am writing to you as a former Great Ormond Street patient to ask you to help us with our project on Congenital Adrenal Hyperplasia and Diabetes. Dr Grant and I would like to find out how you feel about some of your recent experiences, for example your education and career, your present life and your hopes for the future. This knowledge, and your views about the treatment you received, will help us to improve the help that can be given to other girls and women with your condition.

Please will you do two things for us? Firstly complete some questionnaires, which would take you about an hour to do. Then I would like to meet you, either at your home or at the hospital, to hear your views.

The answers to the questionnaires and everything we discuss will be completely confidential and will be used only for the project. No one will know what you have said and the questionnaires will be given a number so that yours cannot be identified.

I have asked your doctor to give you a consent form as well as this letter. As soon as you feel sure that you would like to help us, please sign the consent form and send it in the envelope to me at The Hospital for Sick Children. I will then make contact with you to arrange when and where we can meet, and send you the questionnaires.

If you have any questions about the project, your doctor will explain things to you, or you can telephone me on 071-405-9200 extension 5596.

Thank you very much for taking the time to help us. I look forward to meeting you.

Yours sincerely

Mrs Brenda May
Clinical Psychologist
CONSENT FORM

Congenital adrenal hyperplasia and diabetes mellitus follow up study

I agree to participate in this follow up study as described by Dr and Mrs Brenda May on the understanding that any personal information will be treated in strict confidence and will not be released to any other person without my permission. I also understand that signing this consent form does not prevent me from withdrawing from the study if I wish.

Signature --------------------------- Date ---------------------

Address ---------------------------
---------------------------------
---------------------------------
---------------------------------

Tel ---------------------------
Appendix I (h) (ii) Example of consent form – Co-operating adult hospital

PATIENT VOLUNTEER'S CONSENT FORM

Consultant: ........................................ Investigator: Ms. Brenda May BA, Dip. Psych...

Purpose of the study and brief description of procedure to be carried out

We hope to find out how women with Diabetes are getting on in various areas of their lives now that they are adults. You will be asked to complete four short questionnaires which will take about twenty minutes to do. You will also be asked to agree to be interviewed at your home on such topics as your education, career, hobbies and social interests, attitudes towards marriage, and close relationships. You will also be asked about your visits to hospital now, and about your memories of attending as a child. Your views will enable us to improve the treatment given to others like yourself and to those children who are attending hospital with the same problem.

Everything you say will be treated in the strictest confidence and will not be released without your permission. The study will give group results, and your individual views will not be reported. Your treatment at the hospital will not be affected in any way.

This study has been explained to me and I understand:

(a) What the study involves
(b) That refusal to participate will not affect my treatment in any way
(c) That I may withdraw at any time

I therefore agree to take part in this study

Signature of Patient.................................. Date................................

I HAVE BEEN PRESENT WHILE THE PROCEDURE HAS BEEN EXPLAINED TO THE PATIENT AND I HAVE WITNESSED HIS/HER CONSENT TO TAKE PART

Signature of Witness.................................. Date.............................
(The Witness should be a person not connected with the study)

Full name and address of patient: ........................................
........................................................................
........................................................................
........................................................................
........................................................................
........................................................................

TEL NO.................................................
Dear

Some time ago, we contacted you about our attempts to find out how things were going for women with congenital adrenal hyperplasia.

I have now seen a number of women and have been given some very useful information by them about how things have been since they left Great Ormond Street. They have told me both good, and not so good, things about their careers, their relationships, their sex-life, how they get on at their visits to hospital, how their corrective surgery worked out, and their memories of what it was like as they were growing up - particularly as teenagers.

We need this information so that we can give the best advice and help to those young girls and their parents, who have the same problem as you did.

In order to make sure that I have got a true picture, I need to see more women, please may I see you? All the information I get will be grouped together so, although I need to hear about your experiences, the answers will be part of a group. Your views will be completely confidential.

Most people have chosen to meet at their home, but I have seen some at the hospital. The questionnaires have been taking about twenty minutes to do, and the discussions have lasted about one and a half to two hours.

Please send the consent form attached to this letter, back to me in the pre-paid envelope so that I can contact you to arrange a time and place to meet. If you would like to speak to me before returning the letter, please telephone me on extension 5596.

Thank you for taking the time to help us, and I look forward to meeting you.

Yours sincerely,

BRENDA HAY
Dear

We are often asked by the parents of young girls with congenital adrenal hyperplasia about the long-term aspects with regards to marriage etc, but there is still very little published information about this aspect of the condition. We have recently obtained funding from Action Research to carry out a follow-up study on girls with congenital adrenal hyperplasia who were seen at Great Ormond Street as children and I am writing to ask you whether she is still under your care, or if not, who is now looking after her.

Enclosed is a copy of the procedures involved in this research project and we hope you feel that we can contact her to ask for her help with our follow-up study.

While we would be pleased to make direct contact with her, perhaps you would prefer to speak to her yourself?

We would be extremely grateful for your help.

Yours sincerely

Dr D B Grant MD FRCP DCH
Consultant Paediatric Endocrinologist

Mrs Brenda May
Clinical Psychologist

enc.
APPENDIX II. DATA SUMMARY SHEET. HOSPITAL RECORDS
DATA SUMMARY SHEET.  HOSPITAL RECORDS (G.O.S.)

Subject No............
               d.o.b.  ..........  

1 NEO-NATAL
Birth weight ...........................................  Not Stated

2 SCHOOL PROGRESS:
Not stated

3 MENSTRAL FUNCTION:
Age at menarche.........................................  Not stated
Regular/irregular.....................................  Not stated

COMMENTS:

4 GROWTH

<table>
<thead>
<tr>
<th>7yrs(pc)</th>
<th>16yrs(pc)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Height</td>
<td>Not stated</td>
</tr>
<tr>
<td>Weight</td>
<td>Not stated</td>
</tr>
<tr>
<td>Mother's height</td>
<td>Not stated</td>
</tr>
<tr>
<td>Father's height</td>
<td>Not stated</td>
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5 DEVELOPMENT OF SEXUAL CHARACTERISTICS

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<tr>
<th>BREAST DEVELOPMENT</th>
<th>AGE</th>
<th>STAGE</th>
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<tr>
<td></td>
<td>.....</td>
<td>......</td>
</tr>
<tr>
<td>Pubic hair development</td>
<td>.....</td>
<td>......</td>
</tr>
</tbody>
</table>

6 FAMILY

Position in family.....................................  Not stated
Father's occupation.................................  Not stated
Mother’s occupation.......................... Not stated
Parent’s marital status......................... Not stated

7 CAH

Age at diagnosis............................... Not stated
S.L. or S.V. Not stated
Other family members with CAH Not stated

.............

Genital status at birth
Normal
Clitoris enlarged
Clitoris enlarged with some fusion of labia but separate urethra and vulva
Clitoris enlarged with single opening in perineum
Clitoris enlarged, urethra in male position

Comments Not stated

8 SURGERY

Initial Surgery Date Not stated
Clitoral recession
Clitoridectomy
Vulvoplasty
Vaginoplasty
Vaginal dilation

Non-related surgery/admission to hospital
Date Diagnosis Not stated

Comments
9 CAH PHYSICAL CHARACTERISTICS

<table>
<thead>
<tr>
<th>Characteristic</th>
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<td>Cushingoid</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Muscular/male habitus</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hirsute</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Other</td>
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<td></td>
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10 PSYCHOLOGICAL PROBLEMS

<table>
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<tr>
<th>Comments</th>
<th>Date</th>
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11 TREATMENT COMPLIANCE

<table>
<thead>
<tr>
<th>No. appointments made</th>
<th>No. DNA</th>
<th>% DNA</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
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<table>
<thead>
<tr>
<th>Comments</th>
</tr>
</thead>
</table>

12 GENERAL COMMENTS
APPENDIX III. SUBJECT CHARACTERISTICS

a) CAH interviewed group

b) CAH not interviewed group

c) Diabetic group
<table>
<thead>
<tr>
<th>Subject Number</th>
<th>Prader Classification at Birth</th>
<th>Salt Loser (SL) or Simple</th>
<th>Age at Diagnosis</th>
<th>Referred to Pediatric Physiological Services</th>
<th>Age at First Central Operation (Years)</th>
<th>Age at Interview (Years)</th>
<th>Marital Status</th>
<th>Fertility Status</th>
<th>Employment Status</th>
</tr>
</thead>
<tbody>
<tr>
<td>C01</td>
<td>3 SL Birth no 18</td>
<td></td>
<td>2 24.3</td>
<td>Single</td>
<td>Unknown</td>
<td>Housewife</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C03</td>
<td>4 SL 4/52 Birth no 3</td>
<td></td>
<td>3 37.1</td>
<td>Married</td>
<td>Treatment</td>
<td>Unemployed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C04</td>
<td>4 SL Birth yes 1</td>
<td></td>
<td>1 22.6</td>
<td>Single</td>
<td>Hysterectomy</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C07</td>
<td>4 SV Birth no 1</td>
<td></td>
<td>1 18.4</td>
<td>Single</td>
<td>Unknown</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C08</td>
<td>4 SL 2/52 Birth no 1</td>
<td></td>
<td>1 22.2</td>
<td>Single</td>
<td>Unknown</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C09</td>
<td>3 SL not known no 2</td>
<td></td>
<td>2 23.3</td>
<td>Single</td>
<td>Unknown</td>
<td>Unemployed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C10</td>
<td>4 SL 1/2 Birth yes 2</td>
<td></td>
<td>2 26.1</td>
<td>Single</td>
<td>Abortion</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C13</td>
<td>4 SL 1/2 Birth yes 1</td>
<td></td>
<td>1 24.9</td>
<td>Single</td>
<td>Unknown</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C15</td>
<td>4 SL 4/12 Birth yes 1</td>
<td></td>
<td>1 21.4</td>
<td>Single</td>
<td>Unknown</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C17</td>
<td>2 SL 1/52 Birth yes 1</td>
<td></td>
<td>1 19.3</td>
<td>Single</td>
<td>Unknown</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C28</td>
<td>4 SV Birth no 1</td>
<td></td>
<td>1 28.2</td>
<td>Married</td>
<td>Treatment</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C29</td>
<td>3 SL Birth yes 7</td>
<td></td>
<td>7 29.7</td>
<td>Married</td>
<td>Unknown</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C33</td>
<td>4 SV 1/52 Birth yes 2</td>
<td></td>
<td>2 25.8</td>
<td>Single</td>
<td>Unknown</td>
<td>Employed</td>
<td></td>
<td></td>
<td></td>
</tr>
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CAH/Diabetic group comparisons  
N.S.
APPENDIX IV. PROPOSED INTERVIEW TOPICS
APPENDIX IV. PROPOSED INTERVIEW TOPICS

1 Education
   type of school attended
   school leaving examinations taken
   after-school education
   qualifications achieved

2 Employment
   parents' employment
   own current employment
      level of satisfaction
      reasons for dissatisfaction
   career plans
   current income band
   if married/co-habiting, partner's income band

3 Chronic condition
   understanding of condition
   perceived effects of condition
   paediatric treatment
   current treatment
   current health status
   perception of the future as it relates to the condition

4 Psychological/behavioural problems
   alcohol consumption in past week
      reasons for drinking?
   cigarette consumption in past week
      reasons for smoking
   eating problems
   aggressive behaviour
   drug use
      reason for use
   clinical anxieties
5 Social activities

- sports
- hobbies.

6 Friendships

- purpose of friendship
- important attributes of friendship
- school friendships
- current friendships
- satisfaction with current friendships

7 Dating behaviours

- dates
  - age at commencement
  - parents attitudes
  - frequency of dates
  - number of partners
- behaviours/activities on dates
  - type of outings
  - pair/group activity
- patterns of dating for peers
- motivation
- current status of sexual partnerships
- past relationships
  - length of relationships
  - reasons for finishing/by whom

8 Sexual behaviours

- sources of sex education
- age at start of masturbation
appendix iv. proposed interview topics

self-perception of sexual interest levels
sexual stimuli
sexual experiences
problems during sexual experiences
motivations for sexual experiences
satisfaction with sexual experiences

9 Other relationships

with siblings
perception of parents' marriage
criteria for good marriage
choice of career/marriage
power strategies within relationships

10 Attitudes towards parenthood

knowledge of effects of condition on parenthood
reasons for wanting/not wanting children
effects of parenthood on life-style
perception of self as a parent
APPENDIX V. EXAMPLES OF INTERVIEW TRANSCRIPTS

a) Interview with woman with diabetes 281

b) Interview with woman with CAH 299
BM - Can we start off by me asking you to give a brief description of your education - what kind of secondary school did you go to?

D08 - Just an ordinary sort of secondary school in C (BM- a comprehensive school?) yes.

BM - How did you get on there academically?

D08 - absolutely brilliant, had a fantastic head mistress, she knew all about my diabetes, she used to keep boiled sweets in her drawer and a bottle of Lucozade, in case I ever had a hypo, if I ever was bad which was very very rarely, she used to just give me that, sit me in the first-aid room stay with me, not leave me, see how I was after 10 minutes if I was alright, sent me back.

BM - What a nice lady! What about school activities, you know?

D08 - I done the lot, done the lot, swimming, games and everything, I used to play for the school 5-a-side football team, and we used to do it against the Metropolitan Police, used to do that about 3 or 4 days a week, er....the school girls attitudes towards my diabetes was bad, (really?) yes, they were very very ignorant,(so what did they do then?) Say we'd been doing sport and I'd gone into a hypo, they used to think it was a joke, cos my mouth used to go, and nobody would ever help, they'd just leave me sitting there. I'd say, "I've got to get something to eat, go in my school-bag, and get the chocolate out - I'm ill" and they'd leave me! When my head-mistress sat down and explained to them they were as good as gold-

BM - So YOU hadn't explained to them?

D08 - I had, but they didn't believe me-they thought I was just sky-larking around.. I even done, when we were in the 2nd year we had to do a topic, so I done it all on the diabetics, insulin, how it was made, what made it what you had to do, I even sat in the class room and injected, the syringe into my leg, and believe it or not I got a very good grade for that, but of course as you go from year to year, you change classes, and you change school girls,(right) in the end I thought - well! !- I did have some very special school-friends and they understood and they were with me from the 1st year up till the 6th year,

BM - And if they were around they would know but the others wouldn't? ( D08 - yes that's it). And what about the other teachers, your headmistress was wonderful, but what about the others?

D08 - I can't say much about them, a lot of them, they weren't just interested, they knew they had a diabetic, I was the only diabetic throughout the whole school, and...um... they were very ignorant, I mean I'd be sitting there say one day I'd had sport and this other lesson and I'd just sit there and laugh, that's a sign, and I'd get a detention! it was only when I was slurring and I couldn't get the words out, I'd get dragged out of the class-room, and told I was mucking about, and I'd say "I'm having a hypo, I'm a diabetic, I've got to have something to eat, PLEASE let me have something to eat" they would say "don't muck me about"- nearly all my teachers were like that. They changed actually when a new girl came into the school, and she was diagnosed as a diabetic, as well, then their attitude changed, once they'd got another one, and they saw it for themselves, that it wasn't just me,.  

BM - Ummm. did you go on school outings and holidays?

D08 - no they wouldn't let me. Never let me go. We was due to go to France, just for the weekend, Friday to the Monday, I got all ready, filling in my passport, and my French teacher says I'm not taking you, your too much of a responsibility, I was in the 5th form then. I was really upset, I did cry, I sobbed my heart out, and I went to see the head-mistress, and I said your out of order, I'm normal, its only my pancreas that isn't working. I said, I look after myself food wise, I inject myself, and she
said, its up to them, my mum and dad came down to the school and kicked up a stink. So after that I never bothered asking, or filling in the forms or anything.

BM - What a shame....Day outings?

DOS - Yeh, those I was allowed out on ! on one condition that my mum put plenty of glucose in my bag, and a packed lunch....fine. But it was only local that we were going,(right) we didn't really go on much school outings, it was a good school, but they never really done much, things like that.

BM - So it didn't occur very often, that's a shame, .....and how did you get on with the school work?

DOS - fine, I didn't let that get me down.

BM - Did you do O levels and things?

DOS - I did CSEs, but I got the equivalent of Grade 1 which is the equivalent of an O Level and I got Child development, business studies, English, History, ....(Maths?) no, I got unclassified, maths has never never been my strong subject, even now I'm still, when I look at it, I think "Oh my God!" and that's it. But I find with my youngest brother if he's round here staying, I do sit and help him with his homework,

BM - O K  how old were you when you were diagnosed diabetic?

DOS - 6 and a half.

BM - Right - do you remember much about those early days?

DOS - Yep...I remember being diagnosed. Going up into hospital, cos we were living up in Bristol, I was really bad, Mum kept taking me back to the doctors, and they kept saying "you've got a spoiled little brat, all you've got is a child that wants a lot of attention, " Mum said "she keeps wetting the bed! She's losing weight like nobodies business- she's not herself! " Mum said "she keeps wetting the bed! She's losing weight like nobodies business- she's not herself! " it was "No. there's nothing wrong with her," It got to the stage where my weight went down to about 2 stone, and I was like a rake, and I was just weeing all the time, it was all I could do, drinking... in the end my mum just kept giving me sweets to shut me up, so in the end my mum said to my dad, "There's definitely something wrong with her, I'm taking her back to London, so they took me back to my old local doctors he took a urine sample, and said get her up to XXX hospital straight away, if you don't she's going to die. So they rushed me up to the hospital, and I remember going on 2 drips, in my arms, and them saying to mum she's diabetic, and then my granddad had a stroke, with the worry of it all.

BM - Is there anybody else in the family?

DOS - Yes, my cousin, cos my mum wasn't close to her, it didn't trigger, apparently they said I'd had it from birth, but it had taken all that time to get out. When I was in hospital, I remember I was sitting on the bed and the nurse saying you've got to do your injection now, and with a kid you're frightened you know, no-one had shown me how to do it, so I was sitting there, and I said where do I do it, and she said, in your leg. I remember this so clear, so I sat there and I got my calf muscle, and I stuck it in there......She shouted at me, so.... much... I jumped off the bed and I run into the bathroom and I cried, and I wouldn't come out, they phoned my mum and dad up, come down, your daughter's locked herself in the bathroom. Why?, cos we made her do her injection. Mum come down and I told her I'd done it in the back of my leg....... I didn't know ! I said the nurse shouted at me cos I done the injection in my leg, and my mum said, “Please come out.” I came out about an hour or so later, of course after my injection I'd gone straight into the bathroom, and I'd had nothing to eat, so my mum took the nurse aside, and said, “She's just been diagnosed, has anybody shown her?” "No." "Why" "You've got to learn the hard way" It got to a stage, I was in hospital for about 3 or 4 weeks, I wouldn't do my injection. I used to go into the bathroom, come tea-time-I'd run, into the
bathroom, and I can remember sitting there on the toilet, sitting there crying, saying "You're not going to do it," and the only reason I started doing it was cos my cousin and her husband came down and brought me a big yellow bear, that I've still got, and that one of the nurses bring me a syringe for Teddy, and I used to inject into Teddy and then into myself.

BM - That was good.....Did you have hypos as a child?

D08 - No...I didn't have none what so ever. When I had a hypo it was when I was 14 and I ended up in hospital. What I'd done was, I'd got up, and no-one had explained this, but what I'd done was I'd taken my evening insulin in the morning, so I thought I'd be alright, I could take my morning one in the evening, but it was a stronger dose, so the next thing I knew was I was lying on the bed, and some-one calling your tea's ready pet, and I come down stairs slowly, I had saliva coming out of my mouth, I was foaming, and I was sliding down the wall shouting "Go away- go away," My dad says
"Have you been smoking?" "No dad no," "Have you been taking drugs ?" "No dad, no" "She hasn't been out tonight ". So I see the carving knife, I've picked it up and I've lunged at him, and I love my dad to bits ! But it had completely taken over, I didn't know what I was doing, so I went for the carving knife! So my dad says "come on I'm taking her up to the hospital. She's gone mad !" So my dad's laid me in the back of the car, and he's driving, and I' sat up and I've wrapped my arm round his neck, and I'm choking him ! Dad's jumped all the lights, got pulled by the police, and my dad says "she's gone Do-dally, she's gone wrong in the head," Got me to the hospital, and the nurse says "taken the wrong dose of insulin,?! Give her sweets!" So they sat and gave me sugary tea, chocolate biscuits, I was heaving cos I don't like things like that, and they just made me eat, and eat, and eat. They said to my dad, "If that ever happens again, she might have a re-occurrence tomorrow, even if she's gone back to normal with her insulin, just give her chocolate."

