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Major Research Project

“Living With This Thing Called Cushing’s”

An Exploratory Account of Patient Experiences, Illness Representations & Identity

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# Table of Contents

**ACKNOWLEDGEMENTS** ................................................................. 5

**ABSTRACT** .................................................................................. 6

**CHAPTER 1: INTRODUCTION** .................................................. 8

**CUSHING’S SYNDROME** .............................................................. 10
  * Normal Glucocorticoid Regulation .................................................. 10
  * Aetiology ....................................................................................... 11
  * Figure 1. Typical Control Loop for Pituitary Hormone Release .......... 13
  * Epidemiology .............................................................................. 14
  * Symptomatology .......................................................................... 14
  * Table 1. Causes of Endogenous Cushing’s Syndrome ...................... 16
  * Table 2. Symptoms in Cushing’s Syndrome .................................... 16
  * Diagnosis & Treatment ................................................................ 18
  * Course & Outcome ..................................................................... 19

**PSYCHOSOCIAL ASPECTS OF CUSHING’S SYNDROME** ............... 22
  * Personality and Cushing’s Syndrome ........................................... 22
  * Patient Beliefs and Quality of Life .............................................. 23
  * A Survey of Patients’ Experiences .............................................. 24
  * An Autobiographical Account .................................................... 26
  * Psychological Treatments ........................................................... 27

**DEVELOPING A PSYCHOSOCIAL PERSPECTIVE ON CUSHING’S SYNDROME** ........................................................ 28
  * Perspectives on Chronic Illness .................................................. 28
  * The Distinct Context of Cushing’s Syndrome ............................... 34

**PHENOMENOLOGY** ................................................................. 36
  * The Phenomenological Method ................................................... 37
  * Points of Origin and Destination .............................................. 39

**THE COMMON-SENSE MODEL OF ILLNESS SELF-REGULATION** . 41
  * Background .............................................................................. 41
  * Components of the CSM ............................................................ 42
    * What counts as a Stimulus? ................................................... 42
    * Illness Representations ......................................................... 42
    * Figure 2. The Common-Sense Model of Illness Self-Regulation .... 45
    * Emotional Representation ................................................... 47
    * Bilevel Representations & The Symmetry Rule ....................... 48
    * Procedures ............................................................................ 49
    * Appraisal .............................................................................. 50
  * Utility of the Common-Sense Model in the Present Study .............. 51
  * Where is the Self in Self-Regulation? ......................................... 53
    * Definition of the Self in the Common-Sense Model ................. 54
    * Regulation of the Illness & Regulation of the Self .................. 55
    * The Social Context, the Self and Illness Regulation ................ 56

**POSSIBLE SELVES** .................................................................. 58
  * Utility of the PSM in the Present Study ....................................... 61

**PERSONAL INFLUENCES** ............................................................ 63
RESEARCH OBJECTIVE & QUESTIONS ............................................................................................... 65
Research Objective ....................................................................................................................... 65
Research Questions ....................................................................................................................... 65

CHAPTER 2: METHOD .................................................................................................................. 67
OVERVIEW .................................................................................................................................. 67
RESEARCH ETHICS APPROVAL ................................................................................................. 68
INTERVIEW SCHEDULE DEVELOPMENT & PILOTING ............................................................. 68
  Initial Interview Schedule ........................................................................................................ 69
  Piloting ..................................................................................................................................... 70
  Revised Interview Schedule ................................................................................................... 72
  Boundaries & Participant Protection ......................................................................................... 74
PROCEDURE ............................................................................................................................... 75
PARTICIPANTS ............................................................................................................................ 77
ANALYSIS .................................................................................................................................... 79
  Table 3. Participant Basic Demographics .............................................................................. 80
VALIDITY CHECKS ...................................................................................................................... 84
REFLECTIONS .............................................................................................................................. 86

CHAPTER 3: RESULTS .................................................................................................................. 90
EMERGING THEMES .................................................................................................................. 90
  Figure 3. A Diagrammatic Representation of Emerging Themes ......................................... 93
EVOLVING UNDERSTANDING ................................................................................................. 94
  Clusters of Understanding ...................................................................................................... 95
    Everyday Causes ....................................................................................................................... 95
    There is Something Wrong .................................................................................................... 98
    Gone and Forgotten ............................................................................................................... 102
    Cushing's Is For Life ............................................................................................................. 108
    A Vague Menace .................................................................................................................... 113
  Multiple Shifting Models ......................................................................................................... 115
  Social Framing of Understanding ............................................................................................. 117
    Shaping & Feedback ............................................................................................................... 118
    Strain of Conflicting Beliefs .................................................................................................. 123
    If Doctors Don't Listen to Me, How Can I Trust Their Advice? ......................................... 124
    Evolving Understanding: Overview ...................................................................................... 127
TRANSFORMATION OF SELF .................................................................................................. 128
  Dissolution ............................................................................................................................... 129
    Losing Positive Selves .......................................................................................................... 129
    Gaining Negative Selves ........................................................................................................ 133
    Reconfiguring ........................................................................................................................ 134
  Social Framing of Identity ....................................................................................................... 138
    Identity-Affirming Contexts .................................................................................................... 139
    Identity-Damaging Contexts .................................................................................................. 141
    Transmutation of Self: Overview .......................................................................................... 145

CHAPTER 4: DISCUSSION .......................................................................................................... 146
HOW DO PATIENTS WITH CUSHING'S UNDERSTAND THEIR ILLNESS? ................................ 147
  An Evolving Understanding ...................................................................................................... 147
  Evaluation, Links and Implications ............................................................................................ 153
  The Dynamic Nature of Representations .................................................................................. 154
  Multiple Understandings ......................................................................................................... 157
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Abstract

Cushing's Syndrome is a rare, insidious and elusive endocrine disorder that disrupts multiple physical and psychological balances. Neuro-endocrine and psychiatric research has described multiple factors relating to course and outcome, though the psychosocial determinants of the patient's experience and adjustment have been largely unexamined. However, clinical opinion, outcome data and first-person accounts hint at significant discrepancies between biochemical status and quality of life in the long recovery period. In this study, I argue for a broad, phenomenological approach to the experiences of people with Cushing's Syndrome, in order to explore significant psychosocial factors relating to adaptation, which could form the basis for future research and clinical practice. From a broad survey of the literature on illness, I have selected two influential frameworks, the Common-Sense Model of Illness Self-Regulation (Leventhal, Leventhal & Cameron, 2001) and the Possible Selves Model (Markus & Nurius, 1986) to establish two research questions that broadly frame this investigation: How do patients with Cushing's Syndrome understand their illness? and How does Cushing's Syndrome -and patients' understanding of it- affect their identity, and how does identity influence representations? In-depth interviews with fifteen participants, recruited through Endocrine and Neurosurgery clinics, were analysed iteratively using Interpretative Phenomenological Analysis (Smith, Flowers & Osborn, 1997). Checks on the analysis involved expert, peer and participant involvement, and self-reflection. Two central themes emerged: an Evolving Understanding of the illness, which is composed of
distinct clusters of understanding that patients evolved while struggling to understand and control their illness; and the *Transmutation of the Self*, which describes the strong impact of the illness -and its perception by patients and their social context- in dissolving and reconfiguring core components of the Self. The discussion describes and links these themes to existing concepts and findings in the literature, noting the degree of overlap with accounts of acute, chronic and functional disorders. Equally, however, the data illuminate several dynamic aspects of the process of Illness Representation, which have received little attention, and propose new integrative hypotheses about the relationship of Identity to Illness Representation. Implications of patients’ accounts for clinical practice are suggested, and finally, limitations and alternative qualitative organisations are discussed.
Chapter 1: Introduction

Imagine: your body slowly but steadily develops a flabby, distended belly, bulky shoulders and asymmetrically thin arms and legs. Looking in the mirror, you can’t help but notice your face becoming round, purple, fat and hairy in all the wrong places. You become too physically weak to even lift a shopping bag, and too tired to get to the supermarket anyway. The slightest of jolts can leave large bruises and angry skin tears, your skin erupts in acne, bones snap with little incitement. This would make anyone irritable, but on top it seems that for you, confusion and dark despair are always round the corner. You are too tired to keep up with friends, too ashamed to show your body to your partner, but too afraid to be alone. In the midst of all this, you can’t think straight, your memory is shot, and you cry at the slightest things. But you’re never quite sure, is there something truly deadly going on, or are you going mad?
At a prevalence of about 5 cases per million per year, the majority of the population will never have even heard of Cushing’s Syndrome, the endocrine disorder that is responsible for the above litany of misery. And the majority of people never will; yet for the people it affects, it forces a lengthy, damaging disruption of health, lifestyle, personality and relationships.

Cushing’s Syndrome is an endocrine disorder of hypersecretion of the hormone cortisol. The sources of the disorder in the body are known, but its fundamental causes are not, despite compelling personal accounts that link the onset with stressful life events. Insidious in onset, it has complex, often vague or masked symptoms such as weight gain, muscle weakness, hypertension, mood changes, and osteoporosis. Though in the early stages these are often misdiagnosed or treated individually, once a diagnosis is established the medical and surgical treatments are effective for the majority of patients. However, it is patients’ experiences before diagnosis, and adaptation after treatment, that pose a challenge. In the scant literature referring to aspects of adjustment, clinicians and researchers frequently hint at significant problems, and studies demonstrate high rates of continuing impairment, often despite a resolved clinical condition.

This study seeks to redress an imbalance in the literature that has to date mainly addressed the biochemical or the psychiatric aspects of Cushing’s Syndrome, and has paid less attention to personal experiences, and individual adjustment. In the following sections, I present an overview of the physiological basis and medical treatment of Cushing’s Syndrome, highlighting the existing psychiatric research findings. I will critically review the few studies that adopt a psychosocial perspective, arguing that b
much more can be done to establish a coherent and clinically useful psychosocial contribution. I suggest that an exploratory, qualitative approach to the personal experience of Cushing's can provide valuable insights into the psychosocial factors of adjusting to the illness, and have distinct clinical utility. Furthermore, the proposed approach can provide a basis for future research and theoretical development, since this disorder presents a unique combination of biological, psychological and social influences. To this end, two broad theoretical frameworks are critically examined and used to formulate guiding research questions.

Cushing's Syndrome

Cushing's Syndrome (CS) is a complex endocrine disorder whose clinical symptoms and biochemical manifestations are secondary to excessive exposure of the body to glucocorticoid hormones, primarily cortisol. The role of cortisol in the human body is complex, and often debated, yet it is broadly involved in the function of the immune system, the body's stress response and metabolism, especially of fat. The following sections describe the various aspects of the syndrome's clinical presentation and treatment.

Normal Glucocorticoid Regulation

In its normal state, cortisol production by the cortex of the adrenal glands is tightly regulated by a complex feedback mechanism between the hypothalamus, pituitary

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1 Drawn from Boscaro (Boscaro et al., 2001), Sonino & Fava (2002), Grossman (2000) and Forsling & Grossman (1986), except where specific references are given.
Adrenocorticotrophic Hormone (ACTH) released into the blood stream by the pituitary gland under the influence of hypothalamic Corticotrophin-Releasing Hormone (CRH), reaches the adrenal glands, where it stimulates the synthesis of cortisol. The blood cortisol level in turn acts as feedback to the pituitary and hypothalamus, to regulate ACTH production; thus, if there is an excess of circulating cortisol, this acts as negative feedback on the pituitary, and inhibits further production, until blood levels return to their baseline.

This simplified control system belies a complex diurnal rhythm of hormonal synthesis. Both ACTH and cortisol vary as a function of the sleep-wake cycle over 24 hours. Thus, peak secretion is seen between 07:00-08:00 hours, with a progressive fall, until midnight and the early morning hours, where secretion is at a minimum. In addition, physiological and psychological stressors result in rapid increase of circulating ACTH independently of cortisol feedback or diurnal regulation.

**Aetiology**

There are three main causal pathways to cortisol excess in CS: *exogenous, endogenous ACTH-dependent* and *endogenous ACTH-independent* (Beauregard, Dickstein & Lacroix, 2002).

- **Exogenous** - Chronic overuse or overexposure to cortisol, taken by patients in the form of medication (such as hydrocortisone) for its anti-inflammatory effects in

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2 See a schematic illustration of the pituitary and how it is controlled in Appendix A.
disorders such as arthritis, asthma, eczema and colitis, or for its immuno-suppressive properties to reduce the risk of transplanted organ rejection, may result in iatrogenic CS.

- **ACTH-Dependent** - When Cushing’s Syndrome is linked to overproduction of ACTH due to a *pituitary adenoma*\(^3\), the term *Cushing’s Disease* (CD) is used. In this condition, which is the most frequently observed cause (see Table 1), aberrant *corticotroph* (ACTH-producing) cells of the anterior pituitary lose their responsiveness to the feedback effects of cortisol, resulting in over-production of ACTH (and consequently over-production of cortisol by the adrenals). Meanwhile, the normal ACTH-producing pituitary cells are inhibited by the high levels of cortisol, resulting in a relative advantage for the abnormal cells, and proliferation (Grossman, 2000). Less frequently, an ACTH-secreting tumour at a different location in the body (*ectopic*) may also be the cause of CS.

- **ACTH-Independent** - Cortisol excess may alternatively be the result of adrenal hyperplasia, adrenal adenomas or carcinomas.

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\(^3\) In general, adenomas (non-malignant cell growths) of the pituitary are not infrequent in the general population, with lifetime incidence being around 10%. Corticotrophic (ACTH-producing) adenomas comprise around 10% of all pituitary adenomas (Russell & Rubinstein, 1999). Most adenomas, though not malignant and rarely metastatic, will typically increase in size during the lifetime and if untreated in the long term can cause pressure effects on the brain and other areas of the pituitary. This can affect the production of different pituitary hormones (eg prolactin, growth hormone), resulting in complex patterns of hormonal imbalances. (Anderson et al., 1999; Russell & Rubinstein, 1999).
Figure 1. Typical Control Loop for Pituitary Hormone Release

The following diagram is a schematic drawing of the control system pituitary hormone release.

Epidemiology

CS occurs in the general population across all age groups in very low rates (typical prevalence: 1 in 250,000; Grossman, 2001). CD accounts for about 70-80% of all cases of CS, with an estimated 0.1-1 new cases of CD per 100,000 population per year. Both CS and CD are more prevalent in females than males, with a ratio around 4:1. In a series of 302 patients, Boscaro et al. (2001) recorded a mean age of 38.4 years (SD 13.5, range 8-75) and a F:M ratio of 3.8:1.

Symptomatology

The glucocorticoid (cortisol) excess in CS, regardless of source, impacts on multiple bodily systems and is associated with a variety of symptoms and signs (see Table 2). It causes protein catabolism (breakdown of proteins), resulting in muscle weakness, osteoporosis and thin skin:

‘The weakness is the worst. Daily you become weaker and weaker, you walk less and less far. Your legs become so weak that you must look for where you may sit before entering a place. You cannot carry a half gallon of milk from the back of a store to the checkout area, you learn to take a cart just for the purpose of leaning on it’ (Gotch, 1994, p.609)

It also causes lipolysis (alterations in fat metabolism), which is evident in characteristic patterns of torso obesity combined with thin limbs, facial plethora (rounded, moon-like, puffed-up face), and a ‘buffalo hump’ (concentration of fat at the rear of the neck). Combined with the thin skin, this can cause easy bruising and characteristic purple striations in the abdomen:
'Cushing's made me feel fat and ugly - with facial hair, ruddy complexion and skin that bruised easily' (Gotch, 1994, p.610).

High levels of glucocorticoids also have *immunosuppressant* effects, leading to an increased susceptibility to illnesses and common infections, and retarded wound healing. They may dysregulate glucose distribution, and can cause *diabetes mellitus*. Due to concurrent adrenal hypersecretion of *androgens* (the family of hormones that includes *testosterone*), *acne, seborrhoea* (greasy skin), *hirsutism* (excessive facial and bodily hair), decreased libido, sexual dysfunction, menstrual irregularities and infertility may result.

Major mental state changes are also a hallmark of CS, and indeed have been remarked upon since the original description of Cushing's Syndrome by Harvey Cushing in 1932 (Lindholm, 2000). Observed symptoms include irritability, depressed mood, mood lability, crying, anxiety, insomnia, social withdrawal and suicidal ideation. Primarily, these are classified as major depressive disorder, with rates of 50-80% generally reported across various studies, with an overall average of 57% for all 490 patients included in a review (Sonino & Fava, 2001). More infrequently, psychotic and other psychiatric illness are diagnosed, though this is likely to be biased by a tendency to publish unusual case presentations (Reed, Watkins & Dobson, 1983; Zielasek *et al.*, 2002).
### Table 1. Causes of *Endogenous* Cushing’s Syndrome.

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>Frequency (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>ACTH-Dependent</td>
<td></td>
</tr>
<tr>
<td>Pituitary-dependent (Cushing’s Disease)</td>
<td>66</td>
</tr>
<tr>
<td>Ectopic ACTH-secreting tumours</td>
<td>7</td>
</tr>
<tr>
<td>ACTH-Independent</td>
<td></td>
</tr>
<tr>
<td>Adrenal adenoma or carcinoma</td>
<td>24</td>
</tr>
<tr>
<td>Macronodular adrenal hyperplasia</td>
<td>3</td>
</tr>
</tbody>
</table>


### Table 2. Symptoms in Cushing’s Syndrome

<table>
<thead>
<tr>
<th>Signs &amp; Symptoms</th>
<th>Frequency (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Truncal obesity</td>
<td>96</td>
</tr>
<tr>
<td>Facial plethora</td>
<td>82</td>
</tr>
<tr>
<td>Glucose intolerance, diabetes</td>
<td>80</td>
</tr>
<tr>
<td>Gonadal dysfunction</td>
<td>74</td>
</tr>
<tr>
<td>Hirsutism, acne</td>
<td>72</td>
</tr>
<tr>
<td>Hypertension</td>
<td>68</td>
</tr>
<tr>
<td>Muscle weakness</td>
<td>64</td>
</tr>
<tr>
<td>Skin atrophy, easy bruising</td>
<td>62</td>
</tr>
<tr>
<td>Mood disorders</td>
<td>58</td>
</tr>
<tr>
<td>Osteoporosis</td>
<td>38</td>
</tr>
<tr>
<td>Oedema</td>
<td>18</td>
</tr>
<tr>
<td>Polydipsia, polyuria</td>
<td>10</td>
</tr>
<tr>
<td>Fungal infections</td>
<td>6</td>
</tr>
</tbody>
</table>

There is some debate about the diagnosis of depression in CS. Strictly within DSM-IV criteria, a diagnosis of depression cannot be made since there is a co-existing organic pathology. Dorn and colleagues (Dorn, Burgess & Dubbert, 1995; Dorn et al., 1997) suggest that the presentation of depressed mood in CS is different to non-endocrine major depression, and suggest the term ‘atypical depression’ to account for the observed symptoms of hyperphagia, hypersomnia, irritability and increased fatigue. They report that 18 of 33 patients (54.6%) at initial admission met DSM-IIIR criteria for a psychiatric illness, with 17 of those 18 meeting their devised criteria for atypical depression and only 3 patients also meeting criteria for major depression (Dorn et al., 1995). However, Sonino & Fava (1998) suggest that these results are at odds with other published studies, and may be accounted for by diagnostic confusion with other features of CS such as weight gain and fatigue, which occur regardless of depression.

Notable cognitive effects have also been identified in CS. Reviews of the available evidence (Belanoff, Gross, Yager & Schatzberg, 2001; Erlanger, Kutner & Jacobs, 1999) identify significant deficits mainly in memory and concentration, consistent with cortisol-induced atrophy of the hippocampus (Starkman, Gebarski, Berent & Schteingart, 1992). A recent study of 19 patients and matched controls using an extensive neuropsychological battery (Forget, Lacroix, Somma & Cohen, 2000) found a wider range of deficits in general IQ, visuo-spatial abilities, reasoning and concept formation, verbal and language performance, as well as nonverbal memory and attention. Half of a separate sample of 33 patients (Gotch, 1994) reported experiencing cognitive problems, leading to problems in daily living and job performance.
Diagnosis & Treatment

Measures of cortisol and ACTH concentration in blood serum and urine are a primary source of diagnostic information. Tests that challenge the HPA axis are also used—for example, small doses of dexamethasone suppress cortisol levels in normal subjects, but not in patients with CS. Bilateral inferior petrosal sinus sampling (a procedure for sampling blood at different locations) can be effectively used to determine the site of ACTH production. Imaging techniques such as CAT or MRI of the sella turcica (the site at the base of the brain occupied by the pituitary) can determine the presence of a pituitary tumour.

The mode of treatment is determined primarily by the disease aetiology. For the most frequent variant, pituitary-dependent Cushing’s Disorder, treatment typically consists of initial short-term drug treatment with cortisol production inhibitors (e.g. metyrapone, ketoconazole) or cortisol antagonists (e.g. mifepristone) in order for the body to restore its immune system function in preparation for surgery. This is followed by trans-sphenoidal surgery to remove the pituitary adenoma. This is the preferred surgical treatment option (Chee, Mathias, James & Kendall-Taylor, 2001; Boscaro, Barzon, Fallo & Sonino, 2001; Reitmeyer, Lee Vance & Laws, 2002) although trans-frontal surgery is also possible for tumours of larger size (macroadenomas). Repeated surgery, combined with radiation treatment or adrenal surgery may also be indicated if cortisol levels have not normalised post-operatively. Hypo-pituitarism, and the need for broad-spectrum hormone replacement, may ensue if the production of other hormones by the pituitary has been affected.
Course & Outcome

The disease process in CS is particularly insidious, with one study showing a mean of 5.4 years until definite diagnosis of CD (Flitsch, Spitzner & Ludecke, 2000). Fatigue, weakness, weight gain and emotional symptoms (particularly anxiety and irritability) were most commonly noted initially (Flitsch et al., 2000). These may result in misdiagnoses of rheumatological or psychiatric illness and ineffective symptomatic treatments.

Clinical outcome audits of surgical treatment of CD indicate a total remission rate of around 94% at two years, however the percentage of patients remaining in remission declines with time: 80% after 5 years, 78% after 7, and 74% after 10 years (Sonino, Zielezny, Fava, Fallo & Boscaro, 1996). Older age, higher clinical severity, and presence of depression are some of the factors that predict less favourable outcomes (Sonino et al., 1996). Reitmeyer et al. (2002) give remission rates of 91% when the tumour is small, quickly detected and entirely removed, falling to 50% for a large, invasive macroadenoma. Depending on the precise criteria used to determine relapse, Newell-Price (2002) claims rates as low as 14-17% following successful surgery. However, in one study, 42% of patients being followed-up suffered some degree of relapse within two years, though the overall mortality risk was moderately but not significantly elevated (Pikkarainen, Sane & Reunanen, 1999).

Following treatment, emotional symptoms generally improve. Drugs that inhibit cortisol synthesis (eg metyrapone), especially in combination with SSRIs, are considered
effective in reducing mood disorders (Sonino & Fava, 2001). One prospective study demonstrated pre-treatment rates of psychiatric diagnoses of 67% falling to 36% at six months and 24% at 12 months post-treatment (Dorn et al., 1997), but only when assessed by clinical interview, not by self-administered scales (Hamilton Depression scale, STAI and most of the scales on the SCL-90R and POMS). Interestingly, this study found no link between changes in measures of psychopathology and HPA axis recovery; also, 4 of the 33 patients did not have a psychiatric diagnosis either before or during CS, but did so following treatment.

Another prospective study of 25 patients (Kelly, Kelly & Faragher, 1996) found that the percentage which did not fulfil criteria for a psychiatric diagnosis increased from 19 to 68% following treatment. The authors concluded that ‘depression, anxiety and somatic symptoms all improved in parallel with the successful restoration of cortisol values towards normal’ (Kelly et al., 1996, p.791). However, there was no statistical analysis of comparing symptom ratings and cortisol values, thus this conclusion seems to be a much weaker inference from the pre-post design, rather than an actual finding. Taken together, these results suggest that links between hormonal status, observed mood and self-rated mood may be less linear than actually considered.

Commonly in CS, significant residual symptoms may persist even after successful treatment. It is noted that patients’ physical and emotional symptoms will remain for a period of years, with slow rates of improvement. Pituitary insufficiency, particularly of growth hormone, and other secondary problems may also contribute to symptoms of impaired health, fatigue and depression (Pikkarainen et al., 1999). This is reflected in
patient reports: six months after surgery, a majority of CD patients rated their state of
health as poor, unimproved or only slightly improved in one study (Flitsch et al., 2000).
In another study of 41 questionnaire respondents (with a median time of 3½ years since
diagnosis, range 1-10) only 18 (44%) considered themselves recovered and 34%
reported depression at the time of completing the questionnaire (Gotch, 1994). Similarly,
in another sample less than half of the treated patients stated they felt fully recovered,
and over half complained of persisting fatigue even after successful treatment
(Pikkarainen et al., 1999).

The reversibility of cognitive deficits has also received some attention. Belanoff et al.
(2001) review several studies that found restoration of hippocampal formation volume
following treatment, and one study (Mauri, Sinforiani, Bono & Vignati, 1993) that
showed significant improvements in verbal memory post-treatment4. However, a one-
year post-treatment follow-up of 13 patients (Forget, Lacroix & Cohen, 2002) found no
changes in cognitive function (except in one task of visual organisation), and the authors
proposed that long-term exposure to cortisol may induce long-term cognitive deficits.

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4 A related question is whether the surgical treatment itself has a negative impact on cognitive function. In
a group of patients with different types of pituitary adenomas (including CD), Guinan et al. (1998) found
no decline in general IQ post-treatment, however found that post-treatment anterograde memory deficits
were related to treatment method but not type of pituitary tumour. This suggests that memory deficits are
associated with surgical treatment independent of whether the target pituitary tumour was prolactin-
secreting, ACTH-secreting etc. Other studies of treatment effects on pituitary adenomas (Grattan-Smith,
Morris, Shores, Batchelor & Sparks, 1992; Peace et al., 1997; Peace, Orme, Padayatty, Godfrey &
Belchetz, 1998) employed retrospective designs and mixed groups, thus it is not possible to factor out the
effects of treatment versus tumour type.
Psychosocial Aspects of Cushing's Syndrome

Advances in the neuroendocrine understanding of CS have not been matched with equal attention to the psycho-social contributions to the course and outcome of the disease. Extensive literature searches of main databases (PsychINFO, Ovid, Ingenta and ScienceDirect) over the past 25 years (1977-2002), using reference list branching as well as keyword and topic searches revealed very few relevant contributions. This section critically reviews the available evidence.

Personality and Cushing's Syndrome

Hinting at more enduring personality influences on the course of the illness, Sonino & Fava (Sonino & Fava, 1998) note that, in their clinical experience, anxiety and irritability can often be present 'long before the onset of Cushing's Disease' (p.145). Sablowski, Pawlik, Ludecke & Herrmann (1986) explored such influences of personality in patients with CS. They administered a battery of personality tests (including the State-Trait Anxiety Inventory) to patients with pituitary adenomas (including CD) in order to 'find out changes or problems which demand special treatment' (p.8). They found that scores on the STAI normalised post-treatment across all groups, but otherwise found no particular pattern of traits to differentiate patients from controls. Flitsch et al. (2000) also failed to find significant differences between patients with CD, acromegaly or hormonally inactive pituitary adenomas on the majority of the personality test scales. Both studies, however, used personality tests much as a different type of outcome
measure, thus the question remains whether aspects of personality substantially interact with and influence the presentation of psychiatric symptoms, and by which mechanism.

**Patient Beliefs and Quality of Life**

A disjunction between physiological and functional status following treatment is frequently noted in the literature. Post-surgically it is recommended to patients that they exercise in order to regain physical fitness and muscle tone, which are lost due to the protein-catabolising effects of excess cortisol. In follow-up studies of post-surgical adaptation, functioning and well-being of patients, it has been observed that a substantial percentage of patients experience disability beyond what would be predicted by their post-operative medical condition, and rehabilitate poorly (Sonino & Fava, 1998). Depression, in particular, may persist for long periods, and its presence cannot be fully accounted for by neuro-endocrine and treatment variables (Dorn *et al.*, 1997; Kelly *et al.*, 1996; Pikkarainen *et al.*, 1999; Sonino & Fava, 1998). Factors such as ‘unrealistic patient hopes for cure’ (Sonino & Fava, 1998, p.43; Sonino *et al.*, 1996; Flitsch *et al.*, 2000) have been suggested as having an important role in this. This implies that beliefs could influence outcome, presumably by determining what health behaviours (e.g. avoidance versus returning to work) are enacted, as well as what emotions are activated (e.g. hope, relief, despair or anger). However a coherent psychological framework for the influence of illness beliefs on rehabilitation, quality of life and outcome has not been elaborated on further in the literature.
A Survey of Patients’ Experiences

One study (Gotch, 1994) aimed at supplementing medical accounts by exploring CS from the patient’s perspective. In this study, 41 patients with CS completed an open-ended questionnaire asking about their experiences of physical and mental effects, the effects on their family and work or school performance and whether they considered themselves recovered. Responses were categorised into themes, and frequencies of similar experiences were calculated.

The data suggest that patients experience CS as very serious and disabling: 91% classified its impact as affecting their lives ‘greatly’ and ‘a lot’. Amongst physical complaints, the most frequent complaints were fatigue (85%) and changes in physical appearance, especially weight gain, (63%). Just one patient reported no physical impairments or changes. Mentally, emotional instability (61%), cognitive impairment (49%) and depression (32%) were the most frequently reported difficulties. Six people did not report any mental effects. In terms of functioning within their wider context, 80% of patients experienced family difficulties while 56% experienced disruption of their work or learning. In terms of coping and improvements in services, patients identified education (56%), maintenance of hope (41%) and prompt diagnosis (32%) as helpful. For those who considered themselves recovered (44%), time to recovery took a mean time of 11 months, and several expressed fears of recurrence.

This study presents valuable first-person descriptions of symptoms, which unequivocally demonstrate the seriousness of the condition for patients as well as vividly illustrating
the quality of their experiences (see quotes on pp.13-14 above). Because it prioritises patients’ categorisation and evaluation of problems, rather than a medical assessment, it conveys a more accurate picture of the lived experience of CS than do symptom frequency lists. This study is also significant in that it widens the scope of attention on disruptions in the family life of patients, an area not previously considered in outcome research. Finally, by eliciting patient’s views for the improvement of services, it also derives helpful suggestions for clinical practice, for example a specific request by patients for education about the disease process and recovery times.

However, despite the vivid quotations, this study is limited to only providing an essentially ‘thin’ description of the experience of CS. Patients are only asked to respond to predetermined categories of ‘mental’ and ‘physical’, and their responses are collapsed into frequencies. In the discussion, the author compares the frequencies obtained with those in other published quantitative surveys, with some discrepant results accounted for by underreporting. If indeed the purpose had been to provide accurate data about the effects of the illness on patients, a quantitative survey with multiple informants would have been a more appropriate vehicle.

Thus, although these descriptions are a useful starting point, there is much more to learn about the psychosocial aspects of CS. It is important to consider experiences not simply as direct effects of the illness, but as complex interactions between the illness and such factors as personal understanding and coping, context and relationships. Further research could also adopt a framework that would give priority to patients’ understanding and meanings and allow ‘thicker’ descriptions to emerge (Charmaz, 1999).
An Autobiographical Account

A subtle interplay between physical health, psychiatric ‘symptoms’, subjective experience and relationships is illustrated in Armstrong’s (1991) autobiographical account of CS. His vivid descriptions are in marked contrast to the accompanying medical account by his treating physician, and expand the frame of reference—for example, he describes how symptoms of irritability were reinforced within family interactions. He also clearly identifies his spiritual beliefs as accounting for his pre- and post-operative composure, even at times when symptoms and loss of function increased:

‘As a result of the progressive sickness, the frustrating delays [in diagnosis] and the fluctuating hormone levels, I experienced extreme mood swings. I felt helpless. During the period of deepest depression, I resigned myself to the fact that if I were to die from this disease, I needed to trust in the will of my God. With this decision, I experienced a sense of peace.’ (p.9)

Describing his recovery, Armstrong also makes specific reference to psychological techniques (relaxation and cognitive-behavioural therapy techniques), social support, and personal beliefs that his experiences had meaning and could be turned into useful knowledge:

‘My belief that this suffering would have meaning for my life and that I would be able to use this illness to assist me in my calling enabled me to focus my energies on recovering from the surgery’ (p.9)

Notably, despite what objectively would appear as a relatively swift process of diagnosis and treatment (six months between first symptoms to surgery, compared to the average of around 5 years; Flitsch et al., 2000) and a positive outcome, Armstrong strongly
described the despair that unmet expectations created. Two months after first noticing the symptoms and being diagnosed:

‘....began the stage of my illness that I can best describe as descent into hell ... The decision [by the doctors to do further tests] was difficult to accept. We had travelled to hospital many times before with the expectation that we would be given a conclusive decision about the condition and its treatment.’ (p.9)

This account highlights the importance of examining a person’s illness experience in its social-relational context in order to adequately understand its impact, and make better predictions about its outcome. The individual’s beliefs and expectancies are also highlighted as central to both emotional state and self-directed health behaviours. Finally, this account poses the question of the efficacy of various psychological treatments in supporting people with CS.

**Psychological Treatments**

In their study of outcome in a series of 33 patients, Dorn *et al.* (Dorn *et al.*, 1997) note that 6 patients saw a therapist on a ‘routine basis’ and psychological intervention was recommended to as many as 12 after remission of hypercortisolism (p.918). Mental health referrals were identified by 6 (15%) of the respondents in the survey by Gotch (1994) as potentially helpful to all patients with CS. In a pamphlet for patients published by the UK Pituitary Foundation, the mental health aspects of CS are summarised and the potential of psychological treatments for depressive symptoms is suggested (Pituitary Foundation, 2003). However, literature searches revealed no reports of psychological therapeutic approaches with people with CS, either adaptations of existing therapies or consideration of particular issues in this population.
Developing a Psychosocial Perspective on Cushing's Syndrome

Although from the existing studies there are several indications of the scope and clinical utility of researching the psychosocial aspects of CS, theoretically and methodologically coherent studies of psychosocial aspects have not been undertaken. This is in contrast to other major physical health conditions (e.g. cancer, epilepsy, stroke, heart disease, chronic pain, etc; Baum, Newman, Weinman, West & McManus, 1997) where there has been a significant volume of psychological research. The following section will provide an overview of the main concepts and topics that have been investigated, in order to frame and inform the current investigation. Given the volume of this literature, this overview will briefly highlight key themes, drawing on reviews from within health psychology (Maes, Leventhal & DeRidder, 1996; Ogden, 1996; Schneiderman, Antoni, Saab & Ironson, 2001; Taylor, 2003) as well as medical sociology (Charmaz, 1995; Charmaz, 1999; Pierret, 2003) and quantitative as well as qualitative data. Specific theoretical models and relevant evidence will be reviewed in later sections.

Perspectives on Chronic Illness

If, following Charmaz (2000), we define illness as the experience of disease, the two processes may not start contemporaneously. One has first to notice the discomfort and label the symptoms, then search for explanations. The path from discomfort to diagnosis is very variable, and may extend from immediate diagnosis to a 'diagnostic limbo' of unclear tests and ineffective treatments. When symptoms have been persistent, intrusive yet vague, sufferers may welcome a 'serious diagnosis' that will remove the burden of
being doubted, dismissed or discredited, and the uncertainty and anxiety. The experience is different when disease appears acutely, and the shock and crisis forces a constriction of time, suspension of 'normal life' and a big mobilisation of resources.

Developing a realistic understanding of the disorder is a core task of the illness experience. How the illness is understood, what is expected, and what procedures can be used to manage the situation are key components, and have been found to be particularly important in a range of conditions (reviewed in Petrie & Weinman, 1997). For example, in a qualitative investigation of representations of Congestive Heart Failure (CHA), Horowitz, Rein & Leventhal (2003) found that suboptimal daily management, and alarmingly delayed responses to crises could be attributed to patients having an acute, rather than chronic, model of the illness, and did not reliably connect symptoms with their illness. Another example is a study on recovery from heart attacks (Petrie, Weinman, Sharpe & Buckley, 1996), which demonstrated that beliefs about seriousness, rather than physical status, predicted return to work, and that those who believed the illness and its causes were controllable were more likely to attend rehabilitation classes.

Heijmans (1999) investigated illness representations, coping and outcome in Addison’s Disease, a disorder of adrenal dysfunction leading to hypo-cortisolism, which biochemically is the 'reverse' of CS. This disorder presents with vague and non-specific symptoms such as weakness, fatigue and weight loss and is treated through long-term replacement medication, which requires that patients are involved in their self-care for the long term. Two distinct clusters of representations were found: a high-seriousness cluster, in which patients saw the illness as more serious, chronic, less controllable, and
with more consequences, and a low-seriousness cluster. Moreover, illness representations were found to be more strongly related to adaptive outcome than was the type of coping.

Ways of coping with illness have also received attention in the literature (DeRidder & Schreurs, 2001; Maes et al., 1996). Strategies have been classified into categories such as seeking social support, problem-focused, emotional-focused, cognitive escape/avoidance, positive reframing and many others (Taylor, 2003). Although proactive, problem-solving strategies are generally related to better adaptive outcomes, what type of coping is more effective depends greatly on the specific context, person and disorder. For example, emotion-focused coping is often more strongly associated with adaptive outcome than problem-focused coping in some types of stable, chronic illness (Maes et al., 1996).

Being diagnosed with a disorder may be met by a host of cognitive and emotional consequences. Denial and minimisation have been observed to be common responses which have the potential to cause long-term complications—for example, by delaying treatment—but may equally help protect a person from overwhelming anxiety and uncertainty in the short term. Anxiety is evoked when people expect a major impact on their lives, yet have little control over the consequences and processes. The realisation and burden of impairment can cause depression, withdrawal and negativity which may in turn have a significant impact on how the person engages in rehabilitation. Though there have been several attempts to create stage models of reaction (e.g. Kubler-Ross,
1970), it is generally acknowledged that different emotional responses may emerge intermittently at different points, or persist for extended periods.

Doctor-patient communication throughout the illness is a crucial factor of outcome, quality of life and patient satisfaction. Issues of trust, rapport, continuity and control have been the focus of considerable attention (Kreps, Arora & Nelson, 2003). A contrast between 'paternalistic' and 'patient-centred' interactions in medical settings has been drawn, and recent policy promotes active patient involvement, choice and shared decision-making.

Serious, chronic illness ‘assaults the body and threatens the integrity of self’ (Charmaz, 1995, p.657). The impact on identity has been characterised as a ‘biographical disruption’ that challenges prior meanings, lifestyles and identities. It forces a person to ‘repeatedly rethink how they live and who they have become’(Charmaz, 1995, p.657). Self-esteem, goals, plans and dreams, roles and relationship balances may all be affected. The altered body and its relationship to the Self becomes particularly problematic: what was once taken for granted and invisible now becomes an enemy, a prison or an alien which threatens to take over.

The experience of illness is inextricably linked to the sociocultural context in which it occurs. How illness is generally conceptualised and what avenues for adaptation are socially sanctioned will all shape the person’s experience of and response to illness. Resources, financial and social, may buffer the damaging effects of illness to a considerable degree, allowing a person to ‘withdraw gracefully’, delegate tasks and
maintain the semblance of a previous normality. Conversely, illness can be even more serious and damaging when people already have few supports, little control, and live in deprivation (Hezlich & Pierret, 1987; Gallant, 2003).

Drawing on decades of interviewing people with a variety of conditions, Charmaz (1995) describes adaptation as one possible way in which people live with chronic illness. In her analysis, adapting implies that one accepts impairments, acknowledges limitations, and alters one’s life and Self in socially sanctioned ways. Adaptation involves engaging in trade-offs between different lifestyles and identities, and finally moving from struggling against, to struggling with the illness. The author describes this as a particular form of surrender, ‘flowing with the experience’ and not defining the illness as a battle to be won, but as an ongoing reality to be embraced: ‘adapting shades into acceptance’ (p.658). Taylor (Taylor, 1983) offers a different perspective, suggesting that coping with the threat of illness involves a search for meaning, a search for mastery and controllability, and a process of maintaining and enhancing self-esteem. Taylor argues that the creation, maintenance and use of ‘illusions’ -positive reinterpretations of reality- is a necessary and essential component of the process of adaptation.