BM - This when you were what, 14 ? So you had already had diabetes 8 years. It sounds as if your parents hadn't had very much explained to them?

D08 - No, neither had I ! All we were told was just measure the food, and how much to give me, I was allowed to have about 10 Smarties to a carbohydrate, unmmm 2 slithers of Kit-kat- 2 portions, no chocolate biscuits, and I was allowed to have a packet of crisps, and an ice-cream, thats all they explained, but they didn't explain if your daughter has a accident with her insulin, she's not on drugs, she's not a druggy, do this- calm her down and give her chocolate, you have to wait until its happened,

BM - Right, ..... and when you were in hospital having been diagnosed, they didn't make you have a hypo?

D08 - NO. No, I don't think they did. I remember having a hypo in the hospital cos I was crying cos I felt all funny. But I don't know whether that was intended or not, (Right) cos being in hospital away from your mum, my mum wasn't allowed to stay, so of course, I didn't want to eat, I wouldn't eat ! You know and that was it !

BM - So can you explain in your own words to me what diabetes is?

D08 - It's just somebody with, - they've got the pancreas in the back of the stomach and the pancreas has packed up, and the insulin isn't making sugar, for your body, so you need insulin, that your pancreas doesn't produce, other than that- no, I can't tell you.

BM - And now-a-days, erm...how much do you juggle about with it yourself, -how many injections of insulin are you on a day?

D08 - Twice a day.

BM - Do you vary those amounts yourself?
D08 - I don't touch 'em! I don't know how to touch 'em. I'll be truthful, I've got NO IDEA how to touch them.

BM - And food do you count in portions or 10 gms of carbohydrates?

D08 - portions. The old fashioned portions!

BM - And do you know how many you have for breakfast kind of thing?

D08 - erm, 3 for breakfast, 2 for mid-morning, 5 for lunch, 2 for mid-afternoon, 6 for tea, and 2 for supper. But I don't bother with the middle ones cos I'm just not hungry.

BM - And do you test your blood sugars?

D08 - Do my bloods when I'm feeling down, - a cold or something like that. As Dr X would tell you, in all the years the doctors never known my blood count to be bad. Because I do look after myself, OK I might have the odd pick of a bit of chocolate, or if the baby comes in and I look at it and say "no," but if he puts it in your mouth and says "please mummy, please" and I says to him "Mummy's not allowed to" and he says "you have a bit" and I think well! I haven't had my mid-morning, or my mid-afternoon snacks so this little piece will do, that's the way I look at it. (yes, yes) Like if I go out, I never touch alcohol, I might have a glass of wine, or once in a blue moon I might have a Bacardi and diet coke, but I'm not a drinker, (Is that because you're diabetic?) No, I don't like drink.

BM - And what about exercise? What does that do to you?

D08 - Nothing! I do a lot of exercise.

BM - When you were at school you were saying if you played games you might get a hypo?

D08 - Yes, when I was at school, I'd get a hypo, but now, nothing.

BM - Do you know what your latest HbA1C was?

D08 - That's when I was pregnant. No I don't. I know it was good, very very good. Cos I had toxaemia, and when I had him the doctor done it and said "Brilliant, its gone back to what it was. It's fantastic."

BM - And how was the pregnancy all the way through?

D08 - eerm Brilliant, apart from the fact that I had two mishaps and neither was my fault. I had some dodgy insulin, two lots, its was out of date. My ketones and my blood count, well my blood count I didn't even get a reading on it, it was just reading Zilch! and my ketones were...... cor!! I phoned Dr X at home, and X kept telling me to have more insulin, and it was going higher and higher, and then I phoned on the Sunday, and says, "Oh Dr X its still bad, and I'm worried about the baby, and X says take more insulin and if its not right tomorrow, come right into hospital, I phoned at 9 o'clock, and said "I'm bad." and my ketones are nearly 40, I said, and my blood count has just gone off the meter! "Straight in hospital" X said, and X saw the insulin was out of date, well you don't look do you ? I got some fresh insulin there and then, and by lunch time I was back down to normal. They put me straight on the monitors for the baby, I was 7 months, and then I had toxaemia. and the doctor said "IN" and I said "You've got to be joking. I'm not coming in here ! I don't do nothing at home anyway. " "For peace of mind just come in, and you can go home tomorrow!" But they kept me in, and they kept me in, and they kept me in until I said, "I've got to go home, I'm going round the bend, there's nothing to do here, cos the hospital was up London I never had any visitors, so I said, I'll make a deal with you, let me go home and I'll have a mid-wife round, you can't beat that can you ? The doctor said "You push a hard bargain " so I said if you don't I'm going to discharge myself cos I'm
bored. I said, “I know I've got to be careful, I know its dangerous, but you've got to let me go. So the
doctor said "fine, just you make sure you have the health visitor " and the health visitor came round
and said, "God, you're swollen, you've got toxaemia, and you've got to go in". “Leave it out, I've just
come out! They told me just as long as I've got my feet up, I just didn't budge.” He [husband] was
getting home from work and doing all the hoovering and the dusting. Then the midwife turned up, I
was just 8 months, and she knocked on the door, and C [husband] said "you're too late, she had it last
night. The baby just didn't want to wait, so she went in and had it last night”. I was in labour for God
knows how long, it started the night before but, last time, cos I'd already lost a baby, my mum just
told me I was constipated, so I didn't take much notice this time, I just sat there on the toilet, and it
was only when I got out the bath, I'd had a show, and the contractions were coming every 2 minutes,
that I thought this is definitely not constipation, I'm definitely in labour, (yes) and I went down there,
and I got down there at twenty to seven, and I didn't have him till ten minutes past one. With a lot of
complications. (BM - And the baby, how was the baby?) Lovely! He spent about 3 weeks in
intensive care. The doctor said he shouldn't have been in that long, he should have been in and out in
24 hours. and XX was very disappointed cos I wasn't allowed to hold him, I was told me and C wasn't
his mother and father, they were, nobody explained to me why he was in there, what was wrong with
him, why I couldn't have him back, and it was only in talking, C turned round and said, just in
talking, " wouldn't it be nice if we could take our boy home" and they put it down in the notes that C
was going to come down and it said, in writing, "Y's father will be down tomorrow, and regardless as
to whether Y has drips in or not, he will be taking him home. Regardless of the baby's condition". C
didn't really say that, he just wanted to have him home. He'd been there for 3 weeks ! None of us were
allowed to hold him. So I demanded to see Y's doctor, and he explained everything to me. If they had
done that before, I wouldn't have worried. But three weeks had passed, and no-one had said about his
blood count or anything. I thought, well because he's not premature, and he's not looking like a sack
of skin, why have they got him in there ! Let's not worry about the parents I The next one I'm not
having there!

BM - Right, that's a very sad story.............O K .....How old were you when you left school?

D08 - 17. I did a year in the 6th form. Originally I wanted to be a nursery nurse, but I didn't like the
thought of another 5 yrs at college,

BM - So you went into the 6th form to start the preliminary nursing course, but after 1 yr, you
chucked that up?

D08 - That's right. (BM - So what did you do?) I went into silver service ,waitress, a hotel up
London. (right) and I stayed there a couple of years. And then I went into shop manager, in S.
Kensington, and I had the actual run of the whole shop, the staff and everything. I stayed there for
years, I loved that, but then I got mugged, and that was it, I handed in my notice and I started getting
jobs down here. I went into being a secretary, and I done that right up until I left to have the baby.

BM - Of all those jobs you've done, which did you enjoy the best then?

D08 - ummmmmm My last firm. I worked for a demolition Co. My boss was fantastic, and I did love
being a secretary.

BM - Any problems with the diabetes when you were working?

D08 - Not while I was working, but I got turned down, (why?) cos you're a diabetic! So I said that's
discrimination, I said I'm normal, as you are, I'm healthier than you are, I have more check-ups than
what you've had, but I got turned down, and another thing I found disgusting, was I passed my
driving test no firm would insure me, I went to the A A , the Pearl, you name it I went to the top, all I
had was a Renault 5, the cheapest quote I had on that was £600, the dearest was just under £1000, in
desperation, we phoned the BDA -£27! I made 19 calls! I was crying. I said you are bang out of
order, they said I could have a hypo and kill some-one, I said YOU could have a heart attack and kill
someone, and I know when I'm having a hypo and can do something about it, you can't with a heart-

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attack. I said I was going to write to the papers, I did but they didn’t publish it. (so are you still driving?) Yes, I love it.

BM - What did your mother and father do? Did they work?

D08 - My dad’s a glazier, Mum was a hairdresser and she worked from home, but as we got older she went into being a night care assistant, So she was with us until 8.30.

BM - How many brothers and sisters do you have?

D08 - I’ve got 4 brothers. I’m the 2nd eldest.

BM - And none of them are diabetic? (D08 No) Now, I’m a bit confused about your marital status. Can you explain?

D08 - Yes. I got married when I was 22, er..., been courting him about 5 yrs, got married and he got a bit heavy handed, knocked me about, started taking all my wages from me. I stuck it for 2 yrs and then I walked out with the clothes on my back. Met C 9 months later, and it took me 9 months to go out with him, I was anti men - I hated men, and I’ve now been with C for 4 yrs in May,. We’re not married, we are getting engaged.

BM - Are you divorced from your husband?

D08 - YEAH. He was sorry and tried to make it up, but I didn’t want to.

BM - And C, what does he do?

D08 - He is in the carpets. Carpet lays, carpet fits,. When he was a boy, his dad bought him a paper stall, and he’s still got that, every Sunday. He’s a hard worker - a good provider. He knows all about my diabetes, what to do, he knows how to inject, he knows how to draw up,

BM - Right. So you’ve had experience of living with two fellows, - has the diabetes been a problem for either of them?

D08 - For my first one. My mum made him draw up the insulin, before I was allowed to marry him, but after that, nothing. With C he has to learn how to do it, do it in front of my mum, if I’m ill with flu and I haven’t got the energy, he does it for me, (and keeps an eye out) Yes. I can’t pick fault.

BM - Now I’ve got a list of salaries here, could you call out the number your husband falls into?

D08 - 4

BM - One or two more questions on the diabetes. Thinking about what you know about it now, do you think you know enough, or are there areas you’d like to know more about?

D08 - I’d like to know a lot more, a lot more.

BM - Can you think of any particular area, or is it a general feeling?

D08 - Just a general feeling. I’d like to know more.

BM - Right. Your son, he was obviously tested

D08 - Yes. He’s clear.
BM - Now, before you had him, did anyone sit down and discuss with you the chances of any child of yours being diabetic?

D08 - No, cos its only hereditary in girls. And X said if I had a girl there's a 90% chance it could be diabetic, or she could have other problems, my cousins got a little girl and she was born partially deaf, partially blind, her insides were on her outside and she had a hole in her heart. She got diagnosed as a diabetic at 2 and a half, and at 3 she died. She's got a healthy little boy, and she's got a little girl now, (BM. Did you know you were having a boy when you were pregnant?) Well you see, I've had quite a few miscarriages, like they were all early, but the one I had before Y - was about 4 and a half months, and when I lost that I said to C "They're girls, they got to be, cos God's saying to me -"S, you can't carry girls- there's got to be something wrong with them, I'll get rid of it ". When I had Y, when I first fell with him, I had no bleeding, I felt healthy, I felt fit, I had morning sickness, but I thought "Brilliant." Then by the time I got to 3 months I still had no bleeding and C said to me "That is a boy", and from 3 mos. we used to call him Y - C would get home from work, kiss my belly and say "Hello boy". I was going for regular scans and I went in there, and I said it was a boy. In the end they said "If that gets any longer, he's going to be popping out your mouth, and they started saying "your son". But I think it was because I kept on and on about it, talking about my son that they told me. Cos I had no problems about miscarriage through that pregnancy.I knew it was a boy.

BM - Right. they didn't do any tests particularly to find out?

D08 - No, Dr X did say did I want to, I went up to XX hospital for a scan on the baby's heart to see if he had a hole in the heart,

BM - O K. Now, ....if you weren't diabetic what difference would it make to your life?

D08 - None.....cos I do everything.

BM - O K and can I ask you about your visits to the diabetic clinic now, how do you feel about those visits?

D08 - They're alright when I go (laughs). If I feel down, I go, but if I don't feel down, I won't go ! DrX saw me when I was pregnant, she might not see me for another 4 years! I don't go, I know I should, I feel fine. (Right- ) I feel healthy, (So why don't you go) Why, cos its such a long wait when you get up there. I don't like sitting around, I don't like sitting indoors.

BM - So if you could be assured that when you got to the hospital you would be in and out?

D08 - I'd go. I'd go every 6 months like they ask me to.

BM - Give me a number between 1-5 for visits.

D08 - 3. I do enjoy it when I go, I enjoy seeing Dr X. I don't enjoy hanging around. If they said to me, S, your appointments at 12, cos they give you an appointment, and I'm still sitting there at 1.00 pm, and they say, " Well, I'm sorry but there's a backlog!" If I went in about 12 or 12.45 I'd go whenever.

BM - and how well do you think you are NOW, a number between 1-5?

D08 - Lovely. 5.

BM - How do you see the future, medically for diabetics, is it going to be the same or are there going to be changes?
D08 - I hope so, changes. What I've always hoped for they'd find a cure, and I'll be the first, be a guinea-pig. I know there are those worse off than what I am, and they're just as frightened of injections as I am, but I've always said to Dr X, when you get a cure, let me be the first. And then there was an article in the newspapers saying that a woman had had a pancreas operation, and it was going fine, I phoned up the hospital and asked how it was going, and they said it was like just a dummy-run, so I said is it on prescription or do you like have to pay for it, I said, "I've got the money here, let me have the operation". So they said it won't be for a couple of years. My mum and Dad have both got the money, C had turned round to me and said, "S. if a cure come round tomorrow, you go for it, he's amazing I think". That's the only thing I wanted as a little girl, I'd love to be a guinea-pig. I mean my eye-sight is fantastic, I've just started wearing contact lenses.

BM - I'm not sure I understand, is it because you want to be experimented on, or you'd do ANYTHING to get rid of the diabetes?

D08 - I want to be experimented on, I'd like to. Being a diabetic don't worry me,

BM - Right. Now you are on 2 injections a day, (D08 Yes) were you on more when you were pregnant?

D08 - Noooo. I was on 2, Dr X was very surprised. It was only when I had an up or down that Dr X put me on more. That was very rarely.

BM - Right. Change of subject. Now you've told me that you don't drink, but can I just ask you how much alcohol you have had in the past week?

D08 - None.

BM - Right. Do you smoke?

D08 - Yes. 2 or 3 a day. Which Dr X says is good compared to a lot, rather I didn't have any, as I put it to the doctor, I don't drink, I don't eat sweets, let me have one enjoyment.

BM - Yes. When do you have those?

D08 - ummmm, It varies, I never have three whole ones in an evening. I would be sick.

BM - I'm interested to know what is going on that makes you feel you would like a cigarette.

D08 - When the baby's in bed. I sit down, I have a cup of tea, and, I have a cigarette. I'm happy!

BM - Lovely.

BM - O K. Would you describe yourself as having a temper?

D08 - . Yes. (BM What do you do?...) Sulk. (BM Do you ever, throw things or hit things?) I used to, but since I've been with C I haven't, I think it was because of my ex-husband. I've calmed right down,

BM - You've calmed right down.

BM - Now, most people in their life go through stressful times. Have you ever felt you could do with some help for worries?
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D08 - No (BM - Not even when you were going through the bad times with your husband?) I just talked to my mum, or my dad. (BM...Or go to your GP?) No, I suffered a bit from the "Baby-blues", but it was alright I started going out again, and it just disappeared.

BM - O K. Another change of subject. You used to play sport at school. Do you play any now?

D08 - No, I do Keep-fit, once a week, I used to go to training-beds, once a week, but I had to stop because C was going to work earlier in the mornings, and then the car broke down, and I do keep-fit here (BM - how) at home. And I walk everywhere.

BM - Right. Now, - friendship. What is it that you look for in a friend? What is it that makes you say "they're a friend"?

D08 - They've got to be warm......with a special friend I like to check 'em out. for a few months, and I find out if they're talking about me,......willing to help out, and just somebody very very special.

BM - Are you the sort of person who would rush round to help others-(D08 Yes) Can you listen to others problems?

D08 - Yes, if they ask my advice I tell them and then I get it thrown back in my face. So nine cases out of ten I just say well do what you thinks best.

[BM presents friendship rating list]

D08 - Similar attitudes 2
    Doing things together 5
    Go to for help 5
    Share feelings with 5
    Share successes and failures 3

BM - Right, and if you have a worry or problem do you find it easy or difficult to talk to people about that worry or problem?

D08 - um, I normally mainly talk to C, or my Mum or my Dad, if I've got a problem I normally talk about it, it's no good bottling it up.

BM - But you would talk to your parents or C rather than a friend?

D08 - Yes. I'm very very close to C, and I'm very close to my Dad, so I'd find that it would be one or the other.

BM - O K. How satisfied are you with the group of friends you have at present....On a scale......?

D08 - (5)

BM - Right. And how many of those friends know about your diabetes?

D08 - All of them, they all understand it.(BM Because you've told them?) Yep. and once again, in front of their children as well, I draw up my insulin, inject, and I show them. I do it in restaurants, I sit there and do it in my arm, I can't do it in my tummy, but I do it in my arm, it would make me pass out, my dad done it once or twice in my stomach and I was out on my back. Dr X keeps saying its so easy, I say please don't make me do it, I don't mind my legs, I use my bottom, but please don't make me use my tummy, it makes me go cold

BM - Now moving from friends to relations, how do you get on with your brothers?
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DoS - Brilliant. My eldest one is very bitchy, my middle one's very intense, he doesn't express his feelings but I'm very close with him, and my baby brother I adore. He's 13 in June, but he's still my baby!

BM - Do they all live around?

DoS - My eldest brother's in the States, he emigrated over there, my G and my F still live with my mum, that's just down the road, see them regular, (BM so you're still very much part of that family) Oh YES.

BM - And your parents, how do you get on with them?

DoS - My mum and my step-dad I idolise. My dad and his girl-friend, I hate his girl-friend cos she broke up my parents marriage, (BM How old were you when that happened?) 23. They had just celebrated their 25th wedding anniversary, and Dad walked out on Christmas Day, and I hated her for it, but now, we've buried the hatchet, I can't say I'm head-over-heels with her, but my dad brought me up to be polite, but she can't do enough for me.

BM - So you've got your father and his girl-friend, and your mother and step-father, and those two couples. Do you see them both? Do they come here?

DoS - Yes, they do (BM It's quite a big family) You're not kidding. I've got brothers, and step-brother and step-sister, and step-nieces and nephews as well. I feel like "The Waltons"! To be truthful with you, my step-dads children, his son, I do call my brother, and his children I call niece and nephew, but his daughter I don't like, she's a stuck up snooty bitch, she sticks her nose up as if you're lower than her, and I can't bother with anything like that, cos we're all equal (BM Right) I love me step-dad to bits, I'm glad my mum met him.

BM - So, ....I don't quite know how to ask this question given the circumstances of what you've got,(both laugh) What I'm interested in is how you see you parents marriage when you were growing up?