Stigma, shame and blame has accompanied illness throughout history (Hezlich & Pierret, 1987). The ill person may be shunned out of fear (e.g. AIDS) or disgust (e.g. breast cancer), and blamed for the problems, their attitude to the illness or the lifestyle that supposedly caused them. What others believe about the illness will influence their attitude to the person, with implicit judgments being made about worth and need. Visibility and functional disability increase the stigma, and the ill person may find social
support evaporating and withdraw into isolation. Research has shown that peer support groups are effective in reducing isolation and building self-esteem as well as proving information about an illness.

A range of disorders such as Chronic Fatigue Syndrome (CFS) or idiopathic back pain share a pattern of vague but persistent symptoms, pain, fatigue, but as yet have no known source or identifiable tissue damage. In these so-called ‘functional’ disorders, the social stigma and professional disbelief significantly shape the experience. As well as coping with the limitations that the illness poses, and not being overwhelmed by disability, patients may struggle to be believed by doctors. In an analysis of narratives of back pain, Lillrank (2003, in press) highlights the struggle for sufferers to obtain a thorough investigation let alone a medical definition or cure. Moral labels -crazy, lazy or hypochondriac- followed cursory tests, and people felt disappointed, insulted and invalidated by physicians' attitudes to their pain. Glenton (2003, in press) suggests that, paradoxically, the need for a diagnosis increases dependency and consequently feelings of invalidation. Turning points away from the desperation and self-doubt engendered occurred when patients were actually listened to and taken seriously. When a ‘real’ diagnosis was given, patients experienced joy and relief, and a restoration to their status as ordinary, honest people.

Finally, the positive consequences of illness and disability are also increasingly being recognised (Thornton, 2002). Personal growth and revaluation, spiritual meaning, more empathy for others and renewed ‘love of life’ are not infrequently reported by patients.
Charmaz (2000) talks about how ‘earlier vulnerability becomes a source of strength as [people] redefine illness as a time for reflection, reassessment and redirection.’ (p.287).

The Distinct Context of Cushing’s Syndrome

As a serious, often chronic condition, CS raises potentially similar issues for patients, and similar psychosocial factors could be important. However, it is also a uniquely complex disorder, as it combines a long, insidious process of increasing dysfunction and distress, with a (frequently) specific surgical treatment. Psychological symptoms are prominent throughout its onset, not just after diagnosis, and are primary effects rather than overlays or secondary consequences. After treatment, restoration takes considerable time, may not be complete and may require long-term medication. The potential for recurrence, which increases with time, also means that there is no well-defined end-point. In effect, CS combines elements of acute, chronic and functional types of illnesses. As a result, it is not clear at this stage how existing models about illness experiences, beliefs and adaptation would apply to CS. It is possible that different factors or configurations are influential in CS compared to other disorders. Equally, the distinct course of this illness may have a distinct impact on the sequence with which such factors apply.

Arguably, the most appropriate research strategy at this early stage of psychosocial research in CS is an inductive, hypothesis-discovering approach. This can uncover and describe important variables, suggest interactions and causal pathways and build a more complex and detailed picture than available at present. Such research would build on
existing knowledge, but also considering the distinctive features of CS and their possibly differential impact. This could then serve as a confident basis for better-targeted, more relevant research into this specific illness. Indeed, such an approach has been advocated for the study of psychosocial aspects of cancer and other illnesses (Davies, Hall, Clarke, Bannon & Hopkins, 1998; Waxler-Morrison, Doll & Hislop, 1995).

A broader argument in support of this approach refers to the tension between methods that 'pull apart' as opposed to 'considering the whole' object of the investigation. Crossley (2000) argues that within the domain of health psychology, the dominant experimental method of isolating variables risks producing a fragmented view of people's experiences of illness. This is not simply an issue of increasing the sophistication of measurement, using more powerful multi-variate statistics and large samples; it is an argument about taking context and personal meaning into account. Crossley claims that strictly experimental approaches “fail to address a crucial, perhaps the crucial psychological dimension of pain, stress and disease: how humans experience, interpret and live with them” (2000, p77). Smith (1996) and Charmaz (1999) similarly argue that what is required to redress the methodological balance is an idiographic, experience-centred perspective, which can be best be achieved through qualitative methods.

Consequently, in order to achieve the research objective of developing an initial account of the psychosocial influences in CS, a phenomenological approach will be adopted. This approach values and prioritises first-person experiences and allows the researcher
to adopt a broad, exploratory view without applying premature or arbitrary limits. The following section will describe the core concepts and principles of this approach.

**Phenomenology**

Phenomenology as a philosophical movement is primarily associated with the ideas and work of Edmund Husserl (1859-1938) and Martin Heidegger (1889-1976). Derived from the Greek term ‘φανώμενα’ (phaenomena — things as they show themselves, as they appear), it is concerned with the study of human consciousness and perception of the world. The core argument, and key tenet, of phenomenology is that there exists an ubiquitous interpretational, meaning-ascribing process between an object and our perception of it (Spinelli, 1989). In other words, what we as humans perceive as reality is our experience of the world filtered through our mental processing, rather than the world itself. For example, we do not perceive an art painting as simply a pattern of shapes, textures and colours — factors such as its location, personal meaning, emotional impact and cultural significance, to name but a few, will automatically and fundamentally come into play to produce an overall experience of the painting.

At the same time, phenomenology does not deny the existence of a real world, which is independent of our perceptions or existence. Neither does it deny that people may share large portions of their assumptions and ‘filters’ with other people, or that some perceptions and interpretations are more likely, useful or dominant. It fundamentally posits that ‘we never perceive only raw matter; just as, similarly, we never perceive only
mental phenomena - we always experience the interaction between the two' (Spinelli, 1989, p.9).

This philosophical premise led to the development of the ‘phenomenological method’, a set of procedures aimed at identifying and clarifying the interpretations we layer upon the world. Husserl, and the school of transcendental phenomenology ultimately intended this method to be used in order to ‘strip away the variants of experience, and so to arrive at a clearer understanding of its invariants’ (Spinelli, 1989, p.18). In psychological research, the phenomenological method has been applied principally with the aim of scrutinising, as clearly, fully and openly as possible, the conscious experience of the individual, as free from presuppositions as possible. An example of this approach is vividly illustrated in the following excerpt of a study of depression:

‘Depression is experienced as the stoppage of time, the emptiness of space, and the reification of others. Time stops: development of myself, of situations and of relationships all grinds to a halt. Everything appears static, dead, with no change except a progressive deterioration like rusting or rotting. Most of all, the future ceases being really future, really new, unknown, fruitful. Rather, the future seems to promise only a dreary repetition of the past …’ (Keen, in Moustakas, 1994, p.97).

The Phenomenological Method

Spinelli (1989) describes the three fundamental, interrelated steps of the phenomenological method:
Epoché - Also known as bracketing, epoché (from the Greek ἐποχή - to abstain, stand back) refers to an investigatory attitude of ‘setting aside predilections, prejudices and predispositions and allowing things, events and people to enter anew into consciousness, and to look and see them again, as if for the first time ... [it is] a way of looking and being, an unfettered stance’ (Moustakas, 1994, p.85). It urges us to identify and suspend, or place within notional ‘brackets’, our expectations and assumptions, and proceed with an ‘open mind’. It is acknowledged that there are very real and practical limits to how many of one’s biases and assumptions one can set aside, how completely, and how consciously; however, the investigator is directed to make continual efforts in this direction. Furthermore, though it is clear this effort can never be complete or final, the very effort of trying to do so means one is more aware to such influences, and more reflective of his or her own position.

‘Describe, don’t Explain’- Following the first step of opening up awareness and setting aside expectations, the second step of the method requires the investigator to avoid any impulse to organise, reduce, explain or theorise about what is observed and experienced. Instead, the investigator is directed to describe, as fully and comprehensively as possible, every aspect and feature of what appears to him or her. Again, it is acknowledged that there are pragmatic limits on how far one can persist in purely describing. However, the net effect of a conscious effort to do so is a fuller, richer and more extensive description of the object of inquiry, one that may well have eluded the investigator who would hasten to apply existing models and theories.
Horizontalisation - Having obtained as complete a description as possible, this step directs the investigator to initially pay equal attention to every aspect of the data, and treat each as having equal value and significance. After due consideration, data will be organised in some way, and judgments can and will be made about which aspects are more central, common or significant. But by avoiding doing this prematurely, the researcher is less likely to prevent new, unexpected ideas and configurations to appear, and less likely to arrive at pre-ordained conclusions.

Taken together, the above aspects of the phenomenological method suggest a particular attitude to research - one characterised by openness, self-reflection, patience and restraint - rather than any specific set of actions or protocols (Moustakas, 1994; Spinelli, 1989). It is distinctly different to the 'natural attitude' of forming quick judgments of people and situations, proceeding on the basis of existing schemata and accepting the 'way things are' without question, which we employ in our everyday life. It is also different from a strictly 'scientific' attitude to research in that it does not proceed by testing specific hypotheses and does not accept the premise of an external 'objective' stance for the investigator.

Points of Origin and Destination

The phenomenological method aspires to provide a 'fresh look', and to start from a basis of broadly, fully and without preconceptions consideration of a research topic. However, as mentioned above, this contains an irreducible tension within: what one selects to study, where, and how, has already been influenced by existing beliefs, theories and
attitudes which the researcher, and the system s/he is part of, subscribe to. Although an investigator who seeks to use the phenomenological method ought not to specify hypotheses to *apply* to the data, it still is the case that s/he will select, then approach a particular domain with some aims, beliefs and biases. Moreover, research must also be a pragmatic enterprise, with start- and end-points, useful directions, and external controls. As argued above, there is a considerably body of research knowledge about people’s experience of serious illness, which can be usefully brought in to guide this study.

The only possible response to this tension is an effort to specify these aims and influences as clearly and thoroughly as possible. Firstly, the investigator’s broad allegiances, interests and beliefs must be identified and, as far as possible, set aside. Secondly, the theoretical models that the researcher has drawn on must be clearly specified. Such theories can be used to set down clear and specific questions that will provide a focus for the research without necessarily demanding adherence to the particular theoretical model. It will then be easier and more effective to bracket these preconceptions, as well as render the researcher’s position more open to scrutiny.

Thus, in the following sections, the main theoretical frameworks guiding this inquiry into the psychosocial aspects of CS will be described: The Common-Sense Model of Illness Self-Regulation by Leventhal and colleagues (Leventhal, Meyer & Nerenz, 1980; Leventhal *et al.*, 2001), and the concept of Possible Selves by Markus & Nurius (1986). These theoretical models will be used to derive specific research questions, which will guide the collection and analysis of data. A further section will address the influences and allegiances that have shaped the formulation of this study.
The Common-Sense Model of Illness Self-Regulation

One of the most dynamic and flexible models about how people understand and respond to illness has been proposed and elaborated by Leventhal and colleagues (Leventhal et al., 1980; Leventhal, Nerenz & Steele, 1984; Leventhal & Diefenbach, 1991; Leventhal, Diefenbach & Leventhal, 1992; Leventhal et al., 1997; Leventhal, Idler & Leventhal, 1999; Leventhal et al., 2001). The Common-Sense Model (CSM) proposes that illness is understood through an implicit matching process between sensations and mental representations (schemata) of illnesses, which are then responsible for activating selected, targeted coping responses, generating feedback that evaluates, and shapes, the illness model. This model has been very influential in the field of health psychology (Ogden, 1996) and has received empirical support (Hagger & Orbell, 2003).

In the following sections I will describe the main influences in the development of the CSM and its main components. I will then address the particular strengths of this model, and discuss how it can provide a framework for this study.

Background

The initial impetus for Leventhal and his colleagues (Leventhal et al., 1980) to develop the CSM came from studies of how people responded to information about illness threats in health promotion and education. They proposed a ‘parallel response’ model which suggested that the mental processing of information (a cognitive path) and fear (an affective path) occurred in parallel, and were controlled by a self-regulating
feedback loop. Over the course of considerable elaboration and extension, a broad, general framework of illness understanding and adaptation emerged. The CSM describes a continual process of dynamic adaptation, in which a person’s understanding of an illness, or the meaning he or she attributes to a symptom, leads him or her to use specific skills and engage in particular actions to prevent, cure or adjust to that illness (see Figure 2, p.45).

Components of the CSM

What counts as a Stimulus?

The CSM initially referred to how individuals respond to a rather narrow range of external stimuli. These were social stimuli, such as being told something by a friend, public health messages on a billboard, information given by a nurse or doctor, as well as more concrete sources such as health information in a magazine, labels on a cigarette pack, etc. Note that the term ‘stimulus’ was used in the behavioural sense of a discrete, observable entity rather than the cognitive sense of a mental event. As the CSM was more generally applied, it was expanded to include reactions to internal stimuli, such as bodily perceptions, perceived signs and symptoms, thoughts and feelings.

Illness Representations

The component of the model that has received the most attention and elaboration is that of Illness Representations (IRs). When a person is faced with a stimulus, the CSM suggests that a mental process of cognitive representation will generate beliefs about its
nature, potential impact, etc. This set of beliefs is generated, or enlisted, to ‘understand what is happening’ – it is the ‘answer’ to the ‘questions’ that are posed by the stimulus. From the examination of both qualitative and factor-analytic studies, Leventhal and colleagues suggest that, although there is great variability in how people understand illness, their beliefs can be organised in five general dimensions or domains:

- **Identity** – ‘what is this?’ - the label given to the illness, and the symptoms, sensations, signs, deficits, etc, which identify the presence of illness.

- **Timeline** – ‘how long will this last?’ and ‘how will it progress?’ – beliefs about onset, course and duration, recovery period, etc. Typical timeline beliefs include *acute*, with a sudden onset and brief course, *chronic*, or *cyclical*, with a recurring pattern.

- **Cause** – ‘what causes it?’ – beliefs about internal (bodily) and external (e.g. chance, accident, pollution) causes of the illness, both biological (e.g. a virus or defective genes) and psychological (stress, personality, behaviour).

- **Control** – ‘is it controllable?’ – beliefs about the potential for cure or control of the illness, by one’s own behaviour, by means of medical or other intervention by others, by chance or luck, by ‘God’s will’, etc.

- **Consequences** – ‘what is its impact?’ – beliefs about its presumed, or predicted, impact on the total life, health and well-being of the person, both in the present and the future.

To illustrate with a simple example, a mental representation of the flu can include a perception that it includes a blocked or runny nose, headaches, fatigue, painful joints,
lack of appetite and fever (identity) a belief that it is caused by a common virus that is easily transmitted from person to person (cause), that it will last no more than a week (timeline), can be controlled with self-administered paracetamol and vitamins and will be combated by the body's own immune system, but not cured by antibiotics (control) and will mean bed rest, taking time off work, and a small expense on over-the-counter medication (consequences).

It is important to note that a common-sense model of an illness is not just seen as a simplistic or inferior version of a medical model of an illness. Although the CSM refers to the 'person-as-scientist', it does not imply that a person's common-sense models could or should be judged by medical scientific standards. Thus, in the above example, vitamin supplements may be included in a person's representation of what can control a flu, though this may not be based on medical evidence. Although people's understanding may clearly be biased, deficient or inappropriate, as judged by medical textbook standards, the CSM seeks to understand the beliefs and perceptions of each person as they stand, and as they guide him or her to action.
Figure 2. The Common-Sense Model of Illness Self-Regulation

Health threat stimuli, both external and internal, generate both an emotional reaction and a parallel cognitive representation. These set the goals for adaptation and guide the selection and enactment of response procedures. An evaluative process then compares the achieved with the desired state, and provides feedback on the basis of which representations and emotional reactions are revised.

Adapted from: Brownlee et al, 2000, p.403; Leventhal et al., 2001, p.21.
Revisions of the CSM have suggested that IRs are a type of schema, in many ways similar to other schemata that have been described in the social-cognitive psychology literature (Kihlstrom & Cantor, 1984; Kahneman & Tversky, 1982). For instance, they may form hierarchies of increasing levels of abstraction (e.g. a more general concept of 'virus' may have instances of flu, HIV, syphilis etc) and may be more or less explicit and conscious at any one time. As with schemata in general, IRs are subject to constant revision by new information, but may also be resistant to revision under certain circumstances.

Furthermore, Leventhal et al. (2001) suggest that, within a certain national, ethnic, cultural, family or other group, typical 'templates' of IRs will be seen as 'common-sense', and adopted by their members. Individuals will then, when faced with an illness, draw on these socially available 'templates', as well as their individual experiences of illness, general knowledge, personality, mood, and many other factors, to generate their own particular IR. Clearly, the CSM does not seek to specify how an individual arrives at a given set of beliefs, rather suggests that at any one time, that individual will hold a set of beliefs about identity, cause, consequences, etc, which will lead to the selection and performance of particular corrective actions.

An additional dimension of coherence has recently been suggested (Moss-Morris et al., 2002). It refers to the person’s beliefs about the 'closeness-of-fit' and congruency between the illness as perceived and the overall illness model. For example, a person may have low coherence beliefs if, after several months of trying various medications prescribed by the doctor, symptoms still fluctuate, and 'the whole thing still doesn’t
make sense'. The authors suggest that coherence is a meta-representational dimension, that is, a representation about a representation, which could be useful in research on disorders with fluctuating, vague symptoms.

Emotional Representation

Less attention has been focused on elaborating the affective component of the response to a health threat. Leventhal and colleagues have described this component variously as 'representation of fear', 'emotional representation' or 'representation of emotional reaction' (Brownlee, Leventhal & Leventhal, 2000). In the original 'parallel response' model, it was intended to represent the immediate, automatic emotional reaction (commonly of fear or anxiety) that occurs in parallel with a search for cognitive meaning, and then triggers particular processes to deal with these emotions. Brownlee et al. (2000) argue that 'automatic, affective reactions such as the pain and distress induced by severe injury... can activate coping responses prior to complex, interpretive reactions' (p.396). This component does not refer to the longer-term changes in mood that can occur in the presence of major or chronic illness.

It is worth noting that this model suggests the emotional reaction is not solely consequent on, and derivative of, the cognitive representation, but occurs in parallel with it. Indeed, the initial figures of the parallel process model did not include bidirectional vertical arrows linking these two processes, as do latter revisions of the model. The updated model thus suggests that the 'meaning' of an illness for a person can affect his or her emotional response, and also an emotionally (physiologically) aroused
state can influence the cognitive components of attention and processing that lead to the representation.

Bilevel Representations & The Symmetry Rule

A core feature of the CSM is the proposed bilevel nature of representations, both concrete and abstract. Abstract components of representations include verbal descriptions or labels, for example ‘flu is caused by a virus’ or ‘smoking causes cancer’. Concrete representations include somatic feelings, sensations and images, such as the memory of the feelings in the body generated by a fever, or an image of a blackened cancerous lung from a medical documentary.

Leventhal and colleagues (2001) argue that illness representations include both abstract and concrete components. Thus, the representation of time-line will include both ‘clock time’ and ‘felt time’, beliefs about consequences will include verbal descriptions (e.g. ‘I will need to use a wheelchair’) as well as images, real or imagined, of what it is like to be in a wheelchair. The CSM proposes that abstract and concrete representations are linked symmetrically: people will apply verbal labels to felt symptoms, and will identify and assign felt symptoms to a given label. The apparently paradoxical labels-to-symptoms route is suggested by experimental studies in which subjects given a false diagnosis then reported higher levels of symptoms typically associated with that diagnosis, despite no change in their objective physical status (reviewed in Brownlee et al., 2000). It is assumed that the symmetry of symptoms and labels is an effect both of past personal experience of an illness (i.e., the combination of ‘what it felt like’ and
'what it was called') as well as a fundamental property of the representational system (Leventhal et al., 2001, p.388).

**Procedures**

Procedures within the CSM include virtually any response that is enlisted, implicitly or explicitly, to prevent, cure or adjust to a health threat. They include the full range of 'coping strategies' and 'illness behaviours' that have been described in the literature, such as self-examination, going to a doctor, taking or stopping medication, using drugs or alcohol excessively, praying, denying that a threat applies to oneself, joining a support group, and so on.

However, Leventhal and colleagues (Leventhal et al., 2001) argue that the term 'coping' is too strongly associated with the various schemes of classifying responses into emotion-focused versus problem-focused, approach or avoidance, active or passive, cognitive or behavioural, etc. They strongly reject this approach as being led by psychometric, rather than theoretical considerations, and ignoring the true context and meaning of adaptive efforts. They argue that a coping procedure cannot be considered outside its context, and cannot be a-priori classified without reference to the specific illness representation it stems from.

The CSM proposes that any response may be a 'coping' response, if it is derived from the representation of the illness, and is aimed at altering some feature encoded in that representation. In keeping with the 'common-sense' theme, it is argued that coping
behaviours may not concur with biomedical understanding of 'optimal' behaviour, but rather will display 'common-sense' logic and internal consistency. For example, a person may be making a 'logical' decision to stop taking medication which he believes should have acted within a week, although his doctor, knowing that it takes a month for effects to become apparent, labels the decision 'non-compliant'.

Appraisal

The CSM suggests that an appraisal process follows the performance of any coping procedure. The targets and expectations set by the representation are compared to the outcome after the procedure is performed – for example, checking the intensity of a headache a few minutes after taking an aspirin. The results provide feedback about the accuracy and adequacy of the representation, and the utility of the coping procedures. On the basis of this feedback, representations may be altered, new procedures used or the existing ones maintained, and standards for appraisal reset.

This cyclical, dynamic process is best illustrated in the context of an ambiguous condition. The CSM suggests that when symptoms are vague and variable, procedures can serve as 'tests' to narrow down the range of options as well as eliminate the problem. As the symmetry rule suggests, labels are sought for symptoms, but there can also be a further search for symptoms when there is a potential label than must be confirmed or disconfirmed - an implicit question of 'am I missing something?' Consequently, when repeated adjustments, tests and medical consultations do not yield
any change, there will be an increasing vigilance for symptoms, and the person may start considering uncommon and alarming models to account for what is happening.

The mental representation of one’s own body, or ‘body-schema’, acts as a yardstick to representations of illness and guides appraisal (Leventhal & Diefenbach, 1991). The first cycle of appraisal, effectively, will be a decision about whether this is a symptom or just a sensation. Having an understanding of one’s body, how it operates and what its common sensations and states are, allows the person to discriminate between self and symptom.

**Utility of the Common-Sense Model in the Present Study**

In what way could the CSM provide a useful framework for this exploratory study of the psychosocial influences in CS? Firstly, it addresses questions and concerns that have, even at a tentative level, been identified in existing research. Both a first-person account, and researchers in the field have suggested that many of the emotional and behavioural consequences could be linked to how patients understand their illness. If, for example, prior to diagnosis a patient interprets the symptoms as indicative of a mental illness, this could lead to him or her seeking to avoid disclosing symptoms out of shame and fear of stigma. The CSM could be very useful in guiding the process of eliciting, exploring and understanding what beliefs about their illness people with CS do hold, and how these affect their adaptation and coping.
Secondly, though it appears to be a complex framework, the CSM nevertheless developed around a simple, intuitively appealing idea of people as active, 'commonsense scientists'. The CSM is thus broad and generic enough not to overly constrain a qualitative, phenomenological investigation. Thirdly, it is a dynamic model, with an explicit feedback mechanism. In a disorder such as CS, with an extended onset, confusing symptoms, and no real social awareness because of its rarity, patients are likely to struggle to understand what is happening, and will likely be reconsidering their ideas repeatedly.

The CSM also includes a metacognitive component, coherence, which provides a novel concept to consider how and why people decide that their representations are good enough, or decide to search further. This model also accommodates bilevel representations (abstract and concrete), thus allowing for conflict within a representation. This can be experienced as a 'gap' between knowing and 'feeling', e.g. 'Yes I know it takes time, but I can just see myself falling apart', which is a property that can be very useful when personal representation and diagnosis may be at odds. Finally, although applied research in the CSM is very much in a stage of development, it has shown the potential for interventions to identify and change maladaptive beliefs and facilitate adaptation and recovery in serious illness (Weinman & Petrie, 1997).

Overall, the CSM captures a key idea about people's response to illness, but also provides a complex analysis of its various process and components, and has achieved wide recognition and empirical support. The section above presented several reasons that support its utility at this stage of exploratory research, and appears to provide a
reasonable fit with what is known about CS. However, as will be argued in the following section, the CSM is less effective at describing how illness representations may interact with a person’s identity. To address this area in more depth, an alternative theoretical model will be described, followed by a discussion of how both models can contribute to specific research aims.

**Where is the Self in Self-Regulation?**

Most common everyday illnesses, symptoms, health threats and treatments do not pose much challenge to our self-concepts or identities. However, everyday tales and research studies of severe and chronic illness, stress and health adversity indicate that changes can be marked and lasting (Charmaz, 1983). Everyday stories about how ‘our neighbour became a totally different man after his heart attack’ or ‘aunt’s brush with breast cancer just totally changed her outlook on life’ feature as a striking part of our shared commonsense models of the effects of illness. This sense of identity change has been vividly described as ‘biographical disruption’ (Bury, 1982) and emerges frequently in the experiences of people with severe illness.

Recently, Leventhal and colleagues (Brownlee et al., 2000; Leventhal et al., 1999) have considered issues of identity change during illness with the CSM. In a footnote, they reflect on their haphazard inclusion of ‘Self’ in the title of the CSM, at times presenting it as ‘the CSM of Illness Cognition’, others as ‘CSM of Illness Self-Regulation’ (Brownlee et al., 2000, p.371). Until recently the CSM has paid more attention to the ‘self’ in a narrow, concrete sense of the body or one’s behaviour. The
CSM usually employs the term ‘self’ as a concrete reference (‘he hurt himself’) but makes little systematic reference to the ‘Self’ in the psychological sense of identity, self-concept or personality (‘I’m not my old Self’). Leventhal and colleagues suggest that ‘the self-system serves as an integrative mechanism at the base of the self-regulative action’ (Brownlee et al., 2000, p.394). They pose two questions: can the Self influence regulation without being altered in the process of self-regulation? How might the Self become a target for change? The following sections will describe how the CSM has been combined with issues of identity, to answer the above questions.

Definition of the Self in the Common-Sense Model

Drawing principally from the social-cognitive literature (Kihlstrom & Cantor, 1984), Leventhal and colleagues define the Self (alternatively termed the representation of Self) as the full set of mental representations that a person holds about him- or herself, generally similar to what has alternatively been described as ‘Self-concept’ or ‘Identities’ (Brownlee et al., 2000). They suggest that representations of the Self share the same structure and properties as illness representations, and can be described using the same five domains. For example, a representation of oneself as ‘a cruel, cold person’ can include beliefs that it originated because of harsh parental discipline (cause), makes intimate relationships difficult (consequences), is ingrained (timeline) and outside one’s control. Similarly, the Self will include abstract, propositional components (verbal descriptions of oneself) as well concrete, experiential components (images or memories of oneself). These representations will also be organised hierarchically, with broader and

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5 In this study I shall explicitly disambiguate the self as noun from the Self as adjective by applying distinct case, except when it appears in direct quotes.
more central schemata at the top, and more peripheral and situation-specific schemata towards the bottom. Notably, this definition of Self refers to a *plurality* of representations, thus rejecting the idea of single, global, context-free representation of oneself.

Regulation of the Illness & Regulation of the Self

Given this definition, how does the CSM see illness regulation and Self interacting? Does every act of regulation involve the Self? Brownlee *et al.* (2000) distinguish between three types of regulation. The first type is essentially problem-solving with the body as a target; it refers to the very basic, nearly reflexive operation of procedures such as adjusting one's posture to counteract muscle pain or tension. It is more akin to problem-solving about problems in the external world. The second, self-regulation, is the process where goals are chosen and set by the person, in relation to both the illness representation and the Self-representation. The Self, in a sense, works alongside the illness representation process, and influences the selection of goals and procedures. For example, this type of regulation would be involved in the case of a person who experiences heavy flu symptoms in the morning, however still goes to work for the day, postponing common-sense procedures for self-care because of an important business meeting and a belief in 'being tough, I'll make it through OK'. Brownlee *et al.* (2000) have also investigated how particular views of the self, e.g. as being in a high-risk group, can increase a sense of personal vulnerability, and influence symptom detection, illness perception and coping. In both these occasions, the Self is involved, but can hardly be said to be as deeply affected by the process of illness regulation.
It is the third process, Self-construction, that refers to the modification and reorganisation of the Self proper. In an analogy with the immune system, Brownlee et al. (2000) suggest that it is illnesses which present new, unknown challenges for the Self that will require its reorganisation and accommodation, with new roles, qualities and skills being needed. Analysing the first-person account of a person with cancer, they point out that changes in identity were moderated by loss of physical functions and roles, and suggest that reconstruction comes from the person’s active problem-solving efforts to accommodate to those losses.

The Social Context, the Self and Illness Regulation

Exploring another avenue, Brownlee et al. (2000) also connect changes in Self-representations with the way the social context interacts with the illness representations of the person. The CSM states that individual illness representation schemata may be drawn from family, friends and media, in essence deriving from a pool of available ‘common-sense ideas’ about illness. But beyond illness representations, identities may be affected when one’s representations of an illness are discrepant, and come into conflict with, those of others. How others see the illness may often have moral and personal implications that are hard to resist internalising. In a very simplified illustration, if a wife labels her husband’s fatigue as ‘laziness’ rather than illness, this is likely to become a feature of their interactions and recriminations; if the son interprets his father’s fatigue as ‘lack of interest’, this will lead to distance, and the loss of a valued

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6 One good example is the ‘vitamin-deficiency’ idea that has recently created controversy, with regulatory bodies arguing that the marketing of vitamin supplements exploits mistaken public beliefs about need, dose, efficacy and safety – ‘they are all natural, so you can’t have too much’.
role for the father; this person will be presented with these challenges regardless of what he may or may not believe about his symptom. As these researchers argue, the ‘fit’ between the patient’s and family’s representations may well impact interactions, relationships and ultimately identity.

Brownlee et al. (2000) point out that doctor-patient representation conflicts also have the potential for making significant impact on identity. This goes beyond the potential for ‘non-compliance’ when a doctor’s advice, based on a medical model, conflicts with the patient’s common-sense ideas. Crucially, the way doctors (and the whole healthcare system) communicate with and interact with the patient may help patients protect and sustain valued identities through the course of severe illness or may be experienced by patients as undermining and devaluing (e.g. loss of control, dignity, independence or Self-esteem) and undermining valued goals and futures. The example of ‘the tumour in bed 9’ is formulaic, but evocative.

Despite pointing to many ideas and promising avenues, however, Brownlee et al. (2000) put down no firm ideas: ‘it is unclear whether the self is involved as a relatively stable entity that moderates coping or as a set of identities and procedures that change from feedback generated from the management of chronic disease’ (p.399). No specific hypotheses or organising concepts are proposed, at best leaving the Self described as an intermediate ‘system’ between the CSM and the social context. The CSM does not support a coherent approach to such issues, leaving a host of important questions unanswered – why exactly is identity challenged by illness? Why are some changes resisted, some endured and others welcomed? What relationship is there between illness
representations and representations of Self? Leventhal and colleagues accept that ‘conceptualising these mechanisms poses a special challenge for theory’ (Brownlee et al., 2000, p.398). Equally, no experimental or applied studies of these issues have been undertaken from within the CSM framework.

In the next section I will present a complementary model of identity that also draws on very similar social-cognitive foundations as the CSM and provides more developed ideas about how the Self interacts with illness representations.

Possible Selves

A broad framework for thinking about identity is provided by Markus & Nurius’ (1986; Markus & Ruvolo, 1989) ‘Possible Selves’ (PSM). Drawing from the same social-cognitive literature as the CSM, they also describe mental representations (schemata) of Self as the constituent blocks of identity (or self-concept). Schemata are considered to be partly constructions and partly summaries of past experiences, relationships, social messages, media images, etc, and may be more- or less- well elaborated. As has been repeatedly demonstrated, schemata influence attention, information processing, memory, expectation and behaviour, functioning to reduce cognitive load while increasing the capacity to predict and control the external (and internal) world (Kihlstrom & Cantor, 1984). Schemata are organised hierarchically and flexibly, thus throughout the lifespan, various schemata will tend to become central and less context-dependent, while others more peripheral or more specific to situations. Markus & Nurius (1986) suggest a
‘working’ Self that consists of an array of selves, drawn from within the pool of those available to the individual, that is responsive to the person’s mood and context. Last but not least, it is because and through schemata of the Self that a person’s experience is imbued with meaning and continuity.

Markus & Nurius (Markus & Nurius, 1986) introduce a novel dimension by proposing that within their working Self people also hold schemata about the Self in the future. These ‘Possible Selves’ may be positive, for example picturing oneself as a successful, contented, powerful, or negative, for example poor, a failure, useless, lonely, mad. There may be ought (or ideal) selves that the person must achieve, and feared selves, that the person must avoid. More than simply propositional, abstract statements, possible selves are vivid, personalised renditions of one’s hopes and fears in technicolor: not simply ‘a failure’, but “sleeping rough, on the dole, a stinking alcoholic old man”.

The first function of possible selves is to provide motivation for goal-planning, adaptation and behaviour. Markus & Nurius (1986) suggest that a person will be motivated to pursue positive, valuable future possible selves, and avoid negative, threatening selves. They suggest that it is these possible selves that encode the cognitive aspect of motivation. They form vivid concepts of what the future can hold, and provide the target for our endeavours, rather than abstract goals and outcomes. For example, within the PSM, a desire for mastery would not be seen as an abstract disposition, or simply a belief, but as a vivid picture of oneself being ‘on top of’ various challenges and situations.
The second significant function is to provide what Markus & Nurius (1986) call the *evaluative context of possibility* for the working Self-schema. What we think of ourselves at a certain place and time, and how we feel about it, depends on what we believe possible for ourselves in that moment, and in life. A failure will be less injurious if it does not affect future hopes and possibilities, and a success will be less motivating if there are many negative possible selves to undermine it. Although in one sense possible selves can never be disproved, a person would likely react with shock and despair if it somehow becomes totally obvious that improbable but valued hopes and dreams are now impossible, or that one’s worst fears have come true. Markus & Nurius (1986) argue that the impact of events can only be accurately assessed if we have some knowledge of the person’s hopes and fears about themselves. For example, the impact of painful symptoms may be much stronger for the person who has ‘ill, disabled’ possible selves. Thus, possible selves both provide a basis for goal-directed action, as well as a means of evaluating the present Self, and the impact of life events.

This framework has been influential in shaping researchers’ understanding of Self and its impact on behaviour. Leventhal *et al.* (1999) indeed refer to possible selves as the concrete, imagery component of the timeline dimension in their definition of Self. The concept of positive future selves essentially parallels the concept of ‘preferred identities’ (Charmaz, 1987; 1995) which has taken a central role in the medical-sociological view of the illness experience. A number of studies have also provided a degree of empirical backing. Markus & Nurius (1986) provide evidence that possible selves have a significant and unique contribution to current self-image and self-esteem, stronger than the influence of *now* selves. Cross & Markus (1991) demonstrated how adults of all
ages can generate strong images of possible selves, arguing that these can act as psychological resources to aid adaptation throughout the lifespan. Another study supported the model’s implication that enhancing representations of the Self in a desired end state can affect performance by sustaining motivation (Ruvolo & Markus, 1992). Recently, Norman & Aron (2003) demonstrated that motivation to attain or avoid important positive selves could be predicted by how available, accessible and controllable that self was. Thus, a well-elaborated self that is retrieved from memory more quickly, and is perceived to be more controllable, will provide stronger motivation. The authors suggest that clinical implications of their findings could include orienting the initial stages of therapeutic engagement towards constructing, rehearsing and maintaining positive identities - in other words, possible selves that sustain hope.

Utility of the PSM in the Present Study

In terms of the present study, these ideas can provide a frame for understanding the interplay between CS, understanding and Self. An intrusive, encroaching illness such as CS could be a powerful stimulus activating many possible selves. We can speculate that the illness experience will generate very negative, fear-saturated possible selves that patients will be very motivated to avoid. Because past selves can serve as possible selves, people with strong, well-elaborated positive past selves of control, success and efficacy, will be in a better position to resist these intrusions and see ‘a different future for themselves’ than other people whose past selves are marked with failure, guilt, shame and hopelessness. Through the link between a self-schema and its impact on attention, goal-setting and information processing, we can see how a person’s
understanding of the illness will be shaped and guided by the types of selves (including the possible selves) active and available. Thus, a ‘relaxed and healthy’ possible self could influence my reasoning about my current symptoms, leading me to perceive the illness as having shorter duration and fewer consequences; conversely, a hopeless, helpless possible self could influence my perception of control over symptoms, and lead to a perception of the illness as endless and all-enveloping.

These suggestions indeed fit with what emerged from a qualitative investigation into changes in the Self for patients with chronic pain (Hellstrom, 2001). Aiming to investigate the temporal dimension to the Self that is suggested in the PSM, this research analysed in-depth interviews of 21 patients with chronic pain using a phenomenological method. Two of the emergent themes are particularly relevant: firstly, the struggle to keep alive and well past schemas of oneself as strong, healthy and active, and minimise the ‘losses’ of Self incurred due to the illness. Secondly, the sense of a locked-in, isolated and entrapped Self, with radically curtailed possibilities, surviving only day by day. This study did not address illness representations amongst patients, and thus draws no conclusions regarding possible links. However, the findings of this study vividly illustrate how identity, and particularly hopes and possibilities, can be affected by illness.

Overall, the PSM provides a useful set of ideas about links between representations of Self, representations of illness and adaptation during the course of illness. It proposes that illness will affect the self-structure through influencing the array of possible selves within the working self, which in turn will influence the representation of the illness.
This model provides a complementary set of theoretical tools to the CSM and has found empirical support, thus can be helpful in providing a guiding context for the present study. Finally, though the aim of this theoretical exposition has not been to provide a synthesis of different models, clearly it has pointed to specific paths in this direction.

**Personal Influences**

Before moving on to specifying the research questions, it is important to specify particular personal attitudes that have also influenced the choices and frames of reference of this study. Within the phenomenological method such *bracketing* is necessary in order to adequately put aside one's own perspective, and allow that of the participants to emerge with as little premature interpretation as possible. Equally, *owning one's perspective* is also a key validity criterion in qualitative research (Elliott, Fischer & Rennie, 1999) as it allows the reader to assess for him/herself to what extent the researcher's position may bias the data and its analysis.

Firstly, the reader may well be surprised at my choice of research area. *Whatever drew you to this?* is a question I have been often called to answer. CS is a very rare disorder, which even general physicians know little about and clinical psychologists even less, if anything. Yet, I remember learning by rote in basic biology in high school that 'the pituitary is the main relay and feedback station between the brain and the endocrine system', which excited my imagination at the time as a possible site of that ineffable, elusive mind-body interface. Thus, a broader, influential interest in research that brings
together the mental and physical, and addresses connections and interactions underlies this choice. I believe that CS could provide an excellent testing ground for investigations on the interaction of mental and physical states in stress & depression, in the way that neuropsychological cases studies of rare, localised brain injuries are potentially very instructive about universal functions of cognition and brain organisation. Yet could this motivation lead me to seek large, universal themes, and militate against listening to patients carefully and noticing the ordinary?

This leads to a second influence: allegiances. It poses the question, who do you want the results for? Some of the people who helped me shape this project and, crucially, recruit participants have been medical researchers and doctors in Endocrinology and Neurosurgery. I have benefited from their help and support, and, in a way, have committed to providing something useful for their practice. Could this not influence my attitude to what participants say about doctors? On the other hand, other people who helped me shape this project have been volunteers of the Pituitary Foundation, themselves Cushing’s patients, and psychologists in clinical health and health research. Their help has also been invaluable, so am I not somehow pulled to provide something useful and affirming for them too?