DoS - Lovely. This is why I can't understand why my dad left my mum, I really can't. My dad used to work abroad, he was at home every week-end, money got tight and he did have to go abroad to work, cos he earn't good money. Dad used to come home and bring his little girl presents, and perfume and things like that, and I always see them as having a happy marriage - they very very rarely argued. Dad would be sitting there he might have had a hard day at work or something like that, and he'd just blow his rag ! But mum was very placid, quiet, and you know, she just used to put up with it, and then my dad walked out on us, (BM It was a massive surprise?) yes, and it broke my heart. It nearly killed me. My dad had gone, I mean I used to get home from work, you may think this is way out but, he'd be sitting on the sofa reading his paper, and I'd come in and move the paper out of the way and sit on his lap.....because he was my dad. He wasn't anybody else's....I used to go everywhere with him......I was proud of him! I could hold my head up and say "THIS is my dad, and I love him". and when he left my mum I just didn't want to know him any more, I hated him, he broke up my mum and my family. When I was pregnant. I didn't tell him, till he saw me walking down the road, when I was 6 months, and he wasn't a bit amused, and my mum said to him "She didn't want you to know, you hurt her " (BM That means you weren't seeing him during those months?) no. Since I've had Y-, going into hospital with toxemia, my dad phoned my mum and she said I was in hospital and got toxemia, and...er...he came down to see me with a curry. and that was it. (BM And the new relationship is...?) absolutely fantastic. I've got my dad back.

BM - And your father and K [stepmother] how do you see them as getting on?
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D08 - er... She rules his life! I don't understand how he's stuck with it cos he's never allowed my mum to rule his life, I mean, at all. She tells him "no" and and tells him"yes" and she tells him this and she tells him that, of course we only get this from my F. He says dad goes in the other bedroom and sulks, or, I say to my Dad do you want to come down for lunch, and he has to ask! has to ask her if its alright! And I think you shouldn't have to ask if you want to come down and see your daughter, you just get in your car and come down and see her! I've never known him to ask, if he wanted to do something - 5 yrs ago, he'd do it! regardless! Sometimes I get a bit annoyed as to how she talks to him, but I will never ever forgive her. I mean I accept my dad and C, but I don't know how I'd feel if they were to get married, but I've accepted them living together, but I don't think deep down, I'll ever forgive her.

BM - And your mother, you seem happier about that.

D08 - YES! she's got a good man, she's got a fantastic man,. She met him the year after my dad left, he's got a very very good job, he's a very important person, and he idolises my mum. He gives her everything. He's due to retire at the end of April and he's taking her to the States, for a month, giving her a cruise, and he absolutely idolises me, cos I call him dad, cos when I was pregnant, I used to say to T "Well come and say hello to your grandson" and I automatically made him feel, he was Y's granddad, and when I had Y and dad came down, I said to T. “come and say hello to your granddad, I said it in front of my dad”. Whereas with Y, a couple of weeks ago with C he called her Nanny and I said "That-is-not-your-nanny!" I said you can call her aunty, but you are not to call her nanny. He's only 15mos old and didn't know any different, but I made a point. While I've got a say, she will be known as "Aunty".but T.-he's "Granddad" and there's no qualms about it!.

BM - So you've had quite an experience of other people's marriages, so what do you think makes a good marriage.what is important in a good marriage?

D08 - Honesty. Love,. ...really honesty.

BM - In what way do you want your relationship with C to be different to these other two?

D08 - I don't want my relationship to be like either of them! (BM Tell me more.) C dominates me dad, which I don't agree with, I mean I believe a man should do what he wants to, if I said to C "Hoover" then I'd expect him to hoover, I wouldn't dominate him. I wouldn't rule his life, if he said to me "I'm going out tonight for a quick drink, just down the road" I'd say "O.K love, ta-ta love." With my mum she also dominates! I think my mother thinks she can dominate all of us- its only now that I've got to 27 that I've started standing on my own two feet and telling her what she can do with it , but she doesn't like it. T. gives her more than I think he should, because my mum's getting very lazy, T has always had an ironing lady, so now mum has an ironing lady, T. he's a fire-investigator, he'll get home from work and mum maybe hasn't bothered to cook dinner, the man has done a hard day's work and he's a hungry man, he gets home and she says, "I can't be bothered to cook", so for peace and quiet, he says we'll go out to dinner.And I think-no- cos when C comes home from work, he's had a hard day, he wants a dinner put in front of him. I'm at home, just like my mum is, O.K. I've got a baby, my mum hasn't, she's got the whole day to herself, she can do what she wants - I-BELIEVE- A -MAN-SHOULD-COME-IN-FROM-WORK-AND-HAVE-A-MEAL-PUT-IN-FRONT-OF-HIM-, alright, not straight away cos I'm busy, but at 7 o'clock when Y's in bed, we sit down and have our dinner together. O.K. if Y's been really bad, up with his teeth, I'm absolutely cream crackered! C will say "Oh we'll get a pizza, " but its not something I have every night of the week. I stayed determined, as when I was courting with him, I don't cook Saturdays, I cook Monday to Friday and Sunday, not Saturday, so when we moved in here, we continued and we had pizza, fish and chips, it doesn't cost a lot and we're happy. But every night he has a dinner put in front of him.

BM - O.K, I've got some things here that I want you to rate as important or not in a happy marriage...etc.

D08 - 5,5,3,3,5,5,4.
BM - Right. You worked until you were pregnant, do you hope to go back to work or not?

D08 - When they're at school, C doesn't want me to go back, cos he says the mother should be there for the child, you've got more chance of up to them going to school, of them not being well, its not right to palm them off to a child minder, (BM Do you agree with that or feel differently) I agree.

BM - So if I were to ask you to chose between having a career and staying at home to look after Y [child]?

D08 - stay at home. No qualms.

BM - I'm now going to go back for a moment to relationships, and ask you how you get your own way with people.

D08 - I sulk (laughs) (BM Is that with everybody?) Only those what are close, like my mum and my dad and C. cos I know they give in (laughs) but friends, no, I just go along with it, have done with it, (BM How long would you keep it up for?) (laughs) Days! till they gave in they always give in after about hour or so,

BM - And by sulking do you mean not talking, sitting in a particular way, or do you actually go out of the room?

D08 - No I just sit here and not talk, I sit here with a mag or a newspaper, and I go "tut", and C says "look at you" he always finishes up laughing his head off, I say to him "don't talk to me I don't want to know!",

BM - O K another change of subject now. Boy friends- how old were you when you had your first "boyfriend"? - you know what I mean by boy-friend. - someone you had a date with, not just a boy who was a friend.

D08 - 16.

BM - And was that a relationship where you went out just the pair of you, or was it part of a group?

D08 - No, I just went out with him, to the pictures or - wait a minute, I was older than that, I was 17, cos I was working at the hotel, 17, Yes, I was an old granny!

BM - Does that mean that your peer group, your friends, were they dating at the same time as you, before you or after?

D08 - Before, I wasn't that interested in boys. I wasn't interested, I didn't like 'em. I was just happy being with girl friends, out with my parents or....

BM - And how long did that first one last?

D08 - erm.....8 months, (BM And who finished it?) I did. (BM Because?) Too serious. he wanted a sexual relationship and I wasn't ready, I was proud of the fact that I was a virgin, and I wanted to keep it, and he couldn't accept it, so I said "Well – go".

BM - O K what else do we need to know? Oh, what happened then were there others?

D08 - I changed my job and I started courting the one I married.
BM - So this was your second boy friend (D08 Yes) And how long did you court him before you married him?

D08 - 4 and a half years. (BM Was he older, same age?) He was just under a year older than me.

BM - And did you tell me how long you were married?

D08 - Yes, 2 years we were married.

BM - Now, I'm going to get more intimate now and ask you about your sexual experience, cos we need to know how diabetes effects women's sexual response. (D08 Yes) Now, how old were you when you learnt about sex?

D08 - erm, My parents were always very open, I suppose I was about 8. It was from my parents just talking about periods and how babies were made and where they came from, that daddy done it, and how they got into mummy, and

BM - So did you have any formal sex education lessons at school?

D08 - No...no...no...I can't remember any. It was a catholic school.

BM - Now how old were you when you first masturbated?

D08 - 27! I just wanted to try it, so I did. (BM And how was it, was it good?) not really, I still didn't know what I was doing!! I just wanted to try it out of curiosity, I saw it on a blue movie and I thought "I'd like to try that" so I did and I thought "Oh, well" and that was it. I far rather fancy it with C !!

BM - Right. So when you got married, was that your first sexual experience?

D08 - Yes, I was 21 and a half and still a virgin. I was proud. I AM proud, I can turn round and say I've been with 2 blokes, one I married and one I'm living with here. Oh yes I'm proud of it, I walked down the aisle a virgin. I said to him, you want to marry me then you'll have to wait till we're married. I said I don't believe in that rubbish, you've got to be a virgin to walk down the aisle. And I said if you don't respect me for that, go. He respected me for it.

BM - Compared with other people that you know, or what you have in your mind, do you think you are less interested in sex, the same as everyone else, or more interested?

D08 - More!! I'm very horny, I'm terrible, a very high sex drive.

BM - O K. so how many times have you made love in the past week?

D08 - Well, I can't say this week, cos I got my bad belly, but last week, ..........about 14 times.

BM - Is that always at night when you go to bed, or do you sometimes like it during the day too?

D08 - During the day. when we're on holiday, or say I'm in the bath, and he comes in the bath with me, or he's in the shower, I mean I've always been so open, it was like that at home with mum and dad being undressed, and you just walk in and you don't bat an eyelid. If C was in the shower, I'd stick my head in and say, can I come in too, he say, "Yeah, go on then" I look forward to it, being in the shower with him, I think its cos I love him so much, I mean I'm just happy snuggling up, I mean, he's 6ft 4, and I just come above his belly button, all I do is I just stand in there and I just hug him, or I'm in the bath and I shout down "Are you coming in then" and he says, "oh go on then", and I say, "I'll go down by the taps!. Coo - its overflowing!
BM - This high level of sex interest, has it always been like this or is this a change?

D08 - Since I've been with him, and I've been with him 4 years, and its been like that.

BM - But how was it with your husband?

D08 - No, he was just "in and out"

BM - What turns you on?

D08 - Just him! (BM You said just now that you had been watching a dirty movie, do you like them?) No, it was just my cousin, she said she had one, and I said oh, let me have a look. But I think its just C, I look at him and I think " Ohhhhh, I want you!" It's just him, I mean, as you saw, he's not God's Gift, but he's MINE, and he's special (and you love him) Yeah.

BM - What about fantasies, do you have any fantasies?

D08 - No, oh no, I'm happy with him.

BM - And have you ever felt any sexy feelings towards a woman, (D08 No ) No crushes at school? (D08 No, none what-so-ever) Right. O K. Does making love effect your blood sugar?

D08 - I've had a hypo, (BM Yes? What, while you've been doing it? ) I don't know, my blood sugar went sky high, (BM You said sky high, do you mean high or low?) I don't know, I just went all funny, I just said "you've got to stop, I've got to go and get something to eat. and I come down here and just got hold of a bit of chocolate, and shoved it in, and I said, "I'm sorry, that's never happened.

BM - So you're not in the habit of taking extra, taking jam tarts up to bed with you or anything like that?

D08 - No. I mean, sometimes after we've made love, I wake up about three o'clock in the morning and feel, oh I've got to get something to eat, and I do come down and I get something a biscuit or whatever I can get my hands on and after that I go back up to bed again.

BM - And putting it the other way round, does your blood sugar level effect how sexy you feel? Do you understand what I mean, if your blood sugar is high does that make you feel, 'Oh I can't be bothered', or doesn't it make any difference?

D08 - Yes, actually I have found that, I mean even when I was pregnant with Y. I was having it, but, when I had problems I just couldn't be bothered, but when I had him, everything was back to normal, "Come here I want you"- it was just the month. I had toxaemia, and I felt run down, I wanted to, but I couldn't get off the pill! I said oh don't - just leave me alone.

BM - But in a way that makes me feel it was the toxaemia that was making you feel like that, (yes) You see, we have a bit of a problem don't we cos you don't do your blood sugars, so you don't know when , if its high, so . So just to re-cap on that then- sometimes you think you feel low afterwards in the middle of the night and there have been occasions when you've stopped to get some sugar.

D08 - That was when we were doing it lots and lots and lots of times! I think I was just using up so much energy

BM - O K now then, (list of sexual experiences presented.)

D08 - 1 y, 2 y,3 y, 4 n, 5 y, 6 y, 7 n, 8 y, 9 y, 10 n, 11 y, 12 y, 13 y, 14 y, 15 y, 17 y, 18 y, 19 y, 22 y, 25 y, I'll try anything for a game!
BM - O K. Orgasm, can you describe an orgasm?

D08 - It's lovely... (BM What happens when you orgasm?) I go all hot, and that feeling, you can't describe the feeling, its lovely, its not something that happens regular, I wish it could be all again, its a feeling all over,

BM - Was he experienced?

D08 - Yes, He'd had a lot of girl friends, compared to me, but, like he said, I was the only girl friend he'd ever taken back to his place, and spent the night with, and I was the only one that had ever done that to him, nobody else ever had,

BM - Have you had multiple orgasms?

D08 - We did though, together, it wasn't just me, it was him as well, he said it was as if he'd had an orgasm the same as me, he said he'd never had anything like that before,

BM - Was he inside you when this happened? (D08 Yes) Is he usually inside you when you orgasm (D08 Yes) Does he use his hand to stimulate your, do you know what you clitoris is (D08 Yes, yes) to stimulate your clitoris?

D08 - Regular. (BM Does that bring you to orgasm?) Yes it does, I say to him, "Don't stop" and things like that.

BM - O K. So you have an orgasm, are there occasions when he goes on, perhaps he hasn't come, or perhaps he has, and he goes on making love to you, can you come again?

D08 - Yes.

BM - Are they much the same?

D08 - The second times not normally as powerful, as the first, and being a diabetic I find it's very dry and very hard. (BM Right) and he has a hell of a lot of problems, an awful lot, I feel so sorry,

BM - Yes, .....What contraception do you use?

D08 - The birth pill. (BM So you don't use any jelly?) No, I think I should but I'm embarrassed to, I'm waiting for a free offer!

BM - Had he ever lost his erection?

D08 - About twice, but it was ages and ages he was trying. It didn't matter what he tried, he just couldn't get it going! He said, "Alright, we'll go and make a drink, come back, drink our tea, and try again." He didn't say to me "Its your fault" even though deep down I thought it's my fault, but he said "no its not, you're a diabetic, its one of those things"

BM - Does that mean you don't get very wet down there?

D08 - Yes. It upsets me. Its only to be expected, at first I didn't know what had happened, and then I thought is it because I've only had two blokes, and C said to me, in the "You and Me" magazine there was an article on diabetics, and they said they don't produce the lubricant, they are very dry. He'd read about it and he told me! I thought great, fantastic, at least I know there's nothing wrong with me, and he said, "If at first we don't succeed, we'll try and try again". 

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Appendix V(a) Transcript of interview – woman with diabetes

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BM - And is it ever painful?

D08 - Only when its dry. Just after the baby, just after I stopped bleeding, it was terrible, we laid off for a little while, about three weeks

BM - How long after the baby did you have sex again?

D08 - About 4 weeks. and there again, I only had a caesarean, it wasn't as if I'd had a normal birth...

BM - A caesarean?

D08 - Yes, an emergency caesarean, I nearly lost him, they said to C "She's got a hell of a lot of stitching, she's really stitched up badly". C said to me, are you sure, it's only a month after, and I was saying, “Please I want to”. I was really frightened, I said "Please don't hurt me" but then it was alright - except for this nigging pain just there, but we laid off for 2 or 3 weeks cos I started bleeding all over again, and then it was as good as gold!

BM - Right. ....Do you prefer to make love in the dark or the light?

D08 - Anywhere. (BM It doesn't matter) No.

BM - Why do you have sex?

D08 - Why? Cos I want to, cos I need him, cos I love him,

BM - So for you its a combination of expressing your love, but also cos you feel you want him

D08 - YES.

BM - Have you ever done it when you didn't want to, just to please him?

D08 - No. I don't mind cuddling up to him, but I said to him, could be cos I'm tired or had a heavy day with Y or otherwise the same as with him, sometimes he doesn't, he says "It's not cos I don't love you, come here and we'll cuddle." I'll just snuggle up to him.

BM - Lovely. So all in all then, the whole package, I want to know how satisfied you are at this time in your life, with your sexual experience.

D08 - VERY.

BM - From 1 to 5?

D08 - How about 1 to 10!!!- 11 and a half!!

BM - Right. The next bit is about parenthood, which you are already. Why did you want a baby?

D08 - I've always wanted children. I wanted one with C, we had a good steady loving relationship, we had a secure home, and alright I wasn't married, I'd have rather have been married, but Y came along a bit unplanned, I thought C would make a lovely dad, we did discuss it, it wasn't something I just said "I want!", er......, we talked about it, and decided that this one, cos I'd fallen pregnant, we were going to keep him.

BM - So your miscarriages before, were they unplanned?

D08 - Every one, the birth pill! I was on the pill every time I got pregnant.
BM - What change has it made to you as a person, to your life, having a baby?

D08 - Me, I'm still as mad as ever, if not madder, but I've felt I've become more warm, more caring, I mean I'm very warm and caring to C and to my parents, since I've had Y, I've just found love I never thought I had. He gets smothered in it. He gets away with blue murder, I have completely changed, I won't have people telling me what to do, when to do it, and how to do it, I now find that I'm 27, I've got a baby, my own home, if I've got something to say I'll say it. It has changed me. My dad said, now, I've grown up, even C says I've changed. He couldn't put his finger on it, but he says I'm completely different. whereas before I used to be short tempered, and I'd have ago, but now, I just take a pinch of salt, let things as it happens,

BM - That sounds very positive change, what about the negative things it's brought to you?

D08 - I don't think it has! I can't find having a baby a bad fault!

BM - And will there be another?

D08 - YES, we want to go about the end of August, we'll start trying then. C was anti, cos of having such a bad birth with Y, fine in the pregnancy, but the birth was so bad, and its only just now, he's just started to come around, and agree to another.

BM - Did they explain to you at the hospital why it was so difficult?

D08 - They didn't say nothing to me, I didn't know nothing- I had an epidural, it was C that was seeing everything, I mean before I had the epidural they was sticking all these tubes up me which was painful and everything, and I was crying, and they stuck me on a drip cos they said I wasn't contracting enough and they lost the baby's heart-beat and his pulse-rate -it made me bad, C was shouting to take the f.ing thing out and ummmm in the end they took the drip out then they decided to give me the epidural...I remember it but not very good, I remember having my legs over one doctors shoulder and my other one over C's and I remember them trying to force this tube up me and I was screaming and they gave me the epidural and everything was alright and they found that my placenta had stopped feeding Y. ...............BUT THEY LET ME GO. I can't understand it, from twenty to seven till four minutes passed one. without giving me a caesarean earlier, to stop all the hassle , and then they turned round and said they wasn't sure if my cord was stuck around Y's throat, and he was very distressed and he wasn't pushing, erm and things like that. That put C off. I though it was a big joke didn't I! I mean I was lying there and saying "What's taking you so bleeding long !" wasn't I, "Come on, aint he out yet? " They said "If you lay still and shut up, we'll do it just as quick, " But I was just taking the Mickey out of them, but one thing that they didn't explain to me and they should have done this- when I had the epidural I was laying on the bed and I'm shaking! head to toe! C said "What's wrong with her- what the hell's wrong with her !" "Tie her up". So they tied my legs my arms, and C said "Don't tie her up, what's wrong?" " Oh it's just the epidural" But they didn't say to C, be prepared, she'll shake. He thought I was having a fit! So C thought I was going into one, and he's flown out to ring my mum up, and tell her they are doing an emergency caesarean, and by the time C got back he's missed the birth, and when we had the baby, I said to the nurses "What ever you do, you take my boy out and put him in his dad's arms" She went out and said "You're mother-in-law's got more experience, she can hold him" and ..she..gave..him..straight..to..my..mother! Then later to have Y's first bottle, C was just sitting there, looking at our son, counting his fingers, and looking at him, and they come and took the baby out of C's hands and gave him to my mum and said "You feed him" C said to me "I could have cried!"..........................

BM - I was just looking to see if I have missed anything out. Have you ever had any eating problems?

D08 - Only when my Grandmother died. I stopped eating then (BM How long?) for about two months, until my parents threatened to get Dr.X to me.I used to wrap it up in tissue. (BM Wrap what
up in tissue?) My food. My Nanny died and I felt I'd lost part of my life. I just couldn't eat- the sight
of food! [the baby starts to cry, and BM speaks to him] He doesn't like men, he's alright with
women though.

BM - What about his father, does he play with him?

D08 - Yes, a lot, he idolises him...and his Granddad. But it upset my brothers when Y didn't like
them...he went off of them actually, about 5 months. ago. He starts crying now when they touch him.
It's so embarrassing

BM - One more question and then I'll go home and leave you in peace.

D08 - That's alright, it's been an interesting afternoon actually, I've enjoyed myself.

BM - Is there anything we haven't talked about that you think would be helpful?

D08 - No we've talked about everything I think

BM - And has anything we've talked about upset you?

D08 - No, nothing.
BM - Can we start by thinking about your schooldays? What kind of secondary school did you go to?

C41 - mixed comprehensive. I left at just 16 yrs.

BM - How was it for you academically, How did you get on with the school work?

C41 - Not very well. I couldn't seem to grasp a lot of the writing side, I was good at art, I could do anything with my hands, but when it came down to actually concentrating or anything like that I couldn't seem to grasp it all in.

BM - Did you go to hospital a lot in those days?

C41 - Yes, due to the fact that I was going through adolescence, I think I was about 11 when I started periods, and then they were trying to get my hormone balance working out - I was fainting a lot as well, I wasn't too good, I wasn't too well. I was going in for a week or so at a time.

BM - How many times in a term would you go into hospital?

C41 - It depended how my blood tests came out, sometimes I used to have to attend every week, to O/P to take more blood, and other times it would be a month or 6 weeks or then 3 months or 6 months, according to how the balance was going. I worked out, it's about 7 years of my life I have spent in hospital. At one point when I was younger, I used to go home at weekends and then I couldn't wait to get back home- which was hospital, that was more home for me than it was at home! I missed out on my Art O level cos I was at hospital at the time. I didn't get to take my GCSEs, I just got, I just scraped through with a D you know. I didn't even get a grade for maths I just put my name at the top of the paper. I used to have to go to special maths things cos I couldn't get the grasp of it even then. Reading, it has to be a dam good book but no problems in learning to read.

BM - So, you seem to be feeling that you had the ability if only you hadn't had the absences, is that true?

C41 - I don't really know, I just found it very difficult to grasp things and keep it in my mind. I'd read them and read them and read them and I still couldn't get the grasp of them - couldn't be bothered. My brother, now he's got a photographic memory. He was doing so well he could have gone to university, but he blew it, he just opted out. My brother was the pedestal, I was the "Oh well you'll get a job", but I wanted to go to Art college but my mother couldn't afford to send me so it was "get a job".

BM - Tell me about friends at school?

C41 - er, I had one good friend, she used to live next door to me, she went to the same school, but even then it was an effort to do anything with, you know she'd have to come round and say we are going out, and sometimes I'd just sit up stairs and say -I don't want to go-. I wasn't ill, I just - it was an effort to get conversations and mix. She was a year older than me.

BM - So in class?

C41 - I didn't really mix a lot, I didn't have friends that I went out with at night or anything like that. And I don't now. I don't make an effort to go.

BM - What did you do after school?
C41 - Hairdressing, and I done it for 10 years, I went in as a trainee, and moved a few places, salons. When I was 16 - my father had died anyway when I was 13, that blew my brain up quite considerably - I went off the rails. He dropped dead in front of me that didn't help. I had an awful lot of problems with my mum, she wanted to put me in a home cos I was a bit rebellious, she found me very hard going, I used to go out at 13 and get drunk, go in a coma for 3 days with drink and sometimes with people and sometimes on my own. And then I'd get picked up by the police and my mum, she couldn't cope with it. I was just a bitch to her, looking back on it I was a complete bitch to her. She had a problem with my dad dying anyway and I wasn't helping. She was on Valium tablets and she went a bit wacky as well, I felt just not wanted, and that freaked me out quite a bit. My dad dropped dead in the bedroom like, and she could see his face with all these angels going round - I think it was stress but that freaked me out. My brother had shortly left home as well, he's 4 yrs older than me, which left me carrying it all.

BM - How did your mother take it about your illness?

C41 - My mother and me, we don't really hit it off very well anyway, I was a bit of a daddy's girl. My mother was all for my brother, you know- my son this and my son that. She cared a lot. I'm not saying she didn't, with my illness she made sure I took my tablets and go regularly to the hospital, she did all that but she never asked me how I felt. “You should be lucky you are alive you know, you should be lucky that you are having all this attention”, but no one asked me how I felt. I felt awful, really bad, I felt a freak!

BM - What are your memories of going to the hospital clinics when you were a child?

C41 - Ever since I sussed it all out I used to hate going to the doctor. I used to fight, kick when I used to go- I hated the fact that when I used to go, I don't know what they do now, but they used to put you on the table naked, and I was deformed a bit, I had pubic hair and things, and you were put on this table and there used to be, thousands of people sitting around, to me when I was a kid, and then they used to make you stand naked to weigh you, and that freaks kids out. I noticed when I was at school taking showers, I had more than what most of the girls did. I had pubic hair or I'd got breasts and others were flat chested and hadn't even started periods at about 13 or something. And it doesn't do you any good, I was born with pubic hair apparently. I used to hate going to the hospital, I used to be a right cow. It got a bit better cos when I was 16 they transferred me to X hospital. There was a bit more privacy there and they didn't really bother taking all your clothes off. They weren't really interested in looking at my body.

BM - Do you remember the operations?

C41 - Yes. I was about 5. Apparently I had an enlarged clitoris and they done some surgery to either make a vagina or... I think they had to make the vaginal canal. I used to have a wooden thing pushed into me to keep it open, and I was always pulling it out. And I was in pain, but they just had to insert it again, I have very clear memories, very much so.

BM - What was your understanding of why you were going to hospital?

C41 - To make me better. That's what I was told. I didn't know what was wrong with me! I didn't know if I was any different or what, but I must have been cos I was going there to make me better.

BM - Your mother, did she talk to you about why you were going to hospital?

C41 - She just said it was to make me better, and I had to do the things that they said, have another operation because it would make me better like.

BM - Was there any general discussion in the family, could you ask questions?

BM - How did your mother take it about your illness?

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BM - Was there any general discussion in the family, could you ask questions?
Appendix V(b) Transcript of interview – woman with CAH
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C41 - They just said it was one of them things. When I was born I was born at home, and then I was taken into hospital at 10 days old due to the fact that my foot was twisted round the other way, and while I was there they - the mid-wife had told my mother that a lot of babies are born with pubic hair, it was all right. But when I was in hospital they thought it wasn't right and they started doing some tests. I only just found out and it freaked me out, that I had been put down as a boy when I was born, and I never knew that. And I've got another birth certificate. I found out less than a year ago. I feel completely freaked about that.

BM - You got all your hairdressing qualifications?

C41 - No, I didn't take them. I was going to college and then I moved, but I still did hairdressing. At the end of the day I was quite capable.

BM - Is that the only job?

C41 - No, that was the only one until I got married and moved up here. I couldn't get a hairdressing job cos there wasn't much work, so I had to get something so I went into a pottery place, where I was a fettler and sponger of all things. You fettle off the seam when the pot comes out of the mould, I didn't enjoy it but it was a job. Then I got pregnant and now I'm a residential social worker. I've been doing that for 6 and a half years now. I saw an advertisement, - working with difficult behavioural children - and I thought that'll do me, I was a right little bitch, and I get on great with the kids. It's the staff I can't cope with! Some of them are great like, but I think some of the staff are going in there to sort their own problems out like, its one of those jobs where there's a lot of back-biting and back-stabbing, but I love it. Part-time now, I can't get the child minders to look after the kids. I did City and Guilds in Community Care and Social studies, you have to do it for work anyway so, and my tutor was impressed so she said you ought to go on. I'm generally thinking of it when the kids are grown up I'd like to go back full-time, but I'd like to go more into Section 53, I was thinking of doing, starting off with a GCSE in Psychology, see what that's like and then progress to A level and work my way up. They've got courses that you can go to in the day, they've got crèches now, a couple of days a week I would like to do the Access course, but that's a full-time course, at the end of the day, I can't leave the kids, and I can't leave my job, I need that for the money, I need the independence and it gets me out of the house. And I enjoy it. I've got it in me its just getting me motivated. My work is still male dominated and I tend to mouth off a lot as well. I've got this habit of making these mega-statements I think they get frightened of me to be quite honest, I know at work they do, we had one boss, I was always questioning things, so I was put down on his little list, you know, he wanted me out like,

BM - How old are the kids?

C41 - Seven and two and a half now. Both boys.

BM - Please would you tell me which of these salary groupings you fall into?

C41 - Less than £5000. its about £10,000 if you are F/T. I was doing it F/T but I couldn't cope when my husband left, he deserted me when I was 3 months pregnant with the second one. He went off with another woman. He used to work for the M.of.Def. as a clerical officer.

BM - How are the boys, were you at all concerned about them having the CAH?

C41 - No problems with the CAH, don't know if they had been girls though.

BM - What is CAH?

C41 - Besides the physical abnormalities, as far as I know the adrenal glands pump out hormones, and with any hormones there's different enzymes etc, and they produce the poison and er, another
enzyme that counteracts the poison and all I produce is just poison. So the steroids I take is to put back into the body what I don't produce. That's what I understand what it is.

BM - Where did you learn that?

C41 - I asked, at adult hospital, the doctors. When I was at GOS I had a lot of people coming and asking me, students, cos it was quite a rare case, but I couldn't explain it to them. The doctors didn't really say anything, more so my mother, they talked to my mother, but she said "They weren't very informative with me, so I was left in the dark". So she couldn't tell me cos she didn't know what was going on. I think it was a case where not a lot was known and they couldn't tell cos they didn't know themselves, it was trial and error in themselves and I was used as a guinea-pig. They asked my mum if I could be used as a guinea-pig, I used to take these different drugs, I used to blow up - I mean I was continuously on a diet when I was younger, I still am, I just put on weight, I find it very difficult to get the weight down. They told me up here that it wasn't the steroids, I said it was but........

BM - Are there things you, anything else you would like to know?

C41 - Er,.....Well, I think I know generally the idea, I don't understand the medical terms too much, and as long as I know the basics. When you are born with it, you are born with partly the opposite sex, and you have to have surgery nine times out of ten. I don't know WHY we were born like that, genes that's the only thing I don't know is WHY. Why is my brother O K and I'm not. My mother was advised to have a sterilisation after me cos they didn't know what kind of baby would be born to her. You feel a right freak, I blamed my mother for a lot of it, at one point, I blamed her a lot, not my father. It was like our general relationship - you know cos she used to say, "Oh you are a right cow, a right bitch!" and I used to say, "Well, I didn't want to live. No one asked ME, YOU wanted me alive, so if you want to blame somebody blame yourself, I didn't ask to be born, I didn't want to survive". She shut up then. That's what it's been like.

BM - Any other memories of childhood visits?

C41 - Some of the tests I didn't enjoy. The examinations, I didn't like them! It's was like being physically abused, sexually abused, but it's permitted. I mean all good and well, they had to do it, but, if that was somebody that had abused me when I was a child, they would have been done for it. It's the same feeling to a child, and its something that's been allowed to be done to them, I didn't like that at all.

BM - So how were the hospital visits on a scale from 1 to 5?

C41 - I can't do that cos they were the bad things, but I did enjoy being in hospital it was like my home. I liked it in there but I didn't like the things that happened to me. When I went up for days [to O/P] they didn't always examine me, so it's very hard to put it all together like. Somedays I hated it and some days I loved it cos I used to get a present from my mum as well.

BM - How would you like to see things changed for children going to O/P today?

C41 - I hope they don't have to go through what I went through, they don't have to cos medical science has developed so much that within my area of illness I KNOW they won't have to go through what I went through. They may still have to do the internal examinations, but I've been told that they wait to do the ops until they are older, which I think is a bit dodgy, which I would probably been more freaked out with, than when I was 5 or 6. A better age is as early as possible. When you get to 5 or 6 you are beginning to be aware of it all, I feel sorry if they do it even later.

BM - So you're saying - I think its better for kids now, but also it's worse.
C41 - I think it's better cos they know a lot more about this now, the medical side they know more about it, they don't have to pump kids up with different steroids to see what happens, they don't have to put them on endless diets, they don't have to experiment now, they don't have to be weighed and measured hundreds of people gaping at your body, there is a bit more feeling - not a lump of meat. I understand that they have to be examined cos it's one of those illnesses, they are going to have to go through that, but they won't have to go through - "Wow! we've kept her alive! That's amazing, not many have been able to be kept alive!" I felt that's what happened to me. That's what I've got in my head.

BM - How did your parents cope?

C41 - It was a very very big strain for them. At the time they hadn't really had a success, to live, of one of us [with CAH]. Is it all right to say one of us, now I know there are other people. Mother, I think she must have been really upset especially having been told - "we don't know what the next baby's going to be like you had better be sterilised". That wasn't very nice for her cos she wanted a large family. She was the main one that coped with everything. My father was the one still having to go to work, he though a lot of me, but my mum is the type that takes over. She was the one that was taking me to hospital all the time, and they told her that I probably wouldn't survive until I was three. I could die any time, and I'd be a midget and I was pretty much a freak at the end of the day with everything, there wasn't anything that was going for me. I've proved 'em all wrong really. I did survive, O K I haven't grown since I was eleven, but mum's pretty pleased. It was a lot to do with her getting medication down me at the proper time. If I threw up or that, she'd have to work out how much I'd thrown up in that and give me what she thought was . . . even if I had to have a tooth out, I had to go into hospital to have it out. Where as now, I just have them out round the corner and no problem at all. So she had a lot to put up with you know.

I didn't like her talking about my problems to anyone else I hated my mother telling anyone what was wrong with me. I knew there was something wrong and when I found out I'd had an operation I hated her talking to anyone about my problem. Relations knew, but if she happened to mention my problem like, I hated that cos it was none of their business.

BM - And yet she didn't talk to you about your problem?

C41 - Not a lot, No.....

BM - How much did your brother know about it?

C41 - I think he knew. I think he got very jealous when I was born cos I was spending a lot of time in hospital and my mum was there, and when I went home, we didn't seem to get on. There wasn't a relationship, there, there wasn't with mum and dad really, I didn't feel part of the family. I felt more secure in hospital. The doctor used to pick me up and walk round the wards with him, and I used to spend Christmas there and home was hospital.

BM - How is it going to hospital now?

C41 - Can't really be bothered to be quite honest. I go in and they say - "How are you feeling" and I say - "I've not been feeling too good", - or - "I've been feeling pretty knackered like, - I'm getting aches and pains more frequently", my bones seem to be - ..... I get back pains. They probably feel I'm blooming mental! And they say "Oh you seem alright, your HBs alright, see you in so-and-so". I feel they can't really be bothered. It's pretty boring I find, you wait more than you are in there. I've seen so many doctors, I hardly ever see the doctor I'm supposed to be under, I always see one of his underlings. Half the time you have to re-cap to them what's the matter with you. And I think - what the bloody hell am I doing here? If they don't know, and they are supposed to be treating me, I might as well go home and start taking my tablets and ..... they don't seem to be interested. You might get an interested one and the next 6 months, he's left. I said once that I like a doctor and the one that I
was seeing said: "Well, he's gone to Derby, you can go there if you like him"! - That's not an attitude to take is it? If you are dealing all right with things that's all that matters to them.

BM - How do you see the future medically?

C41 - Nothing they can't do anything for me. They've just got me stable and that's it til I drop dead. Cos I'll never come off them, I've asked them.

BM - Do you ever forget to take them?

C41 - Yes. and I feel pretty grotty. I know something's gone wrong the next morning. I start getting headaches, and feeling really grumpy, and then I take one and I seem to have a shock like and feel I need another one, I sleep a lot.

BM - So what difference has it made to you, how would your life be different if you didn't have CAH?

C41 - Well, now even now, I don't like using the word "normal" but if I didn't have it I'd be normal. Just as anyone else.

BM - How much alcohol have you had in the past week?

C41 - I used to drink a lot, but in the past week I've had a hell of a lot. Eight port and lemonades, plus lagers, I go out once every three weeks, that's when my husband has the kids otherwise I don't go out at all. Don't drink at home. If there was an opportunity of going out more, I would drink more. It's a social thing. In the past I used to drink to buck me up, but not now. If it was in the house I would, but that's why I don't get it in the house, cos I could quite easily go through a lot.

BM - How many cigs in a day?

C41 - 20, I smoke more if I'm worried.

BM - Would you describe yourself as having a temper?

C41 - Very much so. I've got more irritable lately. I'd start shouting and bellowing. I get physical too. I'd attack people if they had annoyed me. I've walked out of rooms before now when people have got up my nose, rather than get physical. It got to a real physical fight with my husband. If I'd had the strength I'd have done him in a couple of times, and I nearly did. He didn't seem to listen, always, I used to try and talk and it was as if I didn't exist, once I had a wooden mallet and I hit him on the knee with that he quickly went into the back bedroom and I was literally hammering away at the door, -and another occasion, I don't remember much about these times, but I seem to gain so much strength he is quite a big guy, I got him on the floor, and I had this bayonet and I just about to stab the hell out of him he had to hit me to get me off, you know. But he had to hit me hard, he had to stop me and he hit me on the side of the head, he was panicking, he couldn't get me off, I was ...he couldn't get me off him. I'm beginning to hit the kids, I'm seeing a psychologist about it. My oldest son looks so much like him and he infuriates me, he's very much like him, I try and ignore him, but he will give that little...just like my husband did, and then I just blow. My oldest son, he's got problems anyway, behavioural problems and problems with the split up of the marriage, because he never used to be like it, he was such a placid little thing. The school referred him to the psychologist, he's calmed down but he tends to get a bit up when he's been to see his dad. I've got bruises on my legs where he's kicked me, I have to physically restrain him, and I feel bad when I've hit him, really bad, I give him a whack round the bum if he's naughty. The counselling has made me not feel so guilty about it being my fault, but I still feel a lot guilty. He's more daddy's boy.

BM - Do you do any exercise, any sport?
C41 - No, not now. I was very very sporty. A lot of cross country running, in the district for throwing and javelin, all the teams, hockey, netball, rounders, basketball, I loved sport. I think you go through a spasm when you are a teenager that it's not the hip thing to do, it was more interesting going to discos or getting boys. Then I started to do a lot of badminton, and I used to do a lot with my husband and now- nothing now. I'd like to but its getting people to look after the kids.

I've been doing a lot of live role-playing, done it for years, that's where I actually met my husband, I still enjoy doing it. I'm a nutter. We started in a castle in Chester, you go there and dress up and act out your fantasy killing monsters. I've got a big gathering coming up this weekend 5/6000 role-players from all Europe - we'll meet in Derby, I could take the kids cos they've got crèche facilities, but I'm not taking them, but we're going as a unit, there are about 15 of my mates go. That is my escaping, that's the way I can forget everything. It's another world. I can get into that world very easily. A lot of people think I'm mad and its Satanism and that sort of rubbish but they just don't understand its very sort of people willing to mouth off before they know the facts. We have got a group up here that we do every two weeks as well, well, my husband and I started up one years ago here, and then he left, so I go to that as well. We go to the local woods and act out, everyone thinks we're nuts but it's a good laugh. I enjoy it.