Such questions are, I believe, inherent in psychological research. The personal and institutional reasons for a research program, and the subtle interplay of allegiances cannot be taken out of the equation. I believe that the effort to maintain a reflective stance, supervision, and making one’s theoretical ideas and aspirations explicit can offer a buffer against personal agendas. I further believe that having multiple contributors
(stakeholders) and not working within an established, funded research project has the advantage of equalising allegiances and promoting neutrality.

Research Objective & Questions

As argued above, neither the personal experience of CS nor more general psychosocial factors regarding adaptation to this illness have been adequately described or explored by existing studies. Despite neuro-endocrine advances in understanding CS, a coherent theoretical picture of psychosocial influences has not emerged, and existing studies have generated little further research.

Research Objective

Thus, the overall objective of this study is to explore the experience of people with CS, in order to provide a better starting point for further analysis of the psychosocial determinants of CS:

*What are the significant aspects of people's experience of Cushing's Syndrome?*

Research Questions

It was argued that the most appropriate methodology for this purpose would be an idiographic, qualitative approach, which highlights and prioritises personal experience and meaning, without prematurely narrowing and rejecting possibilities. Thus, no
specific hypothetical predictions are specified or pursued. However, it was also argued that it would be useful to outline broader theoretical directions, to link in with previous research and knowledge, bring some pragmatic limits to this effort, as well as be transparent about existing theoretical biases implicit in our knowledge and training in psychology.

A major theoretical influence in the domain of people's understanding and adaptation to illness has been the Common-Sense Model. It suggests that individuals draw on socially available, individually constructed mental representations of their illness to direct and evaluate their coping efforts. Thus, this study asks:

*How do patients with Cushing's Syndrome understand their illness?*

The Possible Selves model elaborates the reciprocal influences of illness representations and identity. It suggests that an illness can affect identity through influencing the array of possible selves, which in turn will influence one's representation of the illness:

*How does Cushing's Syndrome -and patients' understanding of it- affect their identity, and how does identity influence representations?*

The following section will describe how these questions guided the study's methodological choices and procedures.
Chapter 2: Method

Overview

This study aims to explore the experiences of people with Cushing's Syndrome, in order to establish a basis for further research into the psychosocial aspects of patients' adaptation. This section describes the methodological implementation of a qualitative, phenomenological approach. It covers the development of the interview schedule through piloting, and describes the recruitment and interviewing procedures. Descriptive statistics and brief biographical details are given to describe the sample of participants. The chosen analytic approach and process are described. The validity and reliability checks which were employed are also described. Finally, a section discusses personal and professional influences on the methodological and analytic stages of this research.
Research Ethics Approval

Ethical approval was obtained from the Head of the relevant clinical departments, the relevant Hospital Ethics committee (see Appendix B) and the relevant NHS Trust Research & Development unit.

Interview Schedule Development & Piloting

A qualitative interview aims primarily at facilitating a personal story to emerge freely and openly, with the least constraint imposed by the interviewer. Simply put, it is concerned more with meaning and less with information. In keeping with phenomenological principles, the structure of the interview and phrasing of the questions must be designed to lead the person to describe, rather than explain, their first-hand experience, and must allow equal weight and time to all aspects of the experience. Much more so than in quantitative interviewing, engagement and the process of the interview is crucially important. The interviewer’s stance, as much as the language and sequence of questioning, must be carefully considered to allow participants the time, space and safety to share as much of their experiences as possible. As a result, the interview often takes on more of a form of a discussion, rather than a question-and-answer session. (Smith, 1995; Taylor, 1998)

This discussion needs to follow the participant’s lead in terms of what is important, rather than only cover the interviewer’s own agenda. Equally, however, specific research
questions have been outlined to guide and focus this study. The total experience of Cushing’s would be too broad and too complex to cover in any one interview; indeed, it would be probably overwhelming for participants as well. What is then required is a balance between an open, exploratory discussion and a more detailed focus on particular areas.

The final interview questions and probes were produced through an extended, iterative process. It was shaped by the research aims, but also by the feedback of pilot participants, senior researchers and clinicians in the field. The following sections will briefly describe some of the main points of this process.

**Initial Interview Schedule**

Four general domains of questioning were derived from the study aims, and included in the initial interview schedule:

- Questions aiming to gain an overall sense of the psychological experience of the person, and how it developed during the course of the illness, e.g. 'Could you tell me, in your own words, what was it like for you at the time?'
- Questions about the person’s understanding of the illness and its implications at different stages of the illness – pre-diagnosis, between diagnosis and treatment, and post-diagnosis, e.g. ‘What did you make of the illness at the time? How did you understand it?’
Questions about impact on identity at different stages of the illness, e.g. ‘Do you think that the illness, and how you understood it, made an impact on you as a person?’

Questions about the future, both the person’s expectations of the illness as well his or her expected challenges and identity changes, e.g. ‘Given what you told me you thought about the illness, and its impact on you, what do you expect will happen in the future?’

Following guidelines (Smith, 1995; Taylor, 1998; Crowley Jack, 1999), these questions were organised from the most general to the most specific, and various possible probes were prepared. The interviewer would thus ask for a general, broad overview of events, to get to know the participant’s story and most salient experiences – e.g. asking ‘Could you maybe start by briefly telling me the ‘story’ of your illness, up to now?’ - before moving to considering the particular issues of interest. Also, the interview was structured to consider the illness in three stages (pre-diagnosis, pre- and post-treatment) and asking the above questions for each particular stage – e.g.: ‘In the period since the diagnosis was made, how do you think that the illness affected your sense of yourself as a person?’.

Piloting

In order to assess the structure, appropriateness and coverage of this interview schedule, three pilot interviews were conducted. The Pituitary Foundation, a charity organisation that provides information and support for people with CS was contacted for assistance.
The Foundation offers patient support through a network of trained ‘phone buddies’, who are themselves people with CS. These individuals have extensive experience in dealing with queries, are prepared to share their stories with others, and have added knowledge of a wide range of stories of other patients. The Foundation suggested a list with the names of several such individuals, which is also publicly available on request from the Foundation’s helpline. An initial phone contact with three ‘phone buddies’ was followed by a letter enclosing the study Information Form (see Appendix C) and the study Consent Form (see Appendix D). All three individuals responded positively, and phone interviews were arranged. Interviews included an extended section requesting feedback and opinions on the questions themselves, their appropriateness for other patients with Cushing’s, and suggested inclusions and exclusions.

After each interview, the data obtained, participants’ feedback and suggestions and my personal experiences of conducting each pilot interview were carefully considered within research supervision. As a result, several rounds of alterations were made to the interview schedule. Firstly, it quickly became apparent that dividing the experience into ‘stages’ was a researcher-imposed division that was not reflected in how the pilot participants saw their experience. In addition, it interrupted the flow of the story, and unnecessarily compartmentalised the discussion. Secondly, the pilot participants found the experience a very positive one, and were very supportive of the approach. Clearly, by virtue of their role as ‘phone buddies’ they all had previous experience of telling their personal stories, but strongly believed that this process in itself is highly beneficial for

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7 The Pituitary Foundation can be contacted on 0870-774-3355 or online at www.pituitary.org.uk
patients, and that many people actively seek opportunities to speak and be heard. Thirdly, it became apparent that, though issues of understanding and identity came up frequently, being asked specific questions appeared to stunt the pilot participants’ own descriptions. Indeed, the richest, most immediate descriptions of the topics of interest were spontaneous, rather than in direct answer to questions.

A further observation which was taken into account was that pilot participants tended to talk of their experiences in a way akin to a giving a medical history, listing symptoms, complaints and treatments, and only few descriptions of personal experience. We considered that this could represent the default way that ‘experienced’ patients talked to doctors, thus we needed to adapt the interview in two further ways. First, to make more explicit this study’s non-medical focus and the interest in experience, and distinguish my role from the medical team more strongly. Second, in the interests of rapport, to allow participants to share this story in full before proceeding to ask about more personal thoughts and feelings which are less likely to have been aired in previous retellings.

**Revised Interview Schedule**

As a result of these considerations, the interview schedule was rethought and redrafted. The revised interview is centred around the opening question:

*Could you think back to the first time you noticed any symptoms, and can you tell me about your experience of the illness since then, in your own words?*
This allows the participant to convey their own story, their own meanings and concerns, and specify their own time-scale, while also allowing the interviewer to gain an overview of the person's experience. It also allows the interviewer to listen intently while the person speaks, thus concretely demonstrating the commitment to an open-ended interview, led by person's own story rather than the interviewer's questions. As argued above, it also allows the participants to start off with the most comfortable version of the story, and thus more be likely to feel contained and comfortable in the interview situation (Crowley Jack, 1999).

Rather than using specific pre-scripted probes, the interviewer would then in the first instance probe this story further, asking for clarifications, examples, picking up on striking descriptions, or following up on descriptions of issues of interest to the study. This would ensure that in the first instance the interview would be following the interviewee's lead, and only then use specific questions if the topics of interest did not come up. The following is an example of the type of clarification that was often used:

Nick: I spoke to various- doctor T and her group of registrars, and the SHO, and I learnt an awful lot more, and that's how I found out what they do as far as the surgery is concerned, on the pituitary gland. But I wanted to know, I'd rather know.

**Interviewer: Why was that?**

.. Because I could then understand it.

**And if you could understand it..?**

I could come to terms with it.. Not understanding is something I- I needed to know, if I don't know, and I cannot come to terms with it.

**When you say come to terms..?**

Come to terms with how they are going to do the operation, what it can do to achieve, and what, you know, I'd rather know if somebody is going to have to do something, behind my nose, getting the tumour out or whatever it might be, poking into my skull, then I wanted to know. But- it was a shock to obviously end up in hospital- but it was much better to know that.. I was now, hopefully, if everything went successfully, on the road to recovery. Because beforehand, he had just been the downward spiral... perhaps I wasn't aware of it at the time, but looking back that's how I would analyse it.
Here, with repeated requests for clarification and reflection, it becomes more apparent
that by looking for information, Nick was trying to establish that there is ‘a road to
recovery’ which would take him out of the ‘downward spiral’ he had previously
experienced. Thus, by asking him to clarify the words ‘understand’ and ‘come to terms
with’, it emerged that creating hope was another function of getting information.

Clearly, such prompting needs to be responsive and immediate, thus cannot be pre-
determined. However, for the instances in which particular domains of interests for this
study did not emerge in the course of the interview, an Interview Guide with particular
prompts was prepared (see Appendix E). For example, if no mention of future
expectations of the illness were mentioned, the prompt ‘What do you expect will happen
with this illness in the future?’ would be used.

**Boundaries & Participant Protection**

Open-ended, unstructured discussions convey very rich data, but also are potentially
more sensitive for participants. Maintaining confidentiality, a solid sense of boundaries
and ‘taking care’ of the participant are indeed more important than collecting data
(Taylor, 1998). To this effect, the interview schedule also included introductory sections
where the interviewer explained the limits of confidentiality, the distinct role of the
researcher to the medical team, the likely time-frame of the interview, and the use of the
data in the research. It also reiterated the participants’ right to skip questions or stop the
interview outright, as they saw fit, without any adverse consequences or judgements. At
the conclusion of the interview, participants were invited to reflect on the process, its
benefits and strains, and ask any questions. Having now experienced the discussion, they were again invited to consider whether they did want a transcript and/or summary of the results, if they had previously declined. Finally, the intended uses of the data (for research, rather than a clinical purpose) were reiterated, and the right to withdraw at any point before publication was reinforced.

Procedure

Eligible patients were identified by a consultant endocrinologist and a consultant surgeon, both specialising in CS, from the clinical database. As CS is a rare disorder, even major treatment centres have few cases at any one time. Thus, to ensure an adequate sample, broad eligibility criteria were set out:

- Adult
- Diagnosis of CD or CS
- Treatment in past 3 years
- No other major medical conditions independent of CS

It was decided to establish a treatment date cut-off so as to interview participants as close as possible to their treatment, with the expectation that this would make the experiences, thoughts, feelings and expectations more vivid and available. 'Time since treatment', rather than 'time since diagnosis' was selected for three reasons: firstly, in CS diagnosis often remains uncertain for long periods, thus it is hard to establish a firm date; second, patients referred for treatment to the consultants who collaborated with this
study had already an established diagnosis from their local hospitals; thirdly, multiple and repeated treatments and recurrences are not uncommon, thus selecting by time since last treatment rules out the fewest people. The exclusion of other major medical conditions was made on the grounds that experiences and treatments for other illness could complicate analysis of the person’s experience in respect to CS alone.

Because both consultants accepted referrals nationally, and interviews were offered to participants in their homes, for practical reasons eligible patients were selected for accessibility, i.e. having home addresses in Greater London or the Home Counties.

An ‘invitation to participate’ letter from the consultants was sent to all potential participants (see Appendix F), with the study Information Sheet (see Appendix C), explaining the nature of the study and asking people to respond directly to the researcher. A response slip and a stamped, self-addressed envelope were included. It was explained in the letter that a follow-up telephone call would be made if no response was received within 2-3 weeks.

People who indicated on the return slip that they did not wish to participate would not be contacted again, and all contact details erased. No negative responses were however received. For people who returned affirmative responses, interviews were arranged at their convenience, at UCL or their homes. If no response was received, a reminder phone call was made, which allowed two participants who had changed address but were interested in participating to be included. People who could not be contacted by either phone or letter were excluded and all contact details erased.
The process of the interview started with an explanation of the nature and purpose of the project by the researcher, and an offer to answer any questions. In particular, the distinct role of the researcher to the clinical team and the person’s medical care was emphasised. The limits of confidentiality were explained, as was the fact that this study was part of a doctoral project in Clinical Psychology. Consent forms were then signed by both parties (see Appendix D). The Consent Form further offered participants to select if they wanted a copy of the interview transcript, and/or a summary of the results when it becomes available. A brief demographic information form (see Appendix G) was completed prior to interview.

The interview was audio-taped, and brief notes were taken by the researcher following the interview. These were intended to help with transcription but also included thoughts about striking themes that emerged in the interview. Following the interview, participants were again invited to ask any questions and discuss any difficult moments, or place any restrictions of use on the information that they disclosed. Backup plans were in place for the eventuality of needing to break confidentiality and/or seek help if participants clearly presented a risk to themselves or others, or were in immediate need of assistance. No such actions were necessary.

Participants

A total of 35 potential participants were identified as eligible by the Consultants. 20 of those had an accessible home address, and were sent invitation letters. None declined,
though 4 were unreachable by either phone or letter. Of the 16 who were interested in participating, 13 were interviewed, two had moved away and one withdrew due to illness. Nine elected to be interviewed at home, while four preferred meetings at University premises.

Two of the three pilot study participants, though outside the study criteria in terms of time since treatment (both had treatment 7 years previously) did meet the overall purpose of selection, which was providing very vivid descriptions of their experiences, and did not have other concurrent major medical conditions. Furthermore, initial analysis of the interviews suggested that the themes emerging were very vivid and similar to those of other participants. It was thus decided to include these two in the analysis. Unless otherwise noted, all analyses refer to the total sample of 15 participants.

The sample size is appropriate by qualitative research requirements (Smith, Jarman & Osborn, 1999) and for the purposes of this study. It also compares favourably with an estimated disease prevalence of 5 cases per million of population. The Pituitary Foundation, which keeps a national register, identified approximately 300 members with CD and 120 with CS on its system.

The mean age of participants was 43.8 (SD 2.85) years. Three participants were male and 12 female, a ratio of 1:4 that is broadly similar to the epidemiological data. 11 participants were white British, 1 white Irish, 1 black British Caribbean, 1 black British African and 1 Afghani. All had a diagnosis of Cushing’s Disorder, except one of the pilot participants who had the Syndrome (adrenal) variant. The mean length since the
most recent surgical treatment\(^8\) was 12.5 months (SD 9.5), with a median of 9 and a range of 2-36 months. Interviews lasted a mean of 94 minutes (SD 8.4; median 98, range 43-145). Transcript texts extended to a mean of 9900 words (SD 1100, median 10200, range 2800-17200)\(^9\).

Basic demographic details are presented in Table 3. Further biographical detail, in the form of brief vignettes for each participant can be found in Appendix H.

**Analysis**

A broad, phenomenological framework was selected for this study and guided the formulation of research questions, the construction of the interview schedule and the process of interviewing. Equally, a phenomenological framework guided the analysis of the interview data.

Several options of specific analytic approaches offer themselves, all drawing from the same common phenomenological principles: The Duquesne method (Giorgi, 1985; Moustakas, 1994), Grounded Theory (Glaser & Strauss, 1967) and Interpretative Phenomenological Analysis (IPA; Smith, 1996). For this study, IPA was selected from among these essentially similar alternatives because of its widening use within health psychology and previous application to similar research topics (e.g. Osborn & Smith, 1998).

\(^8\) Excluding the two pilot participants, thus N=13.

\(^9\) Interview time length was rounded to nearest minute, word-count to nearest hundred words.
Table 3. Participant Basic Demographics

<table>
<thead>
<tr>
<th>Name</th>
<th>Age</th>
<th>Sex</th>
<th>Diagnosis</th>
<th>Months since last op.</th>
<th>Marital Status</th>
<th>Ethnicity</th>
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<td>-</td>
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</tr>
<tr>
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<td>F</td>
<td>CS</td>
<td>-</td>
<td>Married</td>
<td>White British</td>
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<td>CD</td>
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<td>White British</td>
</tr>
<tr>
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<td>F</td>
<td>CD</td>
<td>9</td>
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</tr>
<tr>
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<td>F</td>
<td>CD</td>
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<td>Married</td>
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</tr>
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<td>CS</td>
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<tr>
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<td>CD</td>
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</tr>
<tr>
<td>Glenda</td>
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<td>F</td>
<td>CD</td>
<td>2</td>
<td>Single</td>
<td>White British</td>
</tr>
<tr>
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<td>M</td>
<td>CD</td>
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<td>Married</td>
<td>Black Br.-African</td>
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<tr>
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<tr>
<td>Corrine</td>
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<td>F</td>
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</table>

**Summaries**

<table>
<thead>
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<th>Mean</th>
<th>Median</th>
<th>Married</th>
<th>White British</th>
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<tr>
<td>43.8</td>
<td>12.5</td>
<td>66%</td>
<td>73%</td>
</tr>
</tbody>
</table>

1 Melaney and Fiona were initially recruited as pilot participants. Names and ages have been changed.
As described by Smith and colleagues (Smith, 1995; Smith, 1996; Smith et al., 1999) IPA is a methodological tool for making sense of individual meanings, perceptions and experiences, and prioritises these over facts and information. Equally, it accepts the phenomenological premise that the process of making sense of an individual’s account will always be subject to the researcher’s own perceptions and experiences. As a result, the researcher must try to balance the need to bracket his or her own assumptions in the course of analysis, but also engage his or her full knowledge and experience in interpreting and making sense of the data.

Following suggestions for the implementation of IPA (Smith et al., 1999), the analysis in this study involved the following steps:

1. The analysis started from a broad, quick reading of all interviews, to get an overview of the material. As I had conducted all interviews, I was already familiar with much of their content, however further readings did reveal new information.

2. The two pilot interviews, and six randomly chosen interviews formed one group of analysis. The other group were used at a later stage for verifying and expanding the analysis. This mode of analysis is recommended when there is a larger set of data than can be ‘kept in mind’ at any one time (Smith et al., 1999); additionally, it allows for an internal coherence check.
3. In the first group, each interview was initially read and analysed individually. Notes were made on the margin of each transcript regarding potential themes, significant remarks, ideas and questions. On a further reading, broader themes and concepts were noted on the other margin of the transcript. Themes were drawn both from participants’ own words as well as my labels and interpretations, to form the 1st level of analysis. This level of analysis was closest to the data, and most descriptive. Every effort was made to approach each interview on its own terms. I attempted to set aside ideas and preconceptions drawn from previous interviews, approach each with an open mind and allow the individual’s ideas to emerge, even if contradictory to previous analyses. Nevertheless, there is always the possibility that the first interviews could prime and activate ideas and analytic schemas which influenced the analysis of the latter interviews in the set.