BM - What is a friend?

C41 - A friend to me is someone I can actually be myself with. I've got no fear of them belittling me, they understand they listen to me. They understand me, and I can talk to them, about, not all that's wrong with me, not a lot of people know that. If I do I just mention the side of the tablets and the enzymes, I won't ever, ever mention the physical side. My mother was told I might have hair growing as a beard but touch wood, I haven't had any of those problems yet.

BM - (presents list of friendship attributes)

C41 - similar attitudes- 4
        do things together-5
        help each other-5
        share feelings-5
        share successes and failures.-4. But I'd only, I'd stop, I'd only tell them what I want them to know.

BM - How easy is it for you to share your feelings with friends?

C41 - Bloody hard, people would describe me as not very emotional, not very sympathetic, and with their problems as well, my attitude would be "yeh, yeh, is that all".

BM - If you have a problem, can you share?

C41 - No. I let down my barriers when my husband left cos I was told I was too hard. My mother had a go at me, she said I never showed him any loving feelings or emotions, and I never used to talk to my husband about anything. He did ask me what my problem was, and I said "I've coped with it for the last 30 years and what's it to you to worry about!" And I never told him about anything. So he never knew anything about the CAH, and then when he did leave I started to think, and my mum said you ought to you're so hard, you are hard to everyone. I never looked at myself, you don't do you..... I thought about it and thought, O K I'll give it a go, and when I let my barriers down I got shot on so..... they went back up again. I let them down to my husband and he still walked out. It didn't make a blind bit of difference. I won't try again. When my barriers were down it was the first time I ever told my mum that I loved her in 33 years! She cried. But I can't talk to my mother more now, not really, it's still very hard.
BM - So am I right in thinking that there aren't any people in your life at the moment that you have this kind of true friendship with?

C41 - One. He's male. I've known him for ten or eleven years. I met him the same time as my husband, and he's very much like me. In our feelings, our attitudes, well we are both Virgo and I think that might have something to do with it. If we say well do something we'll do it, we won't dilly-dally - my husband would lie, but we mean what we say. He went through a bad time with a young lady and as I was going through with G and he knows G very well, and the feelings and emotions were talked through. He knows a lot more about me than anyone else does. We go out occasionally, oh there's nothing in it cos he's just a friend, but when I go out socially, that's who I go with and we meet other people that are acquaintances.

BM - Is that enough for you or would you like more?

C41 - I could live without that. I can live with just acquaintances, I don't have to be dependant on people, it doesn't worry me. I feel sorry for people who have to have somebody no matter ...like my friend down the road got rid of her husband but then she soon got somebody else. And it's gone on from there, as soon as that one's gone she's got to have someone else. It's security she feels she needs that, whereas I don't need that. Cos I feel "So what. I'm secure enough. I'm independent, always have been, always been on my own for a long time. In the end my mum got re-married so...I don't need it.

BM - How did your mother and father get on?

C41 - She was always nagging him, poor sod. He used to have to go down the garden to get out of the way like, she was always right, always dominating, he was a big softy anyway, he wanted a quiet life, hard working never saw a lot of him. But my mother was the main one that dealt with the crises, if there were any laws to be dealt out she was there. Nine out of ten times, I used to get the wallops, not my brother, he was a bit of a mummy's boy really.

BM - So was it a good marriage?

C41 - No. They stuck together until he died, about 20 years or something, but in them days it wasn't the thing, you couldn't get an easy divorce could you? And I think people had to work out their differences a lot more than they do today. I don't think their marriage was brilliant. My Auntie J, my dad's sister, she actually told me that my mother buggered off, when my brother was little, with somebody else, and my dad had to go and fetch her, and there was somebody else floating around like that when we used to live down in London, that she was supposedly knocking off. But mother won't tell me things like that, I mean hers was the PERFECT marriage.

BM - What makes a good marriage?

C41 - erm. Well now that's been blown out of my mind, I couldn't say - after what's happened to me. I look at people and their marriage is just as bad, I thought our marriage was all right. I mean I used to blow my top and nag him cos he was that type of person but you had to motivate him, but I think for a good marriage its trust, there's that trust that's there, you've got to work at it, I mean no ones marriage is perfect, if the other person is not willing to give, then forget it! You might as well call it a day. No, I don't want new relationships, that's out of the window. I'm quite happy as I am. My husband we tried to get back together, he said the reasons were that I never talked to him...thats when I let my barriers down and everyone couldn't believe this change, we were getting on so well, he wanted another baby, I didn't, then I said O K we are settling down better, she was gone, tried to make a go of it, and then he buggered off. That was it! Even after I knew he was still lying I said to him what's the point of lying? My husband does argue, he will just sit there, I'm the one who's running round the house shouting my mouth off, calling him all the names under the sun and he would just sit there...and that used to infuriate me! I could feel him turning off, and I wanted him to know how I felt, and he wouldn't listen. And then I used to get violent, I used to hit him, to get
noticed if anything - just a glance at me - bloody look at me! I could have been a piece of shit on the carpet for all he cared. I used to loose my rag and hit him and he used to say "you are mad!" Now if anybody hurts me, I get wild, more so than what I used to be, with my son. My mum says I used to go round hitting kids when I was little, she says I was a right little bitch. I don't remember much in the teen years. Now, I think I've just lost my cool.

**BM** - (presents list of attributes of a good marriage)

**C41** - saying nice things-4  
doing thing-4  
good sex-4  
have interests in common-5  
no money problems-3  
partner is a confident-4  
problem solving- you have got to talk about it. 4.

**BM** - How do you get your own way?

**C41** - Just do it yourself. At the end of the day it's easier to do your own thing than make the hassle, I've never got my own way. I feel I've never got my own way, my husband always used to get his own way, I used to give in. Now, the only way is to ignore everybody else and go and do it. I don't stamp my feet I don't throw things about, I can't put on an act. I'm me, if they don't like it they can go and find someone else, bugger off.

**BM** - How old were you when you had your first boy friend?

**C41** - About 14. Mostly we used to go in a group at that age, we used to have a gang like.

**BM** - Was this the usual age for your group of friends, the usual age?

**C41** - Basically I should say it was average.

**BM** - What were your parents' attitudes towards you having boy friends?

**C41** - Mum didn't like it. She always used to say I used to get the wrong BF [boyfriends], you know. She didn't like them. I didn't have a lot of BF's really, Then at 16 I met D, that I went on to be with for 8 yrs. She didn't like him! I wanted to live with him, but my mum was— she was, I was frightened of my mum actually. To be quite honest I was shit scared of her. Erm...and I couldn't do it, it was alright for my step-sister to go and live with theirs, when they were younger than ME like, and for my brother to do everything under the sun, but she wouldn't allow it with me, you know, she had this bloody moral that she had to stand by. I don't know why she was like that with me, I have no idea. Even now she still tells me how to run my life. When I split with G, "Oh well, we knew it wouldn't last! He was no good to you" and I was feeling really depressed and my mum was going "Well, you know what he's like a real bastard like" I used to think God, I want support, but she wouldn't bother to. My mother-in-law did, she knew more of what was going on than anyone cos, you know, cos I was there and she was upset. She was there when I wanted her. I think a lot of her.

**BM** - So eight years this steady lasted, why did that finish?

**C41** - Me. I thought we was getting into a rut. I don't know really, I felt I wanted to move on, he was a bit of a hippy, we were at the time, he was more into skimping and scrapping jobs, do a job and leave it, and I ended up bloody supporting him. We did a lot, we spent a couple of months abroad rucksacking it, but it just got to the stage where I felt if I didn't leave we'd carry on like it, I mean years is a bloody long time, I think I was 25 when I finished.
When I slept with him I thought "Well he ain't said nothing about me being abnormal, cos he must have had somebody else cos he was a couple of years older than me, and we had a great sex life. Very enjoyable.

BM - Was this a worry for you?

C41 - Yes, that a man would find me not right. That I was different, I didn't know what any woman would look like, cos you don't go and look at people's private parts do you. I was shitting it, but he never said nothing, he was quite happy and I was and off and we went! and had a damn good sex life.

BM - What was your mother's attitude to you going abroad?

C41 - Well she my brother had left home and then I was still at school and she met this guy, well he used to muck her about a bit like, looking back she was like a big school girl, nine out of ten times I used to come home she was never there anyway she was at his house looking after his two kids, she used to phone me up and say she was stopping round there, and one night she phoned up and said "Oh, got some news for you, we are getting married, but don't worry, I've found a place for you to live, and I'll contribute cos you're earning - I was only on about £6 a week," So she got married and she went and sold the maisonette, and I had to get out and she put me in these lodgings, and supported me which she had to in a way, and they lived at his house. So I never saw her for quite a while. I think she was a bit of a shit! As I say we never really got on. We are too much alike with tempers I think. She likes to dominate everything, you've GOT to do what she says. She throws a load of guilt at you, by saying "Well, when I'm dead and buried at least I've got a clear conscience- I never did this to my mother!" and make you feel bloody guilty. Then I move over to HW to be nearer my BF I was living in bed-sit land, and got myself a job and I was off. I lived in a house with a band and it was brilliant.

BM - What happened then, BF wise?

C41 - I had a few, nothing serious like and then I met G. It was less than a year until we married. Now, D we had our ups and downs like, but he wanted me for me, he liked me for what I was, he didn't mind me being a bit over-weight, I'm not the most beautiful woman in the world, he like me. G always said "Oh you're fat". When you first met him he'd give you the world, anything you did, anything you said, he'd agree with, and then of course there was no challenge. You got bored. I actually spoke to this woman he's living with and I called her all the names under the sun, but she's younger than me and she's big, bigger than me. And then he was going "Oh you're too thin". Nothing was ever right, and I don't think anything would have been right, so it was a waste of time trying! What ever you do is wrong, that's what I felt like at the end. But with D I could talk to him we had a better relationship, he knew about the hospitals, I never told him about the physical side, he understood, we used to have quite a few punch-ups like, I was quite sad I broke that one off. He's married now with three kids. It was a damn good time, yes. And now when I go out with my mates, I can have a damn good time, and got myself a job and I was off. I lived in a house with a band and it was brilliant.

BM - I want to turn now to something I feel you have been expecting all afternoon, your sexual experiences. (C41 Yes). Firstly, where did you learn about sex?

C41 - Er, we had one lesson at school I think, and that was about contraception, the basics was books. Medically type books, I used to go to the old library, and experimentation,

BM - How old were you when you first masturbated?

C41 - I wouldn't say I'd masturbated myself it was more a case of, I never got any enjoyment out of it, I was trying to feel what they'd done to me. I don't lie in bed masturbating myself, it's not really a
Appendix V(b) Transcript of interview – woman with CAH
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turn on for me. But I used to put my fingers inside my vagina and say "Well, what they done? Is this supposed to be this low, am I supposed to have this there?" And I used to look at myself in the mirror and think is this normal, is this supposed to be there? Where have they stitched? But I can't honestly say I've masturbated myself for enjoyment. No. It was more finding out about my body and what have they done! I'm still no better off, cos I don't know what any other woman looks like! Even now, I've never thought about it for enjoyment.

BM - What turns you on?

C41 - Basically the foreplay of it all, with a man. Females don't turn me on at all. Males definitely, and they have got to know what turns me on, so I tell them. I like to be fingered, and I like to be licked out, for a long time, I find that more enjoyable, than actually penetration. Not a lot of feeling in my clitoris, I can't actually say there is a real "ohhhh" some women say, nothing like that, it has to be inside, I don't really get a lot of pleasure out of that. I don't know if it's working. There's a lack of sensation there. And feeling my breast and that, its more that [gets me going], I like '69' so I get a lot of enjoyment out of that sucking somebody. So, it's not necessarily penetration all the time. When I used to play with my clitoris, I can't really feel anything, I used to like bizarre sex, my husband thought I was a bit weird, I like to try a number of different positions, different areas, different things, with D it was more experimental. With my husband it wasn't what I was used to, so I used to tend to train him to what I liked, sex toys, not really dressing up, teddy and suspenders and things, I used to like to dress up for him, I enjoyed that as well, baby oil and things, in the open, it was better where people were near, we was all for that, D was the first, I was about 16 then.

BM - Do you get wet?

C41 - Yes, but I dry up very quickly. When I'm turned on, I really do get wet, but I've got to be licked out to get it moving. It takes a bit of a time to get down to it.

BM - Any pain?

C41 - I feel my womb's dropped cos I did get pain after having the kids, but before -no. It seems to be hitting something there, doggy-side, where you get more penetration I get more pain.

BM - Has your partner ever had any penetration problems?

C41 - At the very beginning, I was so tensed up anyway, but no. There does seem to be a block now, not long enough like. I went to see about my womb but they said no it seems O K. but I used to check the coil and I could feel it and it seemed too low.

I've come clean with you, cos I've psyched myself up if it will help others, I said to myself I'll do it, its the first time I've ever spoken about these things to anybody.

BM - Presents list of sexual experiences?

C41 - 5 1 2 3 6 7 8 9 10 11 12 13 14 15 16 17 18 19 20 22 23 25

BM - You mentioned orgasm there, can you describe what it feels like?

C41 - I don't really get a lot of sensation, I get a lot of water. I thought the first time I was probably weeing myself, but I can't seem to get what other people say they experience, the crashing down and the feeling in your stomach, I never have seem to have got that. So I get a hell of a lot of water coming out, it's not wee. I feel I can't stop it coming out, it rushes out, I can't control it, it happens when...different things like, I could be wearing a teddy or suspenders, I could be quite turned on anyway, when I get turned on, the hole where my clitoris was gets hard. Fingerling or licking out is another one. I've got to have foreplay. I think that penetration is the bit that finished it off, I like to
get into the body before that. I like to linger on with the foreplay. I like penetration, but I get more pleasure from the first bit.

BM - When it's over, how do you feel?

C41 - Shattered. If they come quickly, I'd like more, I don't get the gushing with penetration, but I often feel I'd like it go on. I get dead wet and then I want to carry on and on. They usually come before I'm satisfied really.

BM - So all in all, how satisfied with your sex life are you?

C41 - 5 when I was going out together with my husband

BM - Now?

C41 - Each day is different, I could go and get it but I don't want to, like that.

BM - Did you plan the children?

C41 - No. They told me that I would probably never have children, with D I hardly ever used contraception except Durex and I never fell. It was a job to get my periods on a regular basis and I had to go on the pill for a bit, and that didn't agree with me at all, and the periods seemed to settle themselves out a bit better. I've never been really spot on. When I went out with G I must have got pregnant on my wedding night! We had sex before we got married used contraception sometimes, condoms or he withdrew,

BM - How was the pregnancy?

C41 - The first one, I started to loose blood and he wasn't growing, at 7 months I lost blood and went into hospital for a month, and then they induced me. He was about 6.12. I had a normal delivery but they cut me bad. I was going to ordinary antenatal clinic, and I saw a consultant who deals with a lot of unusual cases as it were, my CAH consultant didn't have a lot to do with it.

Then I had the coil fitted. But about then my husband started going a bit funny and he used to sleep the back bedroom anyway. And then when he wanted it, I used to say piss off, anyway. I don't know if he was jealous of me being pregnant or not, he used to spend time in the back bedroom when I was pregnant. There was a 4 year gap, and then I fell for J. that was because I took myself off the coil cos I was having very bad periods, and the way I felt as well, I was having more infections, and lost a lot of weight. So they took it out to be on the safe side as I'd had it in 4 years, but we did use Durex with them both,

BM - How did you feel about becoming pregnant?

C41 - With the first, no, I didn't want him, I didn't want him at all, I was absolutely shocked. I mean I went to the hospital for a check up, and he said "Well we'll test you then to make sure", and I nearly died! He said well I can't do anything about it, you'll have to see your local GP and I went and said to my husband, do you want it, and he said-we'll toss a coin - and it landed on- 'we have him'. But I wish we hadn't in a lot of ways cos we didn't know each other. I think that drew us apart, my husband needs a lot of attention. If you can't give my husband attention he ups and goes somewhere else. That happened. He reckoned I wasn't there listening when he wanted me, he's like a big baby anyway. He couldn't cope with it. The second one we were just getting back together like, we had started a divorce and he wanted to come back and then I got pregnant, I didn't plan it, and I thought we were getting on marvellously well, but he ......one day I came home,cos I was working full time and he'd given up his job 'cos he'd practically got the sack from being down there all the time like, with her, so he had to resign. He was looking after E which he didn't do a very good job with, when I
came home one night, the house was practically deserted, bits and bobs were missing it was about 11 30 at night. The wardrobes were cleared out and he'd gone. I didn't know where the hell he was and I phoned my mother-in-laws and said have you got E and she went "yeah". “Thank God” I said, “He's pissed off”. I never heard from him for about 3 months, my mother-in-law put out a police thing - it wasn't difficult to work out where he was, and he phoned up one day and said what’s the meaning of getting the bloody pigs round my door. The police only went looking for him cos he'd deserted me, and I was pregnant. So I could send all the divorce bits down to him. I'm still going through the divorce, he's been a complete arse-hole. He wanted to come back before J was born - I was in two minds and I said “Well Christmas we'll get together, and he said- “I don't want to!” He seemed to be flipping and flopping you know. I said that's it, no more. He'd got another child by my step-sister, about 14 she was, and he's never really bothered with him...It screwed my brain up considerably. I let down my barriers and that's was it, I got trodden on. But I still want revenge. It’s three years now, I found it tempting but not now! He's hurt me and he's hurt my kids, (the younger one doesn't even know who he is, they go and stay with him) take both of them, they are yours! A adores him, he doesn't seem to understand what he's done, he doesn't want to know, he's still his dad, and A blames me that his dad left, cos I was arguing with him.

The second pregnancy was a normal delivery too, it was a week over-due, the first one I had about 20 stitches, which I though wrecked my fanny, and the second one, I was on my own like, and they were fantastic, and I said for Christ's sake don't cut me like she did, and she said I don't think we need to but she didn't know how much elasticity I was going to have because of the CAH but she gave me a little tiny nick like that and got him out like a pea anyway, and I only had three stitches. But I had a thing about my fanny, they used to say "Oh Mrs X and her fanny!" I said for Christ's sake don't stitch me up too little! I felt good 'cos no one mentioned about my illness, I mean sexual things is a big thing in life isn't it, and when some-ones got something wrong with them.....I think now adays the young people and their parents, I don't know what goes on but I think these kids do need a psychiatrist or psychologist and I think the parents need some counselling as well. I think a lot should be told to them, cos they'll end up screwed up like me. A lot of it stems from the stigma that is put on you ....once I went with A and there was a Health Visitor round the corner and she was filling out the things and she noticed that there were some tests going on for this CAH and she says-has your son got it?- and I says no, its me, and she goes says my brothers got that, and I said well you're the first person in 30 years that I have ever, ever come across, and I hate talking about it to people, and I phoned him up and had one conversation and it was GREAT to relate and I said "I was made to feel a……" and we both said "freak!" It was great cos we had the same feelings. No one knew, you couldn't talk to anyone, you were a freak, what was wrong with you no one could tell you,

BM - What kind of a parent are you?

C41 - I could be more loving, I find it difficult to...I can kiss my little one but I find it hard with my eldest one. I know it sounds selfish but I wish I didn't have him at all. And I could do what I want to do. I don't like being in my situation but I have to put up with it. I shout at them and then I give in anyway, I tend to mouth a lot but I don't seem to do any actions like.

BM - Are there any positive feelings about having children?

C41 - Well they are me, I didn't think I'd ever have them, and I was shitting it thinking they might have what I've got, which I would never have forgiven myself, if they had got it. But thank God they haven't got it. They are hard work. I prefer older kids to younger kids.

BM - Were you ever offered any genetic counselling?

C41 - No, nobody ever mentioned it, they knew I was CAH. I've never heard of it. I just had these worries about the babies, on my own. When my blood went absolute chaotic they didn't know what the bloody hell to do. I know the second baby was tested, but I'm not even sure that the first was, I have noticed that the first has a very small penis, I've taken him to the doctors about that cos I got a bit neurotic about that, the hospital reckon he's fine, its just that he's fat, but I don't want him to be

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ridiculed at school. I tried to breast feed the first, but he was always hungry after six weeks so I stuck a bottle in his mouth, the second one was bottle fed anyway.

There have got to be people there knowing about our people, that can talk to us. Its too late now for me, but I want to see changes, that people aren't going to be shit scared, that these young girls in this day and age they should not be scared of having sex - that's a load of crap, there have got to be trained people.

**BM** – Thank you so much for talking so openly to me. I wonder if anything we have talked about has in any way upset you, or confused you. If so please tell me and we can try and sort things out?

**C41** - No, mostly I was right, I expected most of the things, and I have to be truthful and say that once I got going I found it great to let it all out.
APPENDIX VI. EXAMPLE OF PROCESSES USED IN THE QUALITATIVE ANALYSIS OF THE DATA

a) Outline of procedures used in the qualitative analysis of the data 314

b) Example of initial classification of the raw interview data under separate headings. 315
APPENDIX VI.(a) OUTLINE OF PROCEDURES USED IN THE QUALITATIVE ANALYSIS OF THE DATA

All grounded theory procedures are aimed at the identification, development and relations between concepts. The following is a description of the operations by which data was broken down, conceptualized and put back together again in new ways so as to create an understanding of the meanings of experiences for women with a chronic condition.

The transcripts of the interviews (the data), based on similarities and differences, were separated into discrete items or concepts, these were descriptively named and formed the focal points of the analysis. Concepts may be further composed of lower order components, dimensions that in turn may be composed of a number of elements. Concepts may be related to others to form categories.

These operations are not performed in a step-wise order but are applied flexibly according to the emergence of relationships, questions and checks at any particular time point.
## Appendix VI (b) Example of initial classification of raw data. Page 1 of 2

<table>
<thead>
<tr>
<th>No.</th>
<th>PERCEPTION OF ACADEMIC PROGRESS</th>
<th>ATTITUDE</th>
<th>EX CIRCUMSTANCES</th>
<th>ABSENCES</th>
<th>ACADEMIC PROBLEMS</th>
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</thead>
<tbody>
<tr>
<td>08</td>
<td>I didn't do a lot of work, I didn't like working.</td>
<td>I had a very back attitude, I expected everything to come to me. It was too easy. I wanted to do what I wanted to do.</td>
<td></td>
<td>God had an all. I couldn't get my head round it.</td>
<td></td>
</tr>
<tr>
<td>09</td>
<td>I wasn't that good academically, I struggled.</td>
<td>I couldn't be bothered. I didn't want to go to college so I thought why waste each other's time?</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>I found it very hard to cop at school, it was all too much. I found it better to be taught at home. I wish I could start school again, I've missed out on quite a lot of things.</td>
<td>I was in a calab of hospital, so I didn't go often, and not being well.</td>
<td></td>
<td>I still have to match the words. Need write them with the read directions.</td>
<td></td>
</tr>
<tr>
<td>13</td>
<td>I didn't do particularly well at school.</td>
<td>Some of the teachers just didn't see if it was the individuality. I don't learn well by teachers talking, better by discussion.</td>
<td>I was never pressured by my dad, he didn't insist I stayed on to do As.</td>
<td>I was reasonably well attended.</td>
<td>None - I learnt to read before I went to school.</td>
</tr>
<tr>
<td>No</td>
<td>PERCEPTION OF ACADEMIC PROGRESS</td>
<td>ATTITUDE</td>
<td>EX.CIRCUMSTANCES</td>
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<tr>
<td>001</td>
<td>Academically I did rather well.</td>
<td>I enjoyed my classes.</td>
<td></td>
<td></td>
<td>I struggled a bit in maths.</td>
</tr>
<tr>
<td>002</td>
<td>I liked small groups, it made it a lot easier.</td>
<td></td>
<td></td>
<td>None.</td>
<td></td>
</tr>
<tr>
<td>004</td>
<td>fairly well I think.</td>
<td></td>
<td></td>
<td>I missed 6c cos I was in hospital.</td>
<td></td>
</tr>
<tr>
<td>008</td>
<td>Fine - I ended up failing that (school work) got me down</td>
<td></td>
<td>well, maths was never my strong point but</td>
<td>No difficulties there at all.</td>
<td></td>
</tr>
<tr>
<td>009</td>
<td>I wouldn't say the school was highly academic, but it was nice. I was in the middle of the road.</td>
<td>I wish I'd concentrated more.</td>
<td></td>
<td>I had quite a lot of time off.</td>
<td>None.</td>
</tr>
<tr>
<td>014</td>
<td>These seemed to be 3 groups: those who didn't care, those who wanted to get on, us in the middle.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>018</td>
<td>I suppose at school I got in with the wrong crowd.</td>
<td>I didn't really like school much, I couldn't wait to get out, I hate my exams a get a job.</td>
<td></td>
<td>I had a few problems with maths, but nothing that couldn't be sorted out. I found it difficult to understand, got my GCSEs.</td>
<td></td>
</tr>
<tr>
<td>021</td>
<td>I did quite well through the GCSE exams.</td>
<td></td>
<td></td>
<td>I got to go down a bit - I was nervous, my mind goes all blurry I can't remember anything.</td>
<td></td>
</tr>
</tbody>
</table>
APPENDIX VII. THE CLASSIFICATION OF THE DESCRIPTIONS OF ORGASM
PLEASE READ EACH OF THE NUMBERED STATEMENTS WHICH FOLLOW AND THEN CLASSIFY EACH ONE BY CIRCLING BELOW THE STATEMENT THE ALTERNATIVE WHICH YOU CONSIDER MOST APPROPRIATE.

THE ALTERNATIVES AT THE END OF EACH STATEMENT ARE CODED AS FOLLOWS:—

A = Sexual arousal
O = Orgasm or Climax
DK = Don’t know. (Please try to avoid using this)

THANK YOU FOR YOUR HELP

Please return to Ms. Brenda May, in the stamped addressed envelope.
Appendix VII Classification of the description of orgasm. Page 2 of 6

1. I would say a contraction of the muscles of the vagina- it's nice whatever happens, it's nice! and it doesn't happen immediately, it happens after a while, and once it's happened it goes up and down again, so it's not one long one and then stop, it's like-pulsing, wonderful word. They say the vagina isn't at all sensitive, it's the clitoris. I don't really believe that that's 100% true- I would say it's 70-80% the clitoris and the other plays a part. It's stimulation to both parts in me...what's the point in having the bloke do what he does if it's not important?

A\O/DK

2. I don't know, we both get so excited when we come like. He has to take precautions at the moment cos we don't want anything to happen like. We just get so excited. I don't feel anything inside like,

A\O/DK

3. That's difficult, it's not always the same. It's good, it makes me shake all over. It starts as a little tingle in the clitoris and then it becomes uncontrollable clenches of the muscles, that's it I think.

A\O/DK

4. I'm not too sure, I don't know. I've got myself into such a state but I don't know what it's like. I think I have once. I get that feeling that something's going to happen but nothing does.

A\O/DK

5. It's more of a feeling I think, it's like waves, yes waves, it's hard to explain- more of a feeling, physically and mentally. I suppose the height of excitement. He was actually penetrating me. We time it together actually, we are very into talking actually when we have it, we say "are you ready" "Yes" and then we both go, so we can get it together.

A\O/DK

6. I get the shakes, I get very hot inside.

A\O/DK

7. A nice feeling, he was inside me. Feel it all over really, it lasts a few minutes. After I feel drained out, don't want to keep on sort of thing.

A\O/DK
Appendix VII Classification of the description of orgasm. Page 3 of 6

8. It's a form of extreme excitement. I find orgasm is extreme excitement. The sensation within you is a form of tingling and at the same time a form of pain—can't really say it's pain, but it's enjoyable. I love it, can't get enough of it. It starts within the tract and as time goes on the whole of the body begins to feel the same sensation. I suppose it's the same kind of thing as if you stood on the floor and it vibrates, it slowly becomes...going up your body, the sensation is going through you, it's a terrific form of excitement for me. There are different sorts, there is the orgasm that's the quickie, then there's the middle ground one that you would call a full orgasm, and then there's the third type. I find you both enjoy each other for quite some time, those are not common but nice.

A/0/DK

9. This sort of wave that builds up, you know, runs over. They sort of arrive suddenly, no warning. It's not the case of touching there or rub me there—I'll come in the end.

A/0/DK

10. Text book fashion it's a strong muscular contraction, like a sort of shiver that's nice. I feel it in the genital area. It sometimes makes you feel light headed as well. He's inside me I suppose, but he can bring me with his hand too. I don't have it every time.

A/0/DK

11. You're gone sort of thing, it's nice, it's all over, you tingle. It starts in my belly and then it (shows by circular hand movements).

A/0/DK

12. Tingly...

A/0/DK

13. Sometimes I think I've had one and sometimes I don't. I mean you feel yourself going up and down and things happen. He's usually inside when it happens.

A/0/DK
14. I find it really weird that if I am masturbating I have a totally different orgasm than if I am having sex and I would say that orgasm is terrible excitement, physical excitement, in the vaginal region. If I am masturbating and I'm going to have an orgasm I just will, but I much prefer to be "bonked" cos then I am feeling is about penetration and physical movement in these regions. If I am going to "come" when we're having sex, it's very vaginal, although with a partner it has to be clitoral to start with, to get me going, and then I need him inside to really "come". It lasts longer if I'm masturbating, it's over quickly if I'm actually having sex. I've slept with several people and I've only ever had orgasm with my husband, and only about 3 or 4 times has that been without clitoral stimulation. I can't work out what happened on those occasions to make it that I actually "came" without having to spend hours having to stimulate the clitoris. Sometimes I just can't be bothered, so then I think sex is over-rated.

15. It's just like a feeling that you don't control, that you feel deep inside of you, like an ecstasy, feel it all over, more inside me, the clitoris as well, sometimes it's one or the other, but sometimes both.

16. I just go weak and dizzy and feel faint type of thing. I suppose it just builds up to it, I mean it doesn't happen right from the word go it just grows if you know what I mean, it gets more and then it dies down.

17. It feels good, I feel it all down below, it lasts a few seconds.

18. It just feels really nice. I'm usually clutching something, I usually get really sore hands when I tend to clutch either body parts or a pillow or something. If I have one on my own, or if I try to save it for when he has one he's pretty much doing the same thing that I'm doing cause he's having one too. Sometimes if he's not having one, he's watching me. I can have one without him inside me. I feel it kind of everywhere, kind of the start of it I feel in my feet, which is going to sound really weird, and then towards the peak I just feel it everywhere. I know the blood's kind of pounding, cos if I have a head-ache I can really feel it pounding in my head, pretty much everywhere.
19. I have very strong orgasms, but once I have one I can't go on any more, I have to sort of rest, but sometimes I feel not kind of satisfied, I feel I could go on, I sometimes feel kind of swollen there and I haven't been quite satisfied, but I don't know how to sort it out because when it happens I have to stop and let it all happen. The orgasm, and it totally involves my whole body. It was 100% but it feels as if I could have a rest and start again. It comes up from here right up to the top of my head so it's my whole body and my genital area is pulsing, it goes phmm, phmmm, and it lasts for, well certainly I'd say a minute, and it goes on and on and I want something firm to be there, to sort of hold it. It makes me feel nice all over. I sort of go out of control, I move a lot too, sometimes I have to hold myself because it doesn't, it rarely happens when we are having intercourse, it happens through manual, or if my husband is doing oral sex on me, it totally takes over for these number of seconds.

20. It's a funny sort of experience, a tingle all over. It's your whole body just tingles and you feel absolutely great and on top of the world and on top of the moon and then all of a sudden, that's it, and then you think "Oh, right, fine!"

21. It's a bit like having a coughing fit I suppose, your muscles sort of tense up-yes it's like having a good cough! for a few times then it goes away. Its more likely if he uses his hand.

22. I don't really get a lot of sensation, I get a lot of water. I thought the first time I was probably weeing myself. I can't seem to get what other people say they experience, the crashing down and the feeling in your stomach, I never seem to have got that. So I get a hell of a lot of water coming out, it's not wee, I feel I can't stop it coming out, it rushes out, I can't control it. It happens with different things like, I could be wearing a teddy, or fingering or licking out, I've got to have foreplay, I think that penetration is the bit that finishes it off, I get more pleasure from the first bit.

23. It's just a feeling of climactic release and it's just going through your whole body, not anywhere in particular. It's not like any other feeling, it's like a wave, it's like it's maximum, very pleasurable really. Most often he's inside, but occasionally it's just the stimulation I think usually the clitoris is involved.
24. It's a process of moving up various peaks and a feeling that may be centred in the genitals and then from the genital area to...the orgasm is everywhere else. It's like a sneeze! It's a great relief of tension.

25. It's like a tingly feeling, it's something that gives you an energy rush, and you don't want things to stop. It's good fun when you have one. The tingle is wherever you are being touched or being used in the body, you get a pulse-beat from wherever it's coming. Some days you will be just lying there and it will happen, other days you are exploring each other's bodies. If there is foreplay before the orgasm, then I orgasm first, if there isn't I may not orgasm. Sometimes he's inside me, sometimes not. I like my clitoris to be stimulated, it's relaxing as well, it's like somebody brushing it, I think this is the best thing about it.

26. For me it's a feeling that builds up inside you and then it gets to a point where you almost don't move because...you go almost stiff and just kind of wait, and it's just a wonderful feeling. I suppose it actually starts in the vaginal area, but for me cos I tend to stiffen up, my legs, and it then involves my whole body. I feel tender in my vagina as if you are at the point of something, you know it's coming, and when it breaks it's sort of a rush, it's warmth I suppose. It lasts about a few seconds to about 5 minutes. Although you feel good afterwards, the instant feeling is over very quickly.

27. It's lovely, I go all hot, and that feeling, you can't describe the feeling. It's not something that happens regular, I wish it could be all over again—it's a feeling all over. He's usually inside me when it happens, he uses his hand too to bring me to orgasm, I say to him "Don't stop" and things like that. Sometimes I have it again, the second time it's not as powerful.

28. It doesn't last long enough, it's a really nice feeling. I feel it all over. It's very rare that I can reach orgasm while we are having inter-course, I just don't seem to be able to get to the peak. The reason why I do it is because I know what I'm doing, if he does attempt to do it he's never in the right place for a start, and it used to be embarrassing to say to him "No, you are not in the right place, go down a bit". I know exactly how to find the right spot.
APPENDIX VIII. THE CODA STUDY

a) The invitational letter to parents. 325

b) An example of a completed questionnaire. 326
Dear Parents,

At the recent day conference held at The Institute of Child Health, it was agreed that I would send you a questionnaire asking about your children's CAH and your feelings about it. Leslie Green, of RTMDC has kindly agreed to send this questionnaire to you, and I would be most grateful if you would take the time to complete it and return it to me.

The answers will be very helpful in understanding the needs of the parents and the children with CAH. This understanding will help us to provide the best help for you all.

Thank you for taking the time to do this,

Best wishes,

Brenda May
Clinical Psychologist
The Hospitals for Sick Children
Great Ormond Street
London.
Appendix VIII (b) Coda Study – A completed questionnaire. Page 1 of 5

PLEASE HELP US BY GIVING THIS INFORMATION. IT IS IMPORTANT FOR US TO KNOW THESE THINGS SO THAT WE CAN PROVIDE THE BEST FOR CHILDREN AND THEIR PARENTS WHO COME TO THE HOSPITALS FOR MANAGEMENT OF THEIR CAH. THIS QUESTIONNAIRE IS CONFIDENTIAL AND YOUR INFORMATION IS ANONYMOUS. PLEASE DO NOT DISCUSS THIS WITH ANYONE UNTIL YOU HAVE COMPLETED THE QUESTIONNAIRE.

1. PLEASE GIVE THE FOLLOWING INFORMATION ABOUT YOURSELF:
   a) Your age: 34 years
   b) Are you married or co-habiting/single/divorced (please circle).
   c) Your occupation: Senior Care Assistant
   d) If co-habiting, your partner’s occupation:

2. THE FOLLOWING INFORMATION IS ABOUT YOUR FAMILY
   a) Age and sex of your child(ren) with CAH: 7 years. male

   b) Ages and sex of other in the family children:
      
      | Age | SEX |
      |-----|-----|
      | 8   |     |

   c) If any other members of your family have CAH, please state their relationship(s) to you below.

3. THE FOLLOWING INFORMATION IS ABOUT YOUR CHILDREN’S CAH:
   a) What age was your child(ren) when you were told about the CAH?

   At birth

326
b) How many operations has your child(ren) had?

3

c) Please briefly say what the operations were:

- Cleft lip reduction
- Vaginal reconstruction

d) Please briefly describe any future operations that you know will be necessary because of your child(ren)'s CAH:

More of the above, maybe soon (next year or so) and then probable vaginal 'tidying up' in teens.

e) What is your child(ren)'s present medication?

- 100mg Audrocortione 1 daily
- 10mg Hydrocortisone 1¼ mom 1 eve.

f) On your last visit to the doctor about the CAH, what were you told about your child(ren)'s health?

Very good, but weight gain a little too much and bone age still above actual age by 2 years. Also more pubic hair growth (not too much a health problem but worrying nevertheless).

4. THE FOLLOWING ARE MORE GENERAL QUESTIONS:

a) Describe in your own words what CAH is:

A 'blockage' in the adrenal glands which causes the adrenal gland to produce too much testosterone causing the genital abnormalities. The blockage also causes the minerals & hormones maintained by the adrenal gland difficult to stabilise.

If this has changed as a result of the recent conference you attended, please explain how

Not so much changed, but more understood.

327
b) What can you remember about your feelings when told about your child(ren)'s CAH?

It was the most traumatic event in my life. Terribly upset and tearful, at what the future held for Hannah. Although the consultants were very kind, they didn't give much information about the condition (referring to books continually) having just given birth to Hannah and not knowing what to expect. It was the hardest part.

c) Who else knows about your child(ren)'s CAH? (please tick those who know).

- [ ] Grandparents
- [ ] Aunts/uncles
- [ ] Very good friends
- [ ] Others (please say who)

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d) What has your child(ren) been told about the CAH?

Hannah understands that part of her body doesn't work properly and that is why she takes the tablets - to make it work like other children. Her last operation was when she was 4½ and as it was her 3rd she seemed to accept the situation and didn’t ask any questions.

e) Who told her/him when told?

He/She — the mother.

f) How old was she/he when told?

I have not made a point of explaining it to her, as I do not feel she would understand it all yet. I just answer: 'As you get older and is able to understand.' Has this changed as a result of the recent conference?

YES/NO (please delete as appropriate)

If 'YES' what age did you think was the best age before the conference?

h) Who do you think is the best person to tell the child?

Myself initially. The consultant when she is older, if that is what Hannah wants. Has this changed as a result of the recent conference?

YES/NO

If 'YES' who did you think was the best before the conference?
Appendix VIII (b) Coda Study – A completed questionnaire. Page 4 of 5

i) How easy do you find it to talk to your child(ren) about the CAH? (please put a cross on the line below to show how easy you find it)

[-----------] [------------] [------------] [------------]
very very easy easy quite quite easy easy hard hard

Has this changed as a result of the recent conference?

[YES/NO (please delete as appropriate)]

If "YES" please place a cross on the line below to show how easy it was for you before the conference.

[-----------] [------------] [------------] [------------]
very very easy easy quite quite easy easy hard hard

Please list below the aspects that you find it hard to talk to your child(ren) about

Because of her age and understanding both the medical side and the surgical side.

j) Do you have any problems and/or worries connected with the CAH at the moment? Please briefly say what they are.