4. For each interview, the initial 1st level list of themes, numbering around 20-30, was drawn up and illustrated with quotes (see an example in Appendix I). These themes were further considered, and organised into broader, 2nd level themes, which captured the essence of several underlying 1st level themes (see an example in Appendix J). This process was iterative, moving repeatedly between themes and text, to ensure that new ideas and themes were well grounded in the data. There were commonly around 5-10 2nd level themes for each interview.

5. 2nd level themes across all interviews in the group were brought together and considered as a whole. Noticing patterns, similarities, and divergences, a 3rd
level of themes was drawn up, to cover the most salient and powerful common themes that emerged across individuals. Checking back, and illustrating with quotes, again ensured that the analysis remained grounded, but this 3\textsuperscript{rd} level of analysis was more general, interpretive and abstract than previous levels (see Appendix K). Several less significant and common 2\textsuperscript{nd} level themes were set aside at this point. This judgment was not made on the grounds of frequency alone, but also considered whether the idea conveyed appeared significant or coherently related to other ideas.

6. The annotated, illustrated list of 3\textsuperscript{rd} level themes then formed the basis of the analysis of the seven transcripts in the second group. Each of these seven was initially read individually, with notes, questions and comments written in the margins. The analytic comments for each interview in turn were then compared and contrasted to the 3\textsuperscript{rd} level list, amplifying or clarifying some categories, or suggesting re-categorisations. This process, although more constrained, was still not simply a process of classifying the second set into the format set down by the first set. Rather, counter-examples as well as examples were sought, new evidence for themes that had been set aside previously was considered, and the general fit with the new data was critically assessed. Amendments and additions were made accordingly.

7. The amended list of themes was then illustrated with all relevant quotes and written up. The process of writing up, with its emphasis on pulling together a flowing, meaningful narrative of participants' experiences, suggested further
alterations and editing. From amongst several possibilities for organising and presenting themes, the organisation that would maximise what was distinctive about the experience of CS was selected over other equally possible options. Finally, the extensive list of quotes for each theme was pared down to the most relevant, powerful and evocative excerpts.

Validity Checks

Though it is recognised that the analytic method and process is inevitably influenced by the beliefs and attitudes of the researcher, it is crucial that the process of analysis is not entirely idiosyncratic to the researcher. Thus, a series of ‘credibility checks’ (Elliott et al., 1999) on the process and results of the analysis is necessary in qualitative research, and different types of checks can be used at different stages (Elliott et al., 1999; Popay, Rogers & Williams, 1998; Altheide & Johnson, 1994). The overall aim of these checks is not to establish a type of abstract inter-rater reliability, or any sort of comparison against an abstract gold-standard. Rather, it is a means of audit to ensure that themes are well grounded, supported by adequate illustrations and that inferences are valid. It is also a demonstration of accountability, allowing an external observer to follow the process of analysis step-by-step.

Three types of external verification were employed in this study. Firstly, during the earlier stages of the analysis, discussion with others helped shape the analysis. Emerging themes were presented to and discussed with the research supervisor, small and large
peer groups as well as postgraduate researchers from other disciplines (medicine & philosophy). This was an opportunity to discuss ideas, present tentative themes, get feedback and exchange viewpoints, and resulted in several alterations to the analysis.

Secondly, at a later stage of the analysis, results were checked against actual interview data by two senior colleagues. The internal research supervisor, a senior academic health psychologist with extensive experience in qualitative analysis and published studies using the IPA method, reviewed the resulting themes along with a subset of four interviews. The external research supervisor, a consultant clinical psychologist with extensive experience in health psychology, including endocrine disorders and Cushing’s, and experience in qualitative methods, also reviewed the first draft of the results, along with six interviews. Both were primarily asked to comment on issues of validity and coherence. Additionally, they were asked for their own assessment of the major themes present in the interviews, and whether they would have arrived at distinctly different conclusions. All comments were considered and discussed carefully, and influenced the process of analysis.

Thirdly, I considered ways in which I could involve participants in this verification process. One of the pilot participants, Fiona, who as well as a 'phone buddy' in the Pituitary Foundation was active in organising local support groups and publishing a newsletter, had expressed particular interest in this research and had sent me information about Cushing’s on her own accord. Following an exchange of correspondence, I sent her both the transcript of her own interview, as well as the first draft of the results section. The questions I posed to Fiona were different: I wanted to know whether the
themes I drew out resonate with her own experience and that of others; whether she felt these themes truthfully reflected what was most important to her (and others she knew of), and whether my ideas brought any new insights for her. Her response was very positive and reinforcing of the main messages of the analysis, and discussion with her further influenced my views on the process of analysis.

Finally, I also sent interview transcripts to all six participants who requested them, with a letter requesting any comments and reflections on the transcript, and any new ideas. One response was received by the time of submission. This participant had found the process of the interview and reading the transcript very helpful and illuminating, and judged that the interview had 'undoubtedly' covered the most significant aspects of her experience. Her observations and suggestions were taken into account in forming the results, and prompted further reflection on the selection of thematic labels.

Reflections

Reflecting on the potential influence of my own beliefs and attitudes during this stage of the study was equally important to considering their influence in terms of theoretical background and overall motivations. This was because such parameters could, and did, influence the analytic process. I identified and addressed three particular influences:

Firstly, the impact of training as a clinical psychologist, with its focus on individual formulation. The impetus to form a coherent, linear account, tailored to each individual,
with reference to theories and clinical concepts such as cognitive schemas, or defence mechanisms, was significant. It was nearly reflexive, and felt 'natural'—both distinct markers of bias. Although it perhaps helped in better understanding each individual’s story, it made it more difficult to think more abstractly, broadly and across individuals, pulling together common factors. Additionally, it made it more difficult to see experiences as human experiences, without a clinical lens. For example, for some time I implicitly struggled with the dilemma of whether to consider a person’s pessimism and negativity as a symptom of depression, or, more in keeping with phenomenological principles, as their valid, irreducible experience. This pressure came to light during the course of various discussions with peers, and resulted in a considerably fresh look at my approach to analysis.

Secondly, the depth of medical knowledge about CS I had actually acquired in the course of preparing this study also constrained the analysis. I was indeed expecting that participants could have very different views from their doctors, and knew full well that my objective was to describe those, rather than compare them to the medical model. However, on reflection, it was slightly more difficult to really follow and understand participants’ views when they seemed at odds with received medical wisdom. One striking example is the following: I wondered, at some stage in the analysis, why participants appeared not to understand, even at a common-sense level, that ‘chemical’ bodily restoration would be required for considerable time after surgery; after all, it appeared obvious to me. Reflecting on this, I realised I was implicitly employing a ‘chemical model’ to understand Cushing’s, which I had drawn from medical knowledge about the effects of cortisol. In contrast, participants often talked in terms of an implicit
'surgical' model, where the direct effects of the tumour itself were considered more prominent. Going back to the text provided evidence of the validity of this distinction, and helped considerably in clarifying several different themes.

Finally, a neutral position in the often pitched battle between doctors and patients was hard to maintain. I felt indebted to, as well as surprised by, participants who showed such great interest and concrete commitment in participating: Clare, Peter and Corrine travelled into London from different cities at their own expense, Courtney kept calling back to reschedule cancelled appointments, Gemma scheduled an interview the day before her operation, Ahmed soldiered on despite his poor English, Nick insisted on driving me to and from the station 10 miles from his home. All expressed their interest in telling their story if it could possibly help others, and several found the process of the interview personally very meaningful and healing. More than just talking, in the interviews we developed a relationship and I was moved by their stories of pain and struggle.

At the same time, many participants talked in very negative, angry terms about many of their doctors, and the care they had (not) received. The implicit dilemma for me was how could I truly be neutral in this, without taking on the accusing tone of participants whom I had grown to like, respect and feel indebted to? This could well lead me to find more evidence in favour of particular experiences, adopt a certain language or tone when referring to doctors, or frankly create a good doctors/ bad doctors split so that I could avoid conflicting allegiances. When I reflected on this pressure, I resolved to be additionally cautious when considering themes related to doctor-patient interaction, ask
peers to review my use of language and make sure the evidence was there to support any claims.

This section summarises the methodological process of eliciting and making sense of the illness experiences of people with CS. At all stages and levels of the analytic method, guidelines set out in the literature were consulted, but equally it was important to draw together a coherent process that was most responsive and appropriate for the particular research questions, setting, participants, and myself as researcher and qualitative analyst. The role of peers, supervisors and participants as co-researchers is highlighted as crucial in prompting reflection and shaping the course and outcome of the analysis.
Emerging Themes

CS may manifest and progress in diverse and complex ways, and treatments and outcomes are correspondingly variable. Thus, no single account could fully encompass the broad range of patients' experiences. What did emerge, however, was that the accounts participants gave of their psychological experiences appeared to cluster around two main significant themes.

Firstly, there was a persistent, strongly motivated effort to establish a coherent understanding of the illness within. The narratives provide rich material of participants' sense of themselves as constructing, testing and reconstructing their ideas about the illness, in order to 'come to terms' with what was happening to them. This was labelled
'Evolving Understanding' to evoke a sense of a dynamic, ongoing process. It does not imply that this process is somehow teleological, i.e. aiming towards a 'gold standard' set of beliefs which participants are meant to adopt. Though external observers may consider some beliefs more 'correct' than others, what this theme emphasises is participants' strong motivation to refine their understanding of their own particular symptoms. Equally, it does not imply a linear, singular progression, since in several accounts competing, distinct sets of beliefs appeared to co-occur.

Individual understanding and its development strongly emerged as being continually shaped, motivated and constrained by the attitudes and beliefs of family members, doctors and fellow patients. Participants recalled how husbands, mothers and GPs interpreted their reports of symptoms or illness behaviours as 'a valid illness', 'self-inflicted stress' or 'being hysterical', and reacted accordingly. They talked about how fellow patients validated their experiences and helped them make sense of their illness better. These perceptions influenced patients' own beliefs about their illness and their approach to medical care.

This connects with the second theme to emerge: the 'Transmutation of the Self'. In parallel to the process of understanding the illness, participants engaged with a significant challenge to their sense of Self - in the broadest sense of core beliefs, valued roles, goals and 'selves'. Due to physical restrictions and impairments, as well as their own and other people's interpretations of symptoms, valued identities and roles were

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10 This theme could equally have been labelled redefinition, reworking or redevelopment of the Self. However, I think that the chemical undertones of this word actually convey something of the experience of CS more specifically.
denied or jeopardised; concurrently, negative, threatening identities and impoverished roles accrued. Notably, though, some participants' narratives conveyed a strong sense of a positive, self-enhancing change in outlook, life priorities, valued identities and roles.

Again, the role of social context in defining this process strongly emerged in all participants' accounts. Interactions, relationships and roles within family and medical systems sustained or denied valued aspects of identity. Participants talked about their struggle to retain a sense of themselves faced with others rejecting or dismissing, blaming, not hearing, ignoring or taking over. Equally, they talked about family members and doctors whose care and attention protected, sustained and restored valued identities. The following sections will discuss and illustrate each of the above themes in detail. Figure 3 gives a schematic representation of the relationships and organisation of the broader themes.

It is important to acknowledge that at this stage of analysis, these results are a broad, preliminary organisation of the very rich data that emerged. The results illustrate and extend a broad (and novel) view over the experiences of people with CS. However due to time constraints, many of the questions and topics of interest that this first round of analysis brought up could not be followed up in great detail. It is the task of further analysis (which is ongoing, with a view to publication) to further develop, elaborate, and link these themes.

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The following notation will be used in illustrative quotes: all quotes are indented; excerpts are identified by the participant's name; interviewer's speech is in bold, within brackets if brief; all names have been changed and are presented by initials, e.g. "A [husband]"; significant non-verbal communications, transcription notes or context clarifications are included in square brackets, e.g. [laughs], [sic] or "Mr T [surgeon]"; paraverbal utterances (e.g. "uhm") were not generally transcribed unless clearly significant to the meaning; the notation [...] indicates that some of the speech has been omitted from the quote; silences or speech trailing off is indicated by a short (..) or long (...) ellipsis; fragments or revised sentences are indicated by a dash (- or --), e.g. "I said- well that was before- I told him...".
Figure 3. A Diagrammatic Representation of Emerging Themes

The two main themes and several significant constituent themes are given here. Bidirectional arrows indicate interactions and influences.
Evolving Understanding

From the first symptoms, to diagnosis, treatment and even recurrence, participants talked about being engaged in a continual, active reasoning process to understand what was happening and why. They prioritised and valued understanding, in itself, but also as a necessary guide to action and adaptation:

Understanding helped tremendously. It's just helped to understand it all, it just helped to know, yes, that is why your body is reacting in that way. Then you can actually put a label on all the things that had been happening to me over the past three years, and say yes, that's the cause of it. I think that helped tremendously. That helped tremendously to start coming to terms with it all. (Nick)

I understand what's happening to me. Like understanding the inside of the clock. If you understand, you don't seem to let it get to you so much, at least I don't. (Glenda)

If you understand what's happening, and you understand what the tests are trying to do, it makes it much easier. If you just are being treated and don't know what's happening, you don't seem to be able to get yourself better so quickly. Because a lot of it depends on you making yourself better rather than you sort of drifting into getting better. People who understand what's happening seem to get on a lot better. (Fiona)

And.. yeah... that was the biggest problem, I was not really being able to handle the frustration of not knowing- because I couldn't take it forward, because I couldn't- I don't know how people cope with a long-term illness, when they don't know what is at the end of it, I don't know how they cope with that. no.. (Peter)

The following illustrations bring together some patterns of beliefs that participants expressed about their illness. Taken together, these clusters will illustrate the overall theme. Though there are qualitative changes over time in how participants reasoned about the causes and implications of their illness, it is not my intention to map a rigid sequence of how understanding develops.
Equally, although these beliefs may roughly correspond to different ‘stages’ of the medical process (detecting symptoms, getting a diagnosis, treatment, etc), they were not simply defined by medical developments or symptoms. The actual medical intervention, its timing and outcome varied considerably amongst participants. At the time of interview, some were in the process of having investigations and planning treatment for recurrences, several years after the initial diagnosis and surgery. Others were on ongoing drug treatment; Pamela was waiting for the post-surgical symptoms to resolve, while Claire had received the all-clear letter from the Consultant that very same morning of the interview. These clusters are thus drawn from both ‘here-and-now’ and past evaluations and beliefs.

Clusters of Understanding


Everyday Causes

At the initial stages of the illness, participants talked of understanding their symptoms as common, everyday problems, to be ignored or dealt with by common, known methods. Though widely variable, fatigue and weight gain were frequently the most salient and intrusive symptoms to be initially noted. When identified and interpreted as problems, a wide range of commonsense causes were considered: bereavement, pregnancy, the stress
of a hectic lifestyle or the trials of parenthood, ageing, dietary imbalance or lack of exercise, etc:

That's all I had thought of, maybe I had just run myself into the ground, burnt myself out, that's probably what it is. (Katy)

Well just- because it happened- most of the symptoms happened after I had [the baby]. I just put it down to keeping the weight on after I had her, finding it hard to shift. (Courtney)

[My back] started seizing up, I was re-laying the drive with a big blocks, and I couldn't do anything, the garden stayed in a total mess for three months, because I couldn't bend over. And I thought that lasted a lot longer than a torn muscle, I would have expected it to. (Peter)

I thought it was age, that bit. I thought that was just part of getting old... uhm.. so.. (Sheila)

Well within those five years, I had lost a baby, my brother had gone through a divorce and he was in a financial mess which I had to sort out for him. My mum and dad got divorced so I had to sort her out. Things came out about what my dad try to do me and my brother when we were younger. It was all those things going on... So there was an awful lot happening... I thought it was related somehow to stress. (Melanie).

And it's funny, because quite recently, I understood what diabetes insipidus is, you get very very thirsty and going to the loo a lot. And I had a craving for water, I was drinking like 8 litres of water day [...] This was during the pregnancy, and I just thought I had a craving for water, like you have cravings for all sorts of things. (Gemma)

According to me.. The weight gain was -- there was a- you look for reasons, and the reasons we found, for the weight gain was the fact that I had stopped playing football, and stopped having regular exercise, and the fact that I got married and I was content and enjoying the good life, and then having broken my leg, and therefore exercise, physical exercise had stopped completely, but that coincided with the Cushing's taking a real hold. So, if you look for reasons, you can always find reasons, they might not be the right ones though. (Nick)

I thought that perhaps I was just getting bigger.. because my grandma, she was a big lady, she was only about 4'10"", I think, but she was very big, but she had big bones, made her look quite big. My mother’s sister was quite a large lady, she weighed 18-19 stone, so I thought, just one of those things that run through the family. (Lorraine)

Everything, I put it down to the menopause. (Joanne)
Drawing from their assessment of the causes, participants talked of applying their existing, well-rehearsed ideas and solutions for each symptom, as well as coming up with new ones. These coping strategies also served as concrete tests of the presumed cause:

I was still going to the gym and things like that, but nothing changed really. (Melanie)

But it was all over quite a few months, because when your hair stops growing or falls out, you think oh well perhaps I'm lacking in vitamin C, maybe I'm run down, so you try and deal with that. So you go out and buy vitamin tablets and try and address that issue. The skin, maybe I thought I was allergic or maybe it had something to do with my diet, so I tried to improve that by drinking more water. (Katy)

Literally I would eat things such as boiled cauliflower with nothing on it, that would be my food all day. Admittedly at that time, especially the first time, I wasn't saying to the doctors I'm not eating, because I thought this was up to me, this must be something that I am doing that, you know, maybe having a bit of cheese on your cauliflower, maybe the cheese was too much.. it was that kind of approach. (Gemma)

Ignoring

Some participants said that despite being aware of symptoms and changes, they had not prioritised dealing with them, implicitly or explicitly believing that these were not serious enough to compete with other life priorities for attention:

I don't think, I've always been fit and healthy and my family has been really so you don't get too analytical about what's happening. You're too busy trying to live in normal life and you're ignoring it really. (Fiona)

I didn't like it, but I just- didn't have the energy or the willpower to do anything about it- I just, you know... well, bit of weight...I just ... feel too tired all the time to do anything about it. (Sheila)
Though most participants reported noticing a progression of symptoms, for Clare and Glenda the problems had been constant for so long, it had indeed became ‘the norm’:

For 10-11 years I did not go to the GP for anything, I was never- didn’t get off work, or anything. I just thought I was a freak.. I just never thought that it would be an illness that caused that sort of things, I just- I just carried on, and.. When I woke up in the morning, I had to shave my face [cries] … before I could see anybody…it just got to be routine really, it just got to be the norm really. (Clare)

No, I knew that wasn’t right, eventually I learnt that it wasn’t right. But at the time it had been going on for so long, you just think things in normal, because the symptoms go on for so long. It was normal to you. (Glenda)

There is Something Wrong

Ascribing the problem to a commonsense cause and acting accordingly inevitably failed to provide any relief or reduction of symptoms, and ignoring the symptoms did not make them go away. At some point, there was a qualitative shift in understanding, with an increasing conviction that ‘there was something there’, something palpable, serious, progressive and outside the range of what one could deal with alone:

I didn’t feel that old, you still feel like 21 in your head anyway.. I didn’t feel- it didn’t feel right. It- didn’t feel right, I felt frustrated.. hmm.. (Peter)

So I said [to the doctor] listen, I still have the same symptoms, okay, the blood tests have come out saying that there is nothing wrong with me, but there is. It may not be thyroid or glucose related or whatever, diabetes, there is something there. […] I had convinced myself that there was something wrong with me, I knew there was. I had led these symptoms conquer my life for over one year, 18 months, and I thought no, there is- I wanted a solution. Some- whether it was cancer, whether it was- thankfully it wasn’t, but I just wanted a solution. (Katy)

Suddenly I couldn't play three sets of tennis, almost overnight, you think there's something wrong here, you know. (Fiona).

It was just to the school and back, I used to walk there for exercise. But by the time I got back from there, I just thought, I felt as if I had been on a Grand Prix! I'm telling you, just a five-minute walk which I would do every day, but this was
taking a toll on me. And I thought, there's something got to be wrong with me. (Courtney)

It was a continual thing, little niggly things, it just wouldn't go away. I knew something was wrong but, well, I didn't know what it was. [...] I just thought something was wrong. I didn't think it was cancer or anything like that, I just knew that something was wrong. (Melanie)

An Existing Problem

Pre-existing illnesses, especially ones whose symptoms were in any way similar to CS were often considered as possibilities:

I didn't really know- I mean- I didn't know what was wrong but I just knew there had to be something wrong, because, what do you call them, normal people, don't just get fat looking at a doughnut, whereas I could. Normal women don't have thick dark hair growing in all the places that it shouldn't come out of. [tearful] Periods don't just stop for ten years without there being something wrong. But as to what it was.. I knew it had to be something. I guess I put down to the polycystic ovaries getting worse, and them not being treated. (Pamela)

Well, I had the excess hair, and the skin problems, but I blamed that on the carbamazepine I take for epilepsy. Because that can cause that is well. [...] I had depression when I was young for two years. And now I can go really badly back into depression. But that's why when it started the stress levels and the depression escalated. That's why I thought, I've had it before, I didn't put it down to Cushing's. (Glenda)

I thought it was part of the gynae problem that I had. So, I wasn't trying to find any other symptoms, and think oh what else have I got. I purely thought it was gynaecological. And although I have this fatness around the middle, it didn't really appear to me.. too strange, until it didn't go after I had the hysterectomy. Because I had semi-associated everything that was happening, and whatever was happening, with.. the fibroids, and the gynae problems. (Corrine)

Something Evil Inside

This strong sense of confusion was combined with vivid, threatening images and expectations, and feelings of fear, hopelessness, and despair were conveyed:
So just- I knew that there was something... evil inside, something beyond my control. (Katy)

I feared -- I used to lie there at night, thinking, if this is what it's like to get old, I don't want to get old. And I can honestly say, that had I not been married, happily married and also having the kids, then.. it would probably be quite easy to have contemplated suicide at one stage.. because I couldn't work out what was actually happening to me. (Nick)

I thought there was a growth in there, and I thought it was killing me. Or liable to.. [...] Because I really felt that I was going downhill, and that I was going to end up going out the door. (Sorry?) Going out the door (As in..) as in, a coffin! That's what they used to call it, when I was a child! I thought that was going to be it, I was a downward spiral, into, into the end of life.. Especially, I had visions of this being cancer.. (Peter).

Well, as I said, I was losing my physical condition. I knew somebody with multiple sclerosis, and somebody who had just died from motor neurone -I wasn't particularly considering that. That was along the lines of what I was thinking really. Because they were the only conditions I really knew. (Fiona)

Madness and Lies

Amongst the various beliefs that participants discussed, there were several striking, explicit references to 'mental' causes. Some participants talked of questioning their own perceptions, and their sanity:

But the problem was, because you didn't feel particularly ill you thought it was in your mind, so you are thinking well am I trying to get attention or something like that? [...] When they kept saying it was stress, I thought well, is it mental, is there something mental that I have get sorted? I even went to see a psychiatrist to see if that would help- I felt fine after I got some things off my chest but nothing was improving, so I said no, it can't be that. (Melanie)

You get the stage where all these things are going wrong and you think, is it me really? Is there something desperately wrong or is it me just being a total hypochondriac? (Fiona)

[I considered] that I was going mad, yes. I did consider that. If I hadn't had thought that I was, I would not have taken the pills that they prescribed, despite the family pressure, if I thought it would be a total waste of time, I wouldn't have taken them. But I did, so.. It follows that I thought I was. (Peter)
Frustration and Confusion

The failure to understand came together with confusion, but which came first? When thinking back, Nick at least partly attributed his feelings of confusion, depression and frustration to the illness itself. On the other hand, Peter and Katy attributed them to the failure to understand itself:

Although, I've got to say, you don't actually realise its actually happening. It's such a gradual thing, all the symptoms, and obviously the psychological problems at the end, they are built up, they don't just happen, they must be built up over a period of years. Because surely it can't be that one day- I'm confused! It must be building up to a state of confusion.

Where did you think that confusion came from?
I think it was part of the stress because of the excessively high blood pressure. Whether that can be determined medically, I don't know. Because as soon as my blood pressure went down, it went down as soon after they did the operation, it went very quickly back to normal, and you start thinking straight. That's what I remember anyway, whether I make myself believe that are not, I don't know. Also, I've got excess cortisol now, but I wouldn't say that I'm confused now, and my blood pressure is at the right sort of level (Nick).

I thought that was where my outbursts were coming from. It was the frustration that was making me lash out, not physically, but mentally. I still suspected it might have been -- I don't think it had that much effect, I don't think Cushing's had that much effect on me mentally, I know it can, but I don't think it did with me. It was just literally frustration. What the bugger was this thing! [...] Uhm.. I think now that- my.. the- the- I was given happy pills because I was displaying symptoms of mental problems. Now I don't think those mental problems were necessarily to do with Cushing's, I think they were due to the frustration of having things wrong with me and not knowing what they were, and not being able to deal with them.. I may be totally wrong, it may have been Cushing's, uhm.. (Peter).

Suddenly all these things were happening to my body and I was not in control! (What did that mean to you?) I suppose, angry, frustrated, very upset... just very stressful, just not being able to be on top of things. I've always never done cigarettes, drugs or anything because I've been... in control. And suddenly something has taken over. (Katy)
Failing Faculties

Participants also talked of finding that the skills and faculties (both cognitive and emotional) which they needed to deal with the symptoms had also been affected by the illness. This made the process of understanding and adapting even more fraught:

And I still – at that stage, I didn’t go back to the GP, probably because I couldn’t make a decision to save my life anyway, I was in quite a bad way. (Nick)

My judgment was impaired as well, because on reflection I made some crass decisions at that time. (What do you mean?) Just the business side, just not thinking things through to their logical conclusion. (Peter)

Fiona labelled this the ‘emotional magnification’ effect of CS:

So, everything [in my life at the time] was quite difficult anyway, and it was quite difficult because I had been sick. Things were sort of slipping away. The Cushing’s in itself emotionally magnifies all the things that go wrong anyway. 

When you say ‘emotionally magnifies’, can you explain that a bit more? Well.. Instead of being upset you become totally depressed.. And I don't think you get so many highs, but there are many of lows out there. You feel like crying about silly things, you know, silly things you get quite uptight about. You know, things play on your mind. (Fiona)

Overall, this cluster captures participants’ beliefs that something was wrong, but also the increasing sense of confusion, frustration and distress when it could not be identified or controlled, which led many to question their own sanity.

Gone and Forgotten

With the backdrop of having only vague, fearful images to make sense of what was happening, participants related their often immense relief when eventually they were
given the diagnosis of CS, even if it involved the otherwise frightening image of ‘a brain
tumour’, and the threat of sensitive surgery:

And then when he said but oh, you’ve got a tumour in your head, I said hurrah!,
great, because it was just like the means to an end, it was somebody saying we
have found something. [...] I wanted to kiss him, I thought hurrah, wonderful,
great and- it was funny really because on that consultation, when I went in to see
him.. I thanked him and I said it’s great. [...] So they booked me in for a pituitary
operation a couple of weeks from then. (What was that like for you, that
time?) It was- it was- I couldn't- so excited, it was like going on holiday! I
couldn't get into that theatre soon enough! Because it was a means to an end.
(Katy)

Because I thought, God, one little operation and I'm going to be better,
especially after I had suffered all these horrendous tests that seemed never-
ending and Mr R. [surgeon] said I will do it in three weeks- I was running, and
skipping and jumping like somebody who was- drugged up I guess. I was over
the moon, and everybody who had gone through it with me, as well as my
parents, said this is great news, sorry that you have to have an operation but oh,
Pam, it will be brilliant! (Pamela)

A Light at the End of the Tunnel

There was relief partly because a diagnosis is a concrete, unassailable sign of
understanding: the doctors know; and surely, if it has a name, it must have a treatment. It
is the promise of understanding, combined with the promise of control and restoration:

So to then be told that when they got rid of this disease I could stay at my
weight, possibly even lose more, without having to try so hard, eating the same
foods that everybody else did, fish and chips from the chips shop without feeling
guilty, to go on holiday and eat all the things you love, then come home and not
find that nothing fits you again, that would be important. (Pamela)

Did it make a difference, having a label? Yes, I think it did to me.. If it was
called something, it could be something that could be treated. (Clare)

Did you have some expectations then? I think yes, when they said that I’ve got
Cushing's, I thought it was all, yeah, it was all downhill from now I thought.
They can't tell me there isn’t a cure for it. (Courtney)

Total, total relief, that somebody could tell me why I was ill like that. [...] There
was light at the end of the tunnel...For somebody to actually turn around and
say no, we can cure you, we can get your back onto the road of recovery, and we can get your back being able and fit, was just like a breath of fresh air. (Nick)

When the doctor finally said yes it is Cushing's, I just sat down and felt relieved that there was something wrong and that somebody was going to do something about it. (Melanie)

I Told You So

Significantly, diagnosis also brought relief from the blame, shame and sense of failure that had built up, and anger at the doctors’ dismissive attitude or attributions of the symptoms to ‘cracking up’:

Basically, thank God that there is something out there that I've got, rather than people thinking you're cracking up, I think. (Fiona)

Because I damned well knew there was something wrong with me, and they wouldn't listen. So at first it was a relief to know that I wasn't going crazy, that there was something wrong. (Peter)

I was relieved in a way that I was ill, it wasn't just me putting it on, it wasn't me keeping the weight because of what I ate, after I had the baby, that there was something wrong.. that I wasn’t...(Sheila)

I just felt glad that somebody actually believe the me. It was just relieved that I wasn't talking to myself at last. Nobody would believe be that I felt that there was something wrong with me. It was a huge relief. They say that some people get upset when they are diagnosed, I was so glad! I felt like I could get help at last. (Glenda)

I felt relieved and angry, angry that it wasn't diagnosed earlier.. and let down, basically because I kept going and nobody was taking any note of what I was saying so I felt angry. (Melanie)

Restoration and Undoing

Almost automatically, the initial understanding that participants formed was one in which removal of the tumour would totally get the patient back to how s/he was before; a picture of restoration, undoing, and forgetting:
The Cushing’s was a road to Damascus point. As soon as that word, that
diagnosis was made, everything changed. It was- sorted, it was... sortable.
(Sortable?) Yes, yes, there was an end to the game. (What did that mean?) It
meant that it could be resolved and then I could go on...(Peter)

Did you have any ideas or pictures about what ‘normal’ would be like?
Hah.. What I used to be like, I just wanted to be a happy, healthy, cheerful, go-
getter focused, I just wanted to be a normal person, I just wanted to be what I
was prior to Cushing’s. No better, or maybe, well I thought, I would get older, so
I would make some allowances but... [...] It was very exciting just going for this
operation, you know, when Mr K explained the procedure, I said fine, use a- do
what you like, just get in there, just rid of the bastard! (Katy)

So I was determined I think, determined to get it all out of the way and done.. I
wanted it taken out- the doctor explained how the operation was going to happen
and I thought well I’m not going to get any better. [...] I want it out, out the way.
Because I thought that if I had it out the way I’ll get better really really quickly
and then I could forget all about it. (Melanie).

I thought I’d be better straight away, but obviously it doesn’t work like that! I
was hoping all the symptoms would go and everything.. but.. (Sheila)

Cutting It Out

This understanding of CS implicitly links all the symptoms with the presence of a
tumour in itself, rather than with the overall effects of long-term cortisol overproduction
in the body. It logically follows from this understanding that a surgical procedure is
required to remove a tumour:

I think you hope, and everybody does, you get treated and it gets dealt with
quickly. I mean if you've got cancer or something, they operate within weeks
don't they? (Fiona)

So when they discovered the pituitary tumour it was just a relief. And I thought,
great, at least they will remove that and everything will be fine... (Katy)
I've had operations before, when I was younger I had my tonsils out, because I
had tonsillitis. I had my tonsils out and the tonsillitis went.
So did you compare this operation to that one?
Well yes because every time- I mean I have had my appendix out, I was in
incredible pain, being the sick all the time, they took my appendix out, I was
better. Whenever I've had something done, it's made me better. (Pamela)
Despite going away with only a vague impression from the consultation, Corrine retained a sense of a contained, compact treatment:

**So what did you understand that day?** ... I can't really remember, except that I knew I had to have an operation to get rid of it, and I wanted to organise things so that it could all be done as efficiently as possible.

**Can you tell me about this, get rid of it?**

[...] I can't really remember if they said we can't do anything and I said, well are you sure? Or something like that, so again it was a bit of a nebulous time, but.. I just wanted it.. I had read in the notes that.. my sister-in-law had sent me a short piece from a medical encyclopaedia that she had, and then I looked it up on the Internet, and most of it was a trite description of the operation and the recovery period, and back to work within 6 weeks, and that's all I thought, I didn't think any more than that. So I thought let's get it over with, let's fit it in.. (Corrine)

**A Hopeful Illusion**

Strikingly, participants frequently intimated their awareness that these very vivid, urgent expectations may have been a result of hope more than any concrete knowledge:

And just wanting it to happen overnight rather than waiting, having to take so long. I just wanted a solution quicker please!

**How did you know that there was a solution?**

..mmm... I probably... I probably didn't really...[...].. Erm, I suppose I just-- you-- I didn't-- my head was in the sand a lot, I-I didn't know even then- he'd always questioned Cushing's, Cushing's.. And I looked up the Cushing's web site and gone into that, but I didn't want too much.. information. And a lot of the Cushing's stories I read said if you do your- you'll get back to normal. And that's all I wanted to hear, that you'll get back to normal, once they- remove your-[tumour]. (Katy)

I expected that.. the moment that they took that lump out of my head, I would look and the mirror the next day, and instead of a big round red face, I would be back to myself, I expected to be that quick. But of course it wasn't. Yes, that was my image, that it would be that instant. *(Where did you get that image from?)*

Expectation or hope, I'm not sure [chuckles] just how I thought it would be, I thought it would be weeks and not months, certainly. (Peter)

To tell you the truth, all I wanted was for this thing to go away. That's exactly what I wanted. And.. in reality, it wasn't going to go away, because I wasn't well. I just wanted it to go- and I was, come now, you've had you joke, kind of thing. I needed it to go now, because I have to get on with my life. Not realising
that.. it wasn't going to happen like that. There was something wrong, and I had to face up to, I had a problem. (Courtney)

For Glenda, this expectation was reinforced by the fact that tests were done immediately post-operatively –thus, ‘if they are looking at it now, it must have changed already’:

The cortisols [sic], I knew they were instant. That’s because obviously they were transferring me to do the dexamethasone test straightaway [after the operation], so it had to be instant.

**And if the cortisols were instant, then..?**
Well if the cortisol are affected instantly, that is you stress gland, so I felt that you would get less wound up and less stressed straight away. (Glenda)

Courtney’s reaction to the bad news of a failed operation illustrates quite how powerful this hope for restoration can be:

When I was put into this operation, it was like the opening of this flower, and then, I had it, I went home, and they said oh, I've got the results coming through, come back in a couple of weeks time. Which I did, I went for the results and they said it was unsuccessful. And that was -honestly- you could have knocked me down with a feather at the time. I was in tears, because I thought, I though this was my start of having something special again, back to my normal self, and it was a knock down, I felt really- I felt physically ill, when they actually told me. I was mentally ill, and I just- I just- I just didn't feel that I could go on.

It is yet again hope that is evoked when thinking about her upcoming adrenal operation:

I just want it to - even if - in my heart of hearts, I'd like it to be successful, but if it's not, then I'd like to know.. that I'm living on medication, but.. I would always want to know that there might be something, light at the end of the tunnel. That there might be a breakthrough, where they know exactly what to do, to help people with Cushing's, but.. I don't know, I just feel that this operation is going to be the one for me. (Courtney)

Overall, this cluster captures beliefs about a quick, contained recovery. These beliefs appear to be formed by the relief of getting any diagnosis, a hope for restoration and a
commonsense understanding of what removing a 'tumour' implies, as much as any factual information.

Cushing’s Is For Life

Katy talked about her developing understanding of CS:

I just- I'm still going through the battle, and I don't know.. I believe this is for life not just for Christmas, but I do hope I can work around it...So it could be a long road still. (Katy)

When the Box Burst Open

Forming beliefs about the long-term outlook of CS often occurred through one or more painful moments of disillusionment. The short-term, 'gone-and-forgotten' model of the illness, fashioned out of hope as much as out of knowledge, failed to materialise. Some participants talked of overcoming this disappointment quickly, buoyed by some initial signs of progress and improvement, and by learning more and more information about the illness:

Logically, intellectually, yeah.. It is going to take time. Mentally it was that it was going to happen overnight. Well it didn't. But it wasn't an issue, because I was feeling better. It was gradual, but that was enough to carry me forward. (Peter)

Others, however, were shocked and devastated when things didn’t work out as imagined, and changes in beliefs came at a very high cost to mood, morale and confidence:

And then [in July] I had the operation.. which all the doctors told me was going to be the be-all and end-all, I was going to get better, everything was going to be rosy.. (tearful) ..[...]. After everything that they told me, I was thinking- by
Christmas... I would be feeling a lot better, I would be off the drugs, the weight would start to come off, I could get back to work... and it's just been none of those things. (tearful) And I mean I said to Dr K when we went to see him, if I had known how ill I was going to feel, how depressed I am... (tearful) how upset I get, I would never have had it done. (Pamela)

Because after the operation I thought I'd probably be better and I wasn't, so that was another knock, and I'm still just trying to cope with what's going on with my body and my life. (Courtney)

I'm a bit scared about having an expectation really, in case.. things don't happen that way...It goes back to when I originally went to hospital for treatment, having that expectation, six months and everything will be OK, I don’t want to go through that again.. que sera sera, type.. I don’t have.. plans in my head of what I want to happen. (Clare)

I didn't realise how serious it was until Mr R saw me and said.. very poignantly in my view, he said well Cushing's does affect every single organ and system in the body. That just hit me like a, like a.. bolt of lightning. And it suddenly came home to me how, how dangerous it is, and how, how serious it is. Up until then, I just thought of it as something that I just got okay let’s get rid of it, and move on. [..]All I remember is the shock, the realisation that it was a serious illness, that it was something that would be with me forever, that it wouldn't go, OK I might not be back to work, as quickly as I thought I might be, OK this might affect me for the rest of my life, yes this might have an effect on my heart and my this and my that... like, the box burst open from that point, and I realised, you know, that was the reality of this, from then on for me. (Corrine)

It’s Something You Live With

Yet when this long-term view was established, it gave patients some stability and coherence, even if it was a hard reality to face. When considering recurrences, continuing effects of the illness or side-effects of treatments, or even in evaluating their progress, some participants clearly conveyed a revised model of the illness as a serious, complex, long-term condition:

I've got it and it's something you live with really. [..] It's quite easy to get disillusioned anyway. It takes several years really before you reach a plateau, lots of things take quite long time to get over, physically, bodily and mentally. If I talk to people, they say after a year I don't feel any better and I would say, if
you think about it really you do, but it's going to take two or three years before you really feel your are getting better. (Fiona)

I've still a long way to go, a long way to go. I've just started to go to the various meetings at the PitPat [Pituitary Foundation] and the Cushing's Society now... I thought I'd never be interested, because I thought, well I'm going to be better, I will just close the book, I will be fine, I won't need to know. Whereas I'm realising, no, it's long-term, you might as well get to know more. So I'm still learning.. (Katy).