I am concerned about her pubic hair, her weight and bone growth.

Did you have these before the conference?

[YES/NO]

k) Have there been any problems and/or worries in the past? Please briefly describe what they were giving the age of the child(ren) at the time.

Underweight and extremely difficult to feed up until her 1st birthday.

General worries concerning CAH always.
1) Describe briefly any problems/worries that you think there will be in the future?
   My main concern is the emotional problems Hannah may come across in her teens and again with fertility problems.

Has this changed as a result of the recent conference?
   YES/NO (please delete as appropriate)

   If "YES" please give brief details of how it has changed

5. ARE THERE ANY CHANGES THAT YOU WOULD LIKE TO SEE AT THE HOSPITAL VISITS?
   On the whole we are very pleased with the John Radcliffe.

6. PLEASE ADD ANYTHING ELSE THAT YOU FEEL WOULD BE HELPFUL;
   It would be extremely helpful to have more published information on CAH. One for the parents and another for a child.

DR. D.B. GRANT.
MRS B. MAY

THANK YOU FOR YOUR HELP. PLEASE RETURN THIS FORM TO BRENDA MAY IN THE PREPAID ENVELOPE PROVIDED.
APPENDIX IX  PUBLISHED ARTICLE

May B, Boyle M, Grant D (1996)

A Comparative Study of Sexual Experiences  In pocket
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A Comparative Study of Sexual Experiences

Women with Diabetes and Women with Congenital Adrenal Hyperplasia due to 21-hydroxylase Deficiency

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ACKNOWLEDGE. This research was supported by a grant from Action Research. The authors wish to thank all the women who participated, the psychotherapists who classified the descriptions of orgasm, and the consultants and staff of the hospitals which were involved in the study.

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Abstract

There has been little detailed study of the sexual outcome of women suffering from congenital adrenal hyperplasia, a condition which results in masculinization of the external genitalia and possible genital ambiguity at birth in chromosomal females. This study, combining qualitative and quantitative methods, reports the sexual outcome for a sample of women with congenital adrenal hyperplasia (CAH; N = 19), compared to that for an age-matched sample of women with early diagnosed diabetes (N = 17). The results are given under the headings of sexual interest; sexual activity with partners; reasons for engaging in sexual behaviour; sexual activities; stimuli eliciting a sexual response; physical problems connected with sexual activities, and general issues. CAH women were found to be less sexually experienced in all areas, and reported higher levels of penetration difficulties, pain, and lubrication problems than the diabetic women. It is proposed that the contribution of psychological factors to these problems may be at least as great as that of medical/surgical factors. Implications for management are considered.

Keywords

CAH, diabetes, females, sexual problems
THE PROFOUND SIGNIFICANCE to parents of the sex of their newborn infant is without doubt. Two of the questions most usually asked immediately after the birth are ‘Is it all right?’ and ‘Is it a boy or a girl?’ The answer to the question about the sex of the neonate has immediate and long-term consequences for the interactions of the parents and others with the developing child (Condry & Condry, 1976). There are, however, some medical conditions where the external genitalia are ambiguous, or do not entirely conform with the chromosomal sex of the infant, and for these newborn infants an early definitive answer is not always possible. Congenital adrenal hyperplasia is one condition in which this may occur. Although the condition may affect both males and females, this article is concerned only with females.

Congenital adrenal hyperplasia (CAH) due to 21-Hydroxylase deficiency is one of a group of autosomal recessive genetic disorders in which the activity of the enzymes necessary for adrenal hormone synthesis is deficient. These deficiencies lead to the accumulation of high levels of androgens during fetal development which, in chromosomal females, results in masculinization of the external genitalia although there is no effect on the internal reproductive organs. The degree of masculinization can range from a mild enlargement of the clitoris to a more severe abnormality. In these latter cases, complete fusion of the labia resembles the scrotum, and the urethra may be displaced to the shaft of the penis-like clitoris. The masculine appearance of the external genitalia may result in the mis-classification of the infant as male. About two-thirds of CAH infants also have abnormal regulation of sodium retention which, untreated, results in dehydration and a possible salt-losing crisis, usually occurring in the first few weeks of life. In the untreated course of the condition there is continued postnatal masculinization resulting in further clitoral enlargement. Early excessive growth is followed by premature maturation of the long bones with resulting short adult height. There may be an early appearance of pubic hair, failure to achieve normal puberty and hirsuteness.

In chromosomal females, treatment of CAH consists of surgical and medical components (Cutler & Laue, 1990). The external genitalia are surgically feminized, the necessary procedures depending upon the extent of the masculinization. The procedures may consist of clitoridectomy (amputation of the clitoris), clitoral recession (reduction in size), vulvoplasty (division of the labia) and vaginoplasty (opening and/or enlargement of the vagina). The initial surgery is now usually performed during the first year of life after the hormone imbalance has been stabilized, although vaginoplasty is more often deferred until puberty. Currently, clitoral recession is the surgery most preferred, rather than the more radical clitoridectomy routinely performed in the past. The second treatment component is that of lifelong daily administration of glucocorticoids with the aim of preventing further masculinization and controlling growth by bringing the androgen hormones to near normal levels. For those individuals with salt loss, administration of salt-retaining hormone during childhood is necessary, although in adulthood this is sometimes replaced by extra salt intake with meals.

**Effects of CAH on sexual behaviour in females**

It is now more than four decades since the introduction of such management to infants and children with this condition, and there have been a number of reports of their psychological and sexual outcome (e.g. Ehrhardt, Epstein, & Money, 1986; Ehrhardt, Evers, & Money, 1968; Mosely, Bidder, & Hughes, 1989; Slijper, 1984). While some of these studies indicate a tendency towards homosexual preferences in girls, detailed data on their specific sexual responses, attitudes and behaviours as mature women have been less frequently reported. Mulaikal, Migeon and Rock (1987), for example, reported only on the 'adequate/inadequate introitus' of their sample of CAH women and found from questionnaire data that 35 percent of their sample experienced an 'inadequate vaginal introitus'. They attributed these difficulties to the corrective surgery carried out on these women—vaginal reconstruction and clitoridectomy—rather than to the degree of fetal masculinization. In view of the necessary genital surgery and the possible effects of the prenatal and postnatal high androgen levels, the sexual outcome for these females is of considerable importance, and a fuller understanding of the effects of CAH on female sexual behaviour
would seem necessary for the optimum management of these women.

In addition to the direct effects of surgery, however, other factors may operate indirectly. It seems likely, for example, that sexual behaviour in these women may be influenced by emotions and cognitions related to having a condition which affects the external genitalia. These, as well as surgery itself, may affect sexual responses by increasing anxiety about sexual performance. Although Masters and Johnson (1966) and Beck and Barlow (1984) proposed that such anxiety may play a major role in the development of sexual problems, there has been very little research on this aspect of CAH in women.

As well as effects which may be attributed directly to CAH, there may be more general psychological effects of having a chronic condition necessitating lifelong medical supervision and medication. Attending hospital every three months or so during childhood is likely to affect self-perceptions, for example, as to whether one is ill or well, normal or abnormal, which in turn will influence social and sexual relationships. Some degree of differentiation between these direct and indirect effects would be provided by the comparison of CAH women with another diagnostic group requiring similar levels of medical consultation and medication. Diabetes mellitus fulfils these criteria, and for the present study has the added advantage of possibly resulting in sexual problems. The effects of diabetes on the sexual responses of females are inferred from the erectile problems commonly reported for diabetic men (Fairburn, 1981; Jensen, 1981). These problems in diabetic men are thought to be due to autonomic neuropathy. The parallel problem in women would be decreased vasocongestion and lubrication (Masters & Johnson, 1966), although as Kaplan and Steege (1983) point out, findings in this area have been inconclusive.

Our article therefore has two major aims: the first is to explore in detail the sexual outcome of women with CAH; the second is to attempt to clarify those aspects of sexual outcome which appear to be particularly related to CAH, rather than to the general effects of having a chronic medical condition with possible indirect effects on sexual functioning.

Method

Sample selection and recruitment

Fifty-six girls born between 1953 and 1972, diagnosed as having CAH, were identified from the records of a London specialist children's hospital. Exclusion criteria were: learning difficulties, other medical conditions, non-Caucasian cultural background and a single attendance for a second medical opinion. The fourth criterion was necessary because appropriate paediatric case-note information for these women was not available. Others were excluded because they could not be traced or because the consultant currently in charge of their management did not agree to their being approached. The characteristics of the initial and final samples are presented in Table 1.

<table>
<thead>
<tr>
<th>Categories</th>
<th>CAH N (%)</th>
<th>Diabetic N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Identified from records</td>
<td>56</td>
<td>33</td>
</tr>
<tr>
<td>Exclusions:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Non-Caucasian</td>
<td>0 (0.0)</td>
<td>0</td>
</tr>
<tr>
<td>Other medical conditions</td>
<td>4 (7.1)</td>
<td>0</td>
</tr>
<tr>
<td>Learning difficulties</td>
<td>2 (3.6)</td>
<td>0</td>
</tr>
<tr>
<td>Emigrated</td>
<td>4 (7.1)</td>
<td>0</td>
</tr>
<tr>
<td>Second opinion only</td>
<td>5 (8.9)</td>
<td>0</td>
</tr>
<tr>
<td>Untraceable</td>
<td>4 (7.1)</td>
<td>0</td>
</tr>
<tr>
<td>Sample available</td>
<td>37 (66.1)</td>
<td>33 (100.0)</td>
</tr>
<tr>
<td>Adult consultant not agreed</td>
<td>1 (2.7)</td>
<td>0</td>
</tr>
<tr>
<td>No reply to invitation</td>
<td>17 (45.9)</td>
<td>16 (48.5)</td>
</tr>
<tr>
<td>Final sample</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Agreed to participate</td>
<td>19 (51.3)</td>
<td>17 (51.5)</td>
</tr>
</tbody>
</table>
Table 2. Levels of severity of masculinization at diagnosis and types of surgery for interviewed CAH women

<table>
<thead>
<tr>
<th>Prader category</th>
<th>Clitoral recession</th>
<th>Clitoridectomy</th>
<th>Vulvoplasty</th>
<th>Vaginoplasty</th>
<th>Vaginal dilation</th>
</tr>
</thead>
<tbody>
<tr>
<td>2 or 3 (Mild)</td>
<td>1</td>
<td>3</td>
<td>1</td>
<td>4</td>
<td>1</td>
</tr>
<tr>
<td>N = 5 (27.7%)</td>
<td>4</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4 or 5 (Severe)</td>
<td>1</td>
<td>12</td>
<td>4</td>
<td>10</td>
<td>1</td>
</tr>
<tr>
<td>N = 13 (72.2%)</td>
<td>15</td>
<td>5</td>
<td>14</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>2</td>
<td>15</td>
<td>5</td>
<td>14</td>
<td>2</td>
</tr>
<tr>
<td>N = 18*</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* Data not available in medical records for one woman.

Based on information in the hospital medical notes, the CAH women who agreed to participate and those who did not were statistically compared for age, frequency of pediatric hospital admissions, age at and number of surgical operations, referral to psychological services and the level of external genital abnormality at diagnosis (Prader, 1954). As the groups did not differ significantly on any of these variables, the CAH women who agreed to be interviewed appear to be representative of the total sample.

Prader's classification is based on the level of abnormality at diagnosis and has five categories which range from 1 (normal female genitalia) to 5 (fully masculinized penis and fused labia without testes). Table 2 shows the distribution of the levels of masculinization at diagnosis and the types of surgical repair for the CAH women interviewed. The sample represents a group of severely masculinized women, with clitoridectomy and vaginoplasty being the most frequently performed surgery.

A comparison group of 17 women with early diagnosed diabetes (before eight years of age) was obtained from two London hospitals. Age range and exclusion criteria were identical to those for the CAH group. The method of recruitment differed for the two hospitals: One provided a list of female diabetic patients who fulfilled the criteria; the other requested one of us (BM) to attend the relevant clinics to identify suitable women from the medical notes. Information on the childhood characteristics and management of the diabetic group was not available.

The mean age of the CAH group was 26.06 years (SD = 5.63, range = 18.4–37.1), and that for the diabetic group, 27.01 years (SD = 5.308, range = 18.9–34.75). These means were not significantly different (t = −0.518 df 34, 2-tailed).

The study was submitted to and approved by all relevant hospital Ethical Committees, and assurance of confidentiality was given to all women who were approached. The consultant endocrinologists responsible for the current medical care of the CAH women were identified and asked for agreement for contact to be made. When agreement was given, an initial letter of invitation to be part of the study and a consent form were sent. Initial letters and consent forms were also sent to the diabetic women on the list provided by the hospital. The diabetic women at the second London hospital were approached by one author (BM) at the clinic, given a verbal description of the study, the letter of explanation and the consent form. One follow-up letter was sent to all the CAH and diabetic women who did not respond.

Data collection and analysis
To allow the respondents to express their beliefs and describe their experiences in ways that were meaningful to them, a qualitative method of data collection and analysis was used. Qualitative methods produce findings that are not dependent
on statistical procedures or quantification, but allow the exploration of meanings, ambiguities and contradictions. Although research topics were consistent across all interviews, the use of open-ended questions followed by appropriate prompts provided opportunities for the women to discuss issues and experiences which were of individual importance. The derived themes form the basis for theory-building which is ‘grounded’ in the obtained data (Glaser, 1978; Strauss, 1987). Although the emphasis was on qualitative methodology, some quantitative data were obtained; these included the recalled age at which the women started to date boys and, by use of a rating scale, the women’s perceived satisfaction with their sexual experience.

On receipt of the signed consent form, arrangements for the interview were made by telephone. All CAH and diabetic women, except for one in each group who came to the children’s hospital, were seen in their homes. Each interview was conducted by an experienced clinical psychologist (BM) and lasted about three and a half hours.

Transcripts of the taped interviews were summarized into matrices from which themes were identified and compared (Miles & Huberman, 1984). Where appropriate, statistical tests were applied: ages of respondents at start of dating were compared by t-test, the rating scale data were analysed by Wilcoxon’s Rank Sum Test, and Fisher’s Exact Probability Test was applied to frequency data. The women’s descriptions of their experience of orgasm were independently classified by two experienced psychosexual therapists into ‘Arousal’ or ‘Orgasm’. The therapists were blind to the diagnostic group from which each description came, and agreement of classification was found on 27 of the 28 items (96 percent); the non-agreed description was excluded from further analysis.

The separation of method and theory, and the resulting sections of results and discussion traditionally used in quantitative research reports, is not so appropriate in qualitative research. The three activities identified by Miles and Huberman (1984) as characterizing the analysis of qualitative data—the activities of summarizing and identifying themes, of organizing data and of drawing conclusions—occur concurrently throughout the research project. It is therefore more appropriate to combine the results and discussion in reporting qualitative studies, and this convention is followed in this article.

Results and discussion: Sexual experience

The data on sexual experience is reported under the headings: sexual interest; sexual activity with partners; reasons for engaging in sexual behaviour and stimuli eliciting a sexual response; satisfaction with sexual activities; physical problems connected with sexual activities; and general issues.

Sexual interest

Sexual interest is a broad and non-specific term, and the discussion here is limited to three types of data: that on dating, the women’s own perception of their sexual interest and masturbation.

Dating Dating is a complex collection of activities which includes the development of increasingly intimate relationships and the experience of sexual behaviours (Swensen, 1972). The following discussion is based only on heterosexual dating. Dating—defined here as arranged meetings with one person of the opposite sex with the understanding that sexual contact, however limited, might occur—usually begins in the early teen years: for example, in their in-depth qualitative study of 65 young newly-weds, Mansfield and Collard (1988) report that the recalled mean age for start of dating was 14 years. In this article, the CAH and diabetic women’s recall of their age when beginning dating was not significantly different (t (32) = 1.629, NS 2-tailed) with the mean age for the combined groups being 16.4 years. This is older than Mansfield and Collard’s sample, and compares with the results of Money and Schwartz (1977) who also found their group of CAH girls to be late in starting to date. The similarity between the diabetic and the CAH groups suggests that delay in starting to date may be influenced by having a chronic medical condition rather than being specific to CAH.

Perception of sexual interest Dividing the sample at the mean recalled age for the groups combined, resulted in differences in the wom
en's statements about their current interest in sexual activities. 'Late-daters' (recall of dating later than the total sample mean) in both groups and 'early-daters' (those who recalled started dating before the total sample mean) in the diabetic group saw their current interest in sexual activities as 'average' for their 'normal' peer group. By contrast, early-dating CAH women described themselves as currently less interested in sexual activities than did their peer group. This reported lack of interest may be reflected in the fact that none of the seven early-dating CAH women had boyfriends or partners at the time of interview, whereas all of the early-dating diabetics had relationships, with six of the eight having steady partners and the remaining two, boyfriends. The late-dating CAH and diabetic women show a similar pattern of steady partners and boyfriends with about 50 percent having partners, 20 percent having boyfriends and the remaining 30 percent without either.

These data suggest that the CAH women who had dated early may have a particular pattern of difficulties in this area, and it is of importance to understand why this might be so. Dating is a highly anxiety-provoking activity for all women (Kelly, 1984). The early physical maturity and development of the secondary sexual characteristics frequently seen in CAH girls may have been interpreted by themselves and by the young males in their environment as signals of sexual interest. However, the genital abnormalities and the experience of surgery may have made the young women less able to deal with the intimate behaviours required during dating. The early-dating CAH women described themselves as 'loners' and 'shy', and the anxieties arising from the perceived need to be intimate may have resulted in the curtailment of intimate heterosexual relationships. The following quotes from early-dating CAH women clearly exemplify this:

I get frightened getting too involved, I get worried and back off.
I did [end the relationship], it was getting too involved.
I got cold feet I think, I got out.

Their later reticence to engage in intimate relationships is conveyed in statements such as:

After the first, I never really had a date for years. I liked talking but they were only interested in sex.
No [boyfriends] at present. Sometimes I feel lucky not to have anyone who's moaning, but I see people and I think it would be nice to have that experience, but most of the time it doesn't bother me.

The diabetic women remembered such decisions as being less one-sided: some were taken by themselves, some by the boys and some jointly:

He did, he found someone else!
I did, it got too serious, he wanted it to be sexual and I wasn't ready.
We drifted apart, it was mutual.

Masturbation Masturbatory behaviour may also indicate sexual interest, and the data suggest that the CAH women were inexperienced in this area. Less than half of the CAH group had engaged in this activity; those who had had started in their early teens. By comparison, just over three-quarters of the diabetic women had masturbated since their early teens, and this approximates to normative levels as reported by Hite (1976) and Kinsey, Pomeroy, Martin and Gebhard (1953). An important finding was that those CAH women who had masturbated commonly spoke about masturbation as a necessary medical procedure rather than primarily as a sexual activity:

[masturbation was necessary] to keep it [the vagina] open.
I have done so, everybody does, I used to have to, I would explore—what had [the surgeons] done?
Even now, I've never thought about it for enjoyment.

Sexual activity with partners All of the diabetic group had experienced sexual intercourse. Three (16 percent) of the CAH women had not had this experience, and one other had had intercourse on a single occasion when 'drunk'. In order to compare the range of the specific experiences of the CAH and diabetic women, they were presented with a list of 25 sexual activities (see Table 3) and asked to identify which of these they had ever experienced. Overall, for every activity, the diabetic...
women were more likely to have experienced it than were those with CAH (T (25) = 0, p < .000009, 2-tailed). Although more of the diabetic women had partners at the time of interview, this did not explain the difference in sexual experience as the question asked about total and not current experience.

As can be seen from Table 3, the diabetics reported greater experience in every behaviour listed. The largest areas of discrepancy between the groups were in 'partner put a finger into your vagina', 'your partner ejaculated (came) in your vagina' and 'orgasm'. Other large differences were reported for vaginal penetration, 'your partner ejaculated (came) in your mouth', 'deep kissing', 'your partner ejaculated (came) outside your vagina', caressing and kissing non-sexual areas (dressed and nude), genital caressing and hugging nude. There is very little normative data on particular sexual activities although Breakwell and Fife-Schaw (1992) report percentages of females in the age range 16–20 in the general population engaging in a variety of sexual activities. The age ranges and the descriptions of

<table>
<thead>
<tr>
<th></th>
<th>CAH (N = 17)</th>
<th>Diabetic (N = 18)</th>
<th>Breakwell &amp; Fife-Schaw (N = 1315)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>88</td>
<td>94</td>
<td>-</td>
</tr>
<tr>
<td>2</td>
<td>82</td>
<td>83</td>
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Overall comparison of percentages of CAH and diabetic women reporting listed sexual activities. Wilcoxon Matched-Pairs Signed-Rank Test, T = 0, N = 25, p < 0.01 (2-tailed)
the activities are not entirely comparable with those of the present study.

Breakwell and Fife-Schaw’s data, however, suggest an increase in experience with increasing age. Three similar behaviours were asked about in the two studies: deep kissing, vaginal penetration and anal penetration. The sample of diabetic women in our article is older than Breakwell and Fife-Schaw’s female sample and, in line with these researchers’ finding of increased sexual experience with age, the diabetic women are somewhat more sexually experienced than the general population sample in those experiences looked at in both studies. By contrast, the CAH women, who are also older than Breakwell and Fife-Schaw’s sample, are not more experienced. As can be seen from Table 3, the sexual experiences showing the largest comparative inexperience for the CAH women appear to be of two, obviously related, kinds: those commonly occurring early in sexual activity and which increase arousal and orgasm itself.

The results also suggest that the CAH women are less likely than the diabetics ever to have experienced orgasm during masturbation or sexual activities with partners. The issue of the experience of orgasm is of particular interest in relation to women with CAH as the surgical correction of the external genitalia may affect this response. Information obtained during the discussion of masturbation provided additional data on climactic experience which is relevant here. The diabetic women who had masturbated (over three-quarters) and the smaller percentage (less than half) of those with CAH who had masturbated were asked if they had masturbated to orgasm; about half of those in each group reported that they had. These women, and any other women who reported orgasm during sex with a partner, gave a description of ‘what it felt like’. These descriptions were classified into high arousal or orgasm (Hite, 1976) by independent experienced psychosexual therapists, who were blind as to group membership.

Of the 19 women in the CAH group, 17 had had the opportunity to experience orgasm, whether through masturbation or sex with a partner. Of these, 13 (76.5 percent) reported experiencing orgasm. The independent judges failed to agree on one of these descriptions. Of the 12 remaining descriptions from CAH women, 7 (58.3 percent) were classified as describing orgasm. By contrast, all 17 women in the diabetic group had had the opportunity to experience orgasm through masturbation or with a partner, and 15 (88 percent) of these women reported experiencing orgasm. All of their descriptions were classified by the independent judges as representing orgasm. Although the percentage of orgasmic CAH women in our article is double the proportion—based on women’s self-report—reported in the 1977 study by Money and Schwartz (22 percent), it is still considerably less than for the diabetic group.

There are two possible explanations for this lower level of orgasm: that it is due to the medical aspects of the condition, that is, the genital abnormalities (level of masculinization) and the surgical repair, or to anxiety arising from engaging in sexual behaviour. On the medical side, the relationship among the extent of genital abnormality, corrective surgery and the ability to experience orgasm is difficult to examine as more severe levels of masculinization require more extensive surgery; it is therefore not possible to separate out these two effects.

The combined relationship of the two factors with the experience of orgasm (as judged by the independent raters) was examined by comparing the CAH women who had been classified as ‘severe’ (Prader rating 4 or 5) and ‘mild’ (Prader rating 2 or 3) at birth. No statistical relationship was found between Prader (1954) classification and experiencing orgasm (Fisher’s Exact Probability Test \( p > .05 \)). Nor was there any significant difference between the groups using the women’s own reports of orgasm. This result would not be expected if we assume that severity of genital abnormality at birth and extent of corrective surgery are important factors in later orgasmic experience. Although the sample numbers are small, it can be tentatively suggested that other, social and psychological, factors may be equally important.

Reasone for engaging in sexual behaviour and stimuli eliciting a sexual response

Although about one quarter of each group stated that they occasionally ‘had sex’ because their partners expected it, both groups felt the most common motivation was their own sexual need.
SEXUAL EXPERIENCES OF WOMEN WITH DIABETES AND CAH

For all the women this view was accompanied by a very strong statement that sexual activity was dependent on the existence of a close emotional relationship:

'cos I felt I loved him enough to let him into my body, your body has to be shared with somebody that you love (CAH).
I have to know them, trust them, for a long time (CAH).
I've never been able to distinguish between sex and love (diabetic).
Women have sex for love (diabetic).

The preceding reasons for making love are in line with normative female gender stereotypes (Hite, 1976), with both groups of women reporting the most important sexual stimulus to be that of a 'special person'. Interestingly male characteristics such as 'tight bums', 'muscular shoulders' and 'hairy chests' were mentioned by the CAH but not by diabetic women as stimuli which might elicit sexual interest or behaviour. This focus by the CAH women on male physical characteristics may reflect a more general conceptualization of sexuality in terms of external physical attributes. Two factors may have contributed to this. First, the frequent experiences of the women with CAH at the hospital clinics where there was a strong focus on the external genitalia. Second, although these women subscribe to the view that sex should be part of a close relationship, the difficulties they have with such relationships may lead them to rate external physical aspects more highly than do the diabetics.

All of the diabetic women were very clear about their lack of sexual interest in other women:
I can't say that any woman has ever done anything for me.
The whole idea kind of turns me off.
No, never.

Two (10.5 percent) of the CAH women had experienced lesbian relationships, and another three (16 percent) expressed a strong sense of sexual appreciation of females. They described considering what it would be like to have lesbian relationships, although they were very hesitant about acting on these thoughts. They attributed this hesitancy to an awareness of pressure against such behaviour from their families and friends:

If I see someone and I think 'she's pretty', I don't know—I know it's sort of wrong, you just don't do it do you? In our family that sort of thing is frowned upon, when you have these sort of thoughts you don't share them with anybody 'cos they'll think you're strange!
You look at a woman's body and think 'she's lovely', but I'm not sure about wanting to touch.
Some other female once said, and I agree with her, that another female would be able to please a woman.

It is difficult directly to compare these results with available normative data due to age and marital status differences and to variations in definitions of homosexuality (Hedblom, 1973; Hite, 1976; Kinsey et al, 1953). Overall, the incidence of homosexual behaviour among CAH women in our present study is likely to be similar to that expected in the general population. It is lower than the 29 percent in Money and Schwartz's (1977) CAH sample; it is also lower than the 22 percent reported by Dittmann, M. E. Kappes and M. H. Kappes (1992) but their definition of homosexual orientation included both behaviour and fantasy. The figure obtained in this article is, however, higher than the 5 percent reported for CAH women by Mulaikal and colleagues (1987). Hite (1976) reported that sexual appreciation of other women was frequently mentioned by the women in her sample, but that this did not predict experience with lesbian relationships. The fact that women with CAH may be particularly anxious about intercourse, and may have had negative experiences with men, could make it more likely that they will welcome homosexual relationships. Alternatively, their general anxiety about sex might inhibit both heterosexual and homosexual relationships. The social and psychological issues which surround CAH certainly make it difficult to interpret homosexual preference in terms of prenatal hormonal influences.

Satisfaction with sexual activities
There is obviously no absolute appropriate level of sexual activity and there are likely to be wide individual differences in the levels held to be satisfactory. Both groups of women studied here might be satisfied with their current level of
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sexual activity even if this is low, particularly as some women with CAH reported low interest in the opposite sex. To gain more information on this issue, we asked the women to rate their level of satisfaction on a 5-point scale. The median satisfaction ratings for the CAH and the diabetic women were 3 and 4 respectively; an overall comparison of the distribution of scores for the two groups showed that the satisfaction ratings of the CAH women were significantly lower than those of the diabetic women ($W = 205, N = 16$, $p = .05$, 2-tailed). The two groups of women also reported that different kinds of change would be necessary to increase their satisfaction. Their different responses reflect the CAH women's restricted opportunities for having sex with a male partner and their preoccupation with having 'successful' intercourse. By contrast, the diabetic women would like an extension of their present sexual activity:

If I could have sexual intercourse with a man inside my vagina (CAH).

As soon as I manage to have sex. Oh my God, I'm 19 and still a virgin! (CAH).

I'd like a more active sex life, more of it (CAH).

More time to enjoy it (diabetic).

We don't have enough time for it, in the evenings we are too tired and we settle for a cuddle (diabetic).

**Physical problems in sexual activities**

It is clearly possible that the genital abnormalities and their surgical correction may result in physical difficulties during intercourse for women with CAH, problems connected, for example, with penetration, lubrication, residual pain from tissue repair and reduced sensitivity of the clitoris. Just over half (56 percent) of the CAH women who had experienced sexual intercourse reported problems with penile penetration. These problems did not appear to be related to levels of masculinization, which is in line with Mulaikal's 1987 study, and the problems had decreased for those having regular intercourse. The women themselves attributed these difficulties to tightness or shortness of the vagina or to their operations. For two CAH women (25 percent), failure of penetration, or anxiety about penetration, had resulted in the complete avoidance of heterosexual activities. Penetration was not a problem for the diabetic women with only two (13 percent) mentioning this difficulty in the early days of sexual experience; this was attributed to being a virgin.

Three-quarters of the CAH and half the diabetic groups recalled experiencing some degree of pain during the first few occasions of intercourse. Pain was persistent, however, only for the CAH women, with those reporting early experience of pain also reporting current pain. As with penetration difficulties, current pain did not appear to be related to level of masculinization. Equal proportions of the CAH women who reported current pain had been classified in the 'mild' and the 'severe' Prader categories (1954). As might be expected, those women with CAH who reported current pain also reported current penetration problems.

The attributed cause of pain differed for the two groups. While the CAH women saw it as due to the size of the vagina or 'something there, blocking him getting in', the diabetic women felt their pain was due to 'dryness' of the vagina:

It seems to vary with the position, so I don't know what's in the way (CAH).

I feel my womb has dropped—it seems to be hitting something (CAH).

(When it's dry) when it's dry (diabetic).

When I'm tense and I don't get wet, it is painful (diabetic).

The sensitivity of the clitoris may be reduced or eliminated in CAH women by the surgical procedures of early childhood. Whereas all the diabetic women perceived their clitoris to be sensitive, seven (44 percent) of the 16 CAH women who had masturbated or had sexual intercourse reported a lack of sensitivity. The reported sensitivity or insensitivity was not related to reported pain nor to the level of masculinization. This latter finding may be explained by the radical surgery (clitoridectomy) which was experienced across levels of masculinization by the women in this study. Interestingly, clitoral insensitivity was reported by both orgasmic and non-orgasmic women. Other research, for example, Lightfoot-Klein (1989), also suggests that climax is not dependent on perceived clitoral sensitivity and that fully circumcized infibulated females are capa-
ble of attaining orgasm. In this study, however, those women reporting clitoral insensitivity were twice as likely to have penetration problems.

Lubrication of the vagina is an essential factor in ease of penetration during intercourse and is also believed to be an indicator of arousal. Arousal and, therefore, lubrication are inhibited by anxiety (Masters & Johnson, 1966; Beck & Barlow, 1984). As the data already reported here suggest that sexual anxiety may be common in CAH women, it might be expected that these women would experience lubrication difficulties. Nine (60 percent) of the CAH and six (37.55 percent) of the diabetics reported lubrication problems. The diabetic women, however, believed that 'dryness is common in diabetics', and all of them had used artificial lubricants to alleviate the problem:

Yes, I use jelly sometimes, I know diabetics have problems.
I think I should always use jelly. (My husband) saw in a mag that diabetics don't produce the lubricant. At least I know there's nothing wrong with me.

This practical approach to a medically attributed problem was not apparent in the CAH women who seemed to have neither a medical nor a psychological explanation for their lack of lubrication and had not considered that there might be a remedy:

I think I do (get wet), but I can never tell if it's him or me doing it. I'm often dry, but I've never used jelly.
I do get wet, and then I dry up quickly. It takes a bit of time. ... You just have to work at it.

As might be expected, those with lubrication problems were twice as likely to have penetration problems.

In considering the relationship between the physical problems experienced by the CAH women in this study, and their level of masculinization, we feel that a number of factors need to be taken into account. The sample is small and includes a number of women in the CAH group who have not had sexual experience. The fact that some aspects of surgery were similar for women across levels of abnormality makes any relationship between levels of masculinization and sexual problems difficult to examine. Data from those women who had had clitoridectomy and relevant sexual experience show that the numbers reporting the presence of current pain during intercourse are about the same as those reporting its absence; similarly, almost equal numbers report experiencing orgasm as report not experiencing it. More women, however, report problems with penetration, lubrication and clitoral sensitivity than report the lack of such problems. As can be seen from Table 4, however, the numbers in each category are so small that it is difficult to draw clear conclusions. Further investigation is needed before outcome relationships among levels of masculinization, type of surgery and sexual problems can be clarified.

**General issues**

The majority of the CAH women perceived their surgery as having caused difficulties for them in their sexual behaviour. It was common for them to conceptualize sexual intercourse as 'a hurdle to be got over' and the sexually experienced women remembered extreme worries that they would not be able to achieve penetration:

At the beginning, I was so tensed up any-

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<th>Table 4. Frequencies (and percentages) of physical problems for CAH women having had clitoridectomy</th>
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<td>Problem reported†</td>
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<td>Problem not reported†</td>
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† Expressed as percentages of those having relevant sexual experience.
* Expressed as percentages of total N.
way—there seems to be a block even now, as if it's not long enough.
In the early days, he didn't get fully in, but it's OK now.
It's OK now. It wasn't at the beginning, but it's not deep because of scar tissue, but he's getting further in each time.

Although it is likely that most women experience anxiety about the beginnings of sexual experience, the strong expressions of anxiety by the CAH women were not echoed by the diabetic women. This is perhaps explained by the fact that the CAH women focused their anxiety on their genital abnormalities and were doubtful about the success of the surgery:

I'm very worried about scarring from the operation—it's not normal, it looks knocked about. I've always got the feeling that it's going to close up.
My boy friend says I'm small, but he said it's not surprising cos I'm a virgin. I wanted to be able to do it off my own bat without using dilators. They said you had to do it gradually rather than go 'bash', or you would cause a great deal of damage.

The women with CAH also expressed anxiety about the acceptability of their genitalia to their partners. They recalled their considerable relief when no comment was made about their normality:

... the fear that the man would find out, would he say 'My God, what's the matter with you?'
I must be all right 'cos he didn't say anything.
He said I was doing it properly, but I don't know if I was. I was reassured when he didn't make any comments about it being normal.

It is not surprising that this issue of normality was central in these women's lives, and it is not difficult to understand their uncertainty about the state of their genitalia. As one CAH woman said:

How do you know what you are supposed to look like and how it is supposed to be down there, you never see anybody else do you?

Neither the CAH nor the diabetic women felt that they could talk to their consultants about any sexual problems. Nor did the CAH women see it as appropriate to discuss their anxieties about genital abnormalities with their doctors. There was a general attitude in the CAH group that nothing could be done and that there was no one to go to for help. A popular solution for any sexual difficulties or anxieties was to wait for 'the right man' to come along; it was believed that this event would alleviate any problems, and everything would then be achievable.

Overview

The CAH women were less sexually experienced than was the comparison group of diabetic women, were more reluctant to establish relationships with males and were less satisfied with the frequency of their sexual opportunities. They also reported higher levels of penetration difficulties and a pattern of persistent pain during intercourse. Fewer CAH than diabetic women masturbated or attained orgasm. Adequate lubrication did seem to be a difficulty for both groups; for the diabetic women, this may suggest some parallel with the neuropathy seen in diabetic men. Anxiety about a number of areas concerned with sexual activities was, however, a central problem for the women with CAH. While the pain and the difficulties they experience with penetration may lead them to avoid sexual activities, they will also, of course, reinforce concerns about the normality of the genitalia and be construed as confirmation of 'not being like other women'. Such concerns are in fact common among women (Hite, 1976), but they are given added force among women with CAH by their medical history and by their sexual experiences, even if their partners do not comment on any abnormality. In addition, although both the CAH and the diabetic groups expressed the ideal of sexual intercourse as part of a loving relationship, in practice the women with CAH often construed intercourse as 'a hurdle to be got over'. Many also construed their sexual experiences as 'invasions', 'violations of my body' and 'rape'.

We do not know to what extent these constructions reflect the behaviour of the male partners of women with CAH. Nevertheless, that the CAH women should associate their genitalia with unwanted procedures rather than primarily with sexual need or satisfaction is not surprising.
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The early surgery, vaginoplasty at adolescence and repeated vaginal examinations during visits to outpatient clinics make this very understandable. Indeed, it was notable that the women with CAH also used a language of 'rape', 'invasion' and 'violation' when talking about vaginal examinations and other procedures carried out during visits to paediatric and adult clinics.

Similar findings were reported by Money and Schwartz (1977). The extensiveness and invasiveness of the medical and surgical procedures are, in addition, likely to have contributed to the generally held view of the CAH group that their sexual problems were entirely a result of CAH, and to their striking failure—in contrast to the diabetic women—to consider non-medical influences or methods of alleviation. This all-or-none approach was echoed in the belief, expressed only by women with CAH, that 'the right man' would somehow make everything right. Such a belief is, of course, strongly encouraged by women's romantic fiction (Coward, 1984), and in this case it appeared to be related to the women's sense of helplessness about their problems.

Both groups of women expressed reluctance to ask for professional advice on sexual activities and problems. For the CAH women, however, this reluctance was part of a greater feeling of isolation from all sources of information and understanding about their condition and its effects (May, 1994). Addressing the psychological aspects of CAH will, of course, necessitate the provision of opportunities for open discussion of a number of issues concerned with CAH and its effects on sexual functioning, opportunities that the women do not currently feel are available to them.

The relationship of sexual problems with initial levels of masculinization is difficult to establish from the current data. It is difficult to interpret the distribution of reported sexual problems across initial levels of masculinization because similar surgical techniques were applied across levels of abnormality in the early days of treatment. Further investigation using larger numbers of mildly masculinized women is essential before the direct effects of physical abnormality and surgery on sexual functioning can be clarified. Women with CAH, however, do appear to have some sexual difficulties which are particular to their condition. But it is essential that these problems are not conceptualized as solely medical and/or surgical. Such a model may lead to the belief that the sexual well-being of these women is entirely a function of successful surgical procedures and endocrinological management, while overlooking important psychological factors. For women with CAH, the psychological factors may become independent of the medical aspects and must be addressed in their own right. Indeed, the findings of this study suggest that the psychological consequences of the condition and its management may have just as much influence upon sexual outcome as any of the physical limitations or deficiencies resulting from the initial masculinization.

References
Ehrhardt, A. A., Evers, K., & Money, J. (1968). Influence of androgen and some aspects of sexually dimorphic behavior in women with the


JOURNAL OF HEALTH PSYCHOLOGY

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• encourages the development of sound theoretical analysis in health psychology;
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3. Papers should be as short as is consistent with clear presentation of subject matter. There is no absolute limit on length but 8000 words, including footnotes and references, is a useful maximum. The title should indicate exactly, but as briefly as possible, the subject of the paper. An abstract of 100 to 120 words should precede the main text, accompanied by up to 5 key words and a bio-bibliographical note of 25 to 50 words. The covering letter should indicate whether the author prefers blind or open peer review.

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Tables and figures should have short, descriptive titles. All footnotes to tables and their source(s) should be typed below the tables. Column headings should clearly define the data presented. Camera-ready artwork for all figures must be supplied. Artwork intended for same-size use should be a maximum size of 192:125 mm (page depth: page width).

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