Before after I had the first operation after 14 years, I was saying I have had a serious illness but I'm cured, but now I've had surgery almost every year, it's difficult to kind of say.. I'm cured, you know. Because I just -- don't know. (Gemma)

But I've got to take those steroids for it, for life (Joanna)

I didn't expect it to come back. I thought after 10 years, I'd be quite lucky, because I thought that if it was going to come back it was going to come back within the two were three-year period.. I had no reason to -- that was purely my-interpretation. If the tumour is going to reactivate, if there is something there.. then why would it wait 10 years? You know, so my immediate thought was, after a two to three-year period, no, I'm probably fine, but when I have looked after, read certain passages from the Pituitary Foundation leaflets, magazines etc, you see little snippets there which you didn't pick out before, and people do, people do get it twice. (Nick)

I hope that by the end of the year, I will gradually pick myself up a bit better. I just take each day as it comes and see what happens, really. Can't look too far into the future, and say I'm going to be well by oh October or November, because I might not be, it might be other, other things that you get. Cos some of the treatments have side effects, and you have to learn to cope with that as well. (Lorraine)

Chemical vs. Surgical

Participants now noted the distinction between an acute, compact, ‘surgical’ understanding from a more complex, long-term and multiply determined, ‘chemical’ understanding:

So it has really been more adjusted from the surgical point of view, which is- I think- they will kick it out, and then once the growth is gone and the vision is fine, then.. And the endocrinologists have really been in the background, you know.. Haven't really, I don't know, spend a lot of time investigating, looking at
the Cushing's thing. And I think that sort of gets- missed out. Whereas for me, it's a whole thing, yes the tumour affects the eye, but... then also the chemistry affects everything else... It has certainly been a very difficult case, it hasn't been as easy as that, OK we just remove the tumour and then everything resets itself... and life goes on. (Gemma)

I mean I had read in these textbooks that, you know, as soon as you had the pituitary tumour removed, it was a question of recovering from the surgery, and going back to work and taking up your life. It was a very, it was described in a very.. compact [gestures] fashion, but the reality of it to me was totally different. (Corrine)

You said before, you were expecting to have 'a manual'. Well, I was being a bit flippant, but I didn't realise how complex it is, and I just wanted to know what to do to get better. And I do have many manuals on Cushing's and pituitary related diseases, but it's not quite as clean-cut as that, we are all so different, all so complex, which I am realising, we are all quite individual, and it's quite tailor made, but there are a lot of things that we have in common. But we can deal with it, it's just a matter of getting the hormone level correct. (Katy)

Keep on Fighting

For these participants, this understanding implied that different actions and strategies would need to be employed in order to adapt. Personal resources and competencies were now again at the forefront, efforts were redoubled, and participants talked of the need to combine close medical supervision with personal efforts:

It takes a while, obviously you've got these symptoms, and they are not going to disappear just overnight. So... he [the doctor] couldn't divulge too much information, but I just thought, if I have to keep on fighting, I'm going to keep on fighting, but at least they found what the problem is, I just have to go on and just deal with the- with the hurdles as they come to me. (Katy)

And I am determined to see some resolution to Cushing's problem, I don't want to -- because I find as you get - the surgeons do their job, and you spend another six months fighting, or just leave it, and get on with other things, but I am determined that if there is Cushing's... I'm going to see some end to it. If there is some way of getting rid of it, as far as I know it's not incurable, I would like it to be cured. I don't want to live my life.. it is almost like it's a project now, I'm going to see the end of it. (Gemma).
That’s the only thing I can do, I just am trying to keep myself as healthy as possible. I see people in the support groups, and some of them are quite bad, because they’ve given up- you do have to fight, and you have to work hard to keep yourself going. You do fight every day, to keep yourself going, otherwise if you didn’t you become ill quite easily. (Melanie)

Prepared for the Future

For Peter and Nick, being faced with a recurrence was not worrying, because they felt they had a solid understanding of the disorder, what its treatment implied, and had clear plans of action in place:

It was a very easy decision to make [to have another operation], because I knew what I was going to be going through, I knew what -- it was going to be exactly the same trans-sphenoidal surgery, followed by 4-5 days maximum, but because I wasn't in a bad way physically, I could handle those. [...] Certainly, I was very aware of the disease, very aware of what it can do, I certainly didn’t want to get into -- the force of, of the tumour just pumping more cortisol or ACTH, all the time within me, and putting it off for another period of time and just- it’s not going to get better without it, while that's happening, it’s just going to get worse. (Nick)

So, they said, it’s OK [to not operate yet], but I am conscious that they might be saying we go in there again.. Apparently, it would normally be expected within six months.. or it might reoccur in 9-10 years. So they might have to go in again. But it doesn’t worry me, because if it starts to happen again, I know what it is. And I’m going there every year anyway, so I have a course of action, I know where to go, how to get it dealt with. So it doesn’t intimidate me at all. (Peter)

For Ahmed, the continuing checks, and being known and cared for by his doctors gives him confidence for the long run:

I hope it’s finished, but the doctor he says it maybe, maybe, come back in 2, 5, 10 years. So you should come every year for checkup. I had another appointment this year December, and they will check me every year. So it’s a good thing to have checkups?

Yes, maybe if it happens again the doctor will know, they know because they did it before. And then, when it’s finished, they will tell me, you are finished, it’s normal now. (Ahmed)
Overall, this cluster includes beliefs about CS as an illness that will require long-term involvement, active coping and prolonged medical supervision. These beliefs frequently came about as a result of very strong disappointment, and a change from a surgical to a chemical model of causation.

A Vague Menace

In stark contrast, a distinctly different picture was conveyed by participants who believed the illness to be an unpredictable, uncontrollable, menacing presence:

The tumour seems to be back. And I said, look, I saw you three months ago and it was okay, nothing, you were all very positive and happy, what happened, does this thing stay silent, then something happens and then bang, it grows? Because it doesn't seem to be, its growing a little at a time, it seems to be oh, there is nothing there, it's all clear, and then three months later, we need to operate, when can you come in?! Is it something like I do, something that I don't do? What...?! I feel completely helpless about it, I don't seem to be getting any medical answers to that question. Should I eat, or not eat? So there are doesn't seem to be anything they or that I can control at the moment, about when it grows or when it doesn't. Something clicks, and it grows. [..]

**What is it like to be less certain than other times?**

...uhm... I'm not seeing an end, where before, I was like, I'm just going to go in, and have it done, and done with, now.. I'm kind of thinking, I don't know where the end of this is going to be. (Gemma)

I did feel down a lot. Because I don't understand why I've got it, why I've got the illness. Why me? [...] I worry probably more, now, I now worry that it's going to come back. Because I know it can. From what I had read, and then you need chemo and everything. I don't know if I could cope with all that. (Sheila).

And every time we seem to think we've conquered something, something else comes along and whacks me. We conquered being sick and I thought well we have sorted that out, we are on our way now, then the headaches come, they get rid of the headaches, but now I can't eat anything -- every time we seem to jump a hurdle there seems to be another one in front of me and it seems to be getting bigger, and I'm running out of fight...(Pamela)
Sheila presented a very vague, uncertain understanding of her illness, no sense of why she was taking medication, or what the surgery had actually involved:

I want to know if I’m ever going to get off these steroids. I want to be off them, I don’t want to take them for the rest of my life. I’m just going to have to keep taking them, to compensate for whatever -- I’m not sure whether they just scraped the tumour off the gland, or took away the whole gland. (Sheila)

It Will Come Back to Haunt Me

Believing this state to be uncontrollable, a desperate, desolate future appears:

Maybe that’s why.. I don’t try to lose the weight and everything anything, because I think well it’s going to come back, I’m going to get affected by it again. I’ll have to go through it all again.. Maybe it’s just there, you know, somewhere deep inside, and just comes out every now and again. And I just feel that I’m going to get it again. That it’s going to be one of these things that comes back to haunt me every so often. **You sound like you’re almost convinced.**

Mmm I probably am[..] But it is, it’s still there, it’s going to come back. Especially now, I think, this week, I have thought about it a bit more, now that A [husband]’s going to be away, that I will be on my own and will have to cope with it. And I have visions of going back next month and finding that it has all flared up again, and everything. (Because...?) Because I’ll be on my own and I’ve got no-one to help me cope with it. (Sheila)

So I guess.. In the back of my mind I think, so this [treatment] probably won’t work either, or if it does, what’s going to come up next? And because catering is the only thing I’ve ever done... (tearful) what am I going to do? I can’t work anywhere that smells of anything because it makes me feel sick. I can’t tell the difference between smells, a bunch of flowers, a cup of coffee, a curry, fish and chips, it all smells exactly the same to me, and it’s horrible. And it tastes horrible too. So I suppose it would be the same if you were a hairdresser and had your hands amputated, if that’s all you’ve done, what are you going to do? Because I can’t do anything else... (tearful)... it just seems... it just seems like the end. (Pamela)

For Corrine, CS and depression come together to form a terrifying black hole:

I realise that all of these- well, I don’t realise, my view is that all these things happen to us in life for a reason, and I am a fatalist in that respect.. But at the moment, I can’t see any- it’s like- I can’t, I don’t know how many people with
depression you have spoken to, but it's like I'm underneath that black hole, and it is dragging me further and further back. [...] And I'm just thinking, I'm being sucked into this hole, and this hole, this black hole manifests itself as Cushing's! Because it's easy to say that it is Cushing's that has robbed me of all of this. It might not be Cushing's. (Corrine)

Melanie talked of feeling constantly vulnerable, with her body likely to fail at any time, and the need to work extra hard to keep on the right side of despair:

I'm always doing a lot of things, I'm always rushing about trying to doing lots and I do get tired sometimes. I suppose I've got—sometimes I feel as though eventually I won't be able to do it. I don't think I'm—don't think I'm in control of the illness. So eventually I am going probably get worse.

**Did I get that right, you've got this feeling that maybe you're not in control of the illness?**

Yes, I feel vulnerable and I don't like that either. [...] So you've got to make sure that you look after yourself, you have to look after yourself more than somebody who hasn't got it, because you've got nothing working for you. You've got to do it all yourself, to trying keep everything going as well as possible. And feeling good in yourself, because as long as you feeling good in yourself you'll be able to cope at the, otherwise you'll get further and further into a depression, which is very common in this situation, and you've got to try hard to make sure you keep yourself out of it. (Melanie)

This cluster captures beliefs about an illness that cannot be controlled, cannot be predicted, and whose progress is inexorably negative. Vivid images of malevolence and, a picture of the illness as a persecutor, also emerges.

**Multiple Shifting Models**

As argued previously, participants' understanding evolved in parallel to significant moments of the illness and the medical procedures (e.g. appearance of symptoms, tests, diagnosis, surgery, etc) but did not appear totally or necessarily defined by those alone.
In particular, the ‘gone-and-forgotten’ way of understanding the illness was not simply restricted to when the illness was first diagnosed. Most participants further developed their beliefs, since often the short-term, gone-and-forgotten model of the illness failed to provide adequate understanding. Yet some participants maintained some confidence in this model, and referred to it, explicitly or implicitly, at different times in their illness experiences. As a striking illustration, Gemma had had several previous recurrences and treatments, but still greeted each one with the same expectations:

I had the radiotherapy then, it was supposed to be belt-and-braces -just to make sure that everything was completely.. OK. I don't think it was necessary, but.. Again it was like a big shock, I didn't believe it.. that...

**Can you remember what went through your mind at the time?**

I remember thinking, but they told me it was nothing to do with having babies, you know... I was.. scared, you know. Scared, shocked, I was just- confused, because I was convinced, they had really convinced me that that was it, you had the surgery, you had the radiotherapy, this is it, you know. So this was a complete surprise, right out of the blue. (Gemma)

Similarly, after even an aborted attempt at adrenal surgery, and two failed transphenoidal operations, Courtney maintained a belief that the tumour may have disappeared:

And then I was referred to Dr F. after this first operation. I asked for him specifically to do the testing again, because I said I hadn't had any tests for about three years. So I thought that hopefully everything would have cleared up, but I just wanted to know. As easily as it came, I got Cushing's, but I didn't know how I got Cushing's, I thought it might just as easily have gone away. (Courtney)

There was also evidence that ways of understanding could shift radically from one moment to the next. In one part of the interview, Corrine asserts her full knowledge of
the illness and her expertise at its management, but later on conveys a very confused, incoherent model, where her body feels like ‘a crazy maze’. Compare:

**So, you knew that, that was clear in your mind?** Yes, that all of these subplots, the sickness, the diarrhoea, the complete fatigue, the sleeping, loss of appetite, everything. And at times when I had the extreme Addison's, I recognised that happening, because.. You know, I was put on oxygen.. I knew what I had to have, I knew that I had to have IV cortisol, by this stage I recognised what was happening to my body, and I recognised what I needed. I was able to tell the doctors in the casualty department, so I learned a lot over a very short period of time, about how it was affecting my adrenals, and what the other offshoots of the disease were. (Corrine)

With:

**I must admit I am puzzled. You say [depression] is part of the Cushing's pie chart, so you're putting it in with the illness, yet you are saying it's not?**

Mmm [puzzled look].

**Am I right to be puzzled?** (Yes) Or did I just not get it right?

Yes, no, well, I'm puzzled too. I mean [...] just knowing that the depression existed and that it had a physical element, needing serotonin, I don't feel like that now, I don't think I can isolate.. isolate it. I don't feel I can isolate it from the Cushing's, and I think that the two things, the cortisol and the serotonin, and all these other things are all going on in my body, I just feel it's like a crazy maze, that.. However hard I try to untangle things, one thing is going to give me something else. (Corrine)

**Social Framing of Understanding**

Participants naturally and routinely interspersed their descriptions of their emerging understanding and coping with references to other people’s understanding and advice. They talked about the direct influence of information from others on their own understanding. They also talked about how the quality of relationships with doctors influenced the impact of their advice.
Shaping & Feedback

Because of the rarity of the condition, people were understandably very puzzled when they received the diagnosis:

When they first mentioned Cushing’s, I thought of the actor Peter Cushing, and I thought, what was wrong with him? (Lorraine)

It was a nightmare, because I went on the Internet of course, and I tapped in Cushing’s, and spelt it the wrong way of course, and I came up with very different kinds of lovely cushions, patterned cushions, coloured cushions! (laughs) It wasn't what I wanted to hear about! But after looking up, I actually got a lot of information off the Internet. And even though I've got a lot of information, it was just like -- French, because I didn't understand it? It was like, what are you talking about? (Courtney)

Social information emerged as playing a crucial role throughout the process of developing an understanding, not only after a diagnosis was given. Relatives and friends had an influence in how patients understood their early symptoms, by suggesting labels and causes, suggesting treatments or providing practical help. Drawing on their own beliefs and understanding, they provided another viewpoint on what was happening, and feedback on the presence of symptoms, especially changes of appearance. Also, others were often credited with being the ones to label the symptoms as an illness.

Well I have a brother-in-law who has a science degree and he did a bit of endocrinology, and when he saw me, he felt it was hormone related. And then when I went to see another brother and his wife, who had a mother with thyroid, she said just go check it out, it may not be but.. (Katy)

My husband had the feeling that it was something hormonal, but then he's been involved with animals and dairy herds and things! [laughs] But if he said that, it probably didn't go down too well anyway! (Fiona)

I just put it down to stress you see, and other people say things such as you’re stressed and then you just put it down to stress yourself a lot of the time. In fact the specialist I went to see for ME said you just need to cut down on your hours and then go on holiday. (Melanie)
At that time, what did you make of having to take pills [antidepressants]?
I didn't like it, I don't like taking pills, but I did take them, I did-

How did you agreed to take them?
Because of that pressure from my family. I was- that they were unhappy at my behaviour, and I felt I should do something about it, but the one's course of action is limited to what you are advised. (Peter)

Others’ beliefs about the illness were not only influential in the early stages of the illness, when no diagnosis had yet been established. Given the uncertainty about the root causes of CS, other people may put forward a variety of ideas:

Exactly how it started, it should just go, exactly the same way, that's how I thought. Even now, my mum keeps saying to me, I hope you don't want to have another operation, because how does this sickness start? I don't know, so she says, don't you think it can go away the same way? And I said, I don't really know. Because they've said I have got a tumour on that gland, and if they take the gland out, then it should be able to work itself out. I said, some people do get ill, mom, so I'm going to try and people have said it’s worked. (Courtney)

They think it's stress, and they keep on saying to me, maybe you should think about giving up work. Well, okay.. I don't know. So they -- from that point of view, I now have a pressure. I think they are fed up of returning to the same position every year with a me, because I think it is also difficult for them, and I am the one going for surgery, but they are there as well, the fact that they are wondering, or hoping that I'm going to be okay. Certainly my son, he’s 7 now, is beginning to notice and the last time I went into hospital he was very tearful. So they are putting pressure on me to give up my job. (Gemma)

Marking the Illness

Relatives also often make the call to the doctor, thus framing the moment where the illness was no longer seen as a routine problem:

Then my wife turned around, she said that this is absolutely ridiculous, I am making you- I know you don’t want to go back to the GP, I am making you an appointment for what they call an MOT check, a health check, with a nurse. (Nick)
At that point- I'm unsure- my wife and my family were convinced that I was behaving oddly.. and I don't doubt.. that's true. But I wasn't aware of it. But they persuaded- and in fact my son, my oldest son, he took me to the doctors, and said, you've got to do something about this, and he put me on these- these happy pills, which kept me sort of quiet, you know, but.. (Peter)

My relations said that there is something wrong with you, you are bloated out, your face is round, it's really red. At that stage I realised I had battled with it, I had tried to address all the issues, and at that stage I thought, well, I'll go in to the doctor and get an MOT. (Katy)

Friends and family were often the ones to note and celebrate progress, allowing the person to hold onto progress and have confidence in his or her efforts, when disillusionment set in:

My seven-year-old [son], sometimes he gets up, and he says oh mummy you look different today! The other day he said to me, you got to try that Slim-Fast thing, I said to him why? You are a bit big..! So he is beginning to notice as well, mummy changes, sometimes he says oh, you look thinner today, well thank you! (Gemma)

It's not so noticeable to you, but the people around you start noticing that you get better quickly actually. [..] And I started to lose weight, and everybody was saying how much better I looked and how much fitter you're getting. I think it was my husband's conception as well, so if I said oh I'm sort of grotty, he would say oh no you're so much better it's unbelievable. It's quite easy to get disillusioned anyway. (Fiona)

Isolation

Conversely, participants experienced intense isolation and despair when no help in understanding was forthcoming from others, and nobody could or was prepared to, listen:

I was lonely because nobody seemed to be taking any notice of me. (Melanie)

No-one understands what I'm going through.. Although they are there for me, and I just feel all alone with this. (Sheila)
At the time of the first diagnosis, there was no availability of literature or anything like that, no support foundation or support groups. I wasn't aware of any, nobody directed me to any. (Nick)

People in the Same Boat

For some participants, speaking to fellow patients was very valuable. It appeared that its particular value was in the first-hand, direct, concrete nature of the advice that peers could provide:

So I rang a couple of those [on the Pituitary Foundation phone buddies list], because- I think this is about a month after the operation- just feeling, I still feel very rough, I'm just not right, it's just not happening fast enough, I just needed reassurance. Because, rare as it is, I had not spoken to anybody with it, with anything related to what I had. So I phoned a couple of people who had had these things for years [...] But they were all reassuring, they said its going to take two years for your hair to get back, don't run -- try to run before you can walk. So they were very reassuring, so that put me back feeling positive. They said that you have to take at least one year before your metabolism get back into place, and, you know, it was only six weeks. [...] [After speaking to fellow patients] I went oh God, not another goalpost being moved (grumbles)... but at least, rather than speaking to the medical profession, speaking to people in the same boat as me, so I felt that was better in retrospect, because yes, you can imagine losing your hair, yes it made sense, its going to take a while for it is to grow back. But at least if- it does come back, at least if I know there is light at the end of the tunnel, I can cope with that. (Katy)

Establishing Comparisons

Fellow patients also afforded a crucial comparison function: participants drew conclusions about their illness on the basis of stories of fellow patients and their outcomes:

I think at that stage I had met two people with Cushing's, so I learnt more from them, about what to expect. They both had Cushing's, both had had pituitary problems, and then had the adrenals out, so I suppose I knew what to expect along the lines. I probably got a fairly good idea of what to expect, quite quickly, from those two. (Fiona)
I was told at one stage at the hospital that the combination of Cushing's and the back problem was one in a million chance, I didn't want to hear that, because I didn't want to be one in a million [chuckles] I wanted to be one in sort of 10,000 [chuckles] that sounds daft, but I remember thinking that, at the time. [...] I think it was the probability, others had had it and been cured, if I've got it, I'll be cured. If it really was one in one million, and nobody else had really had it, then what chance had you got? (Peter)

Well, I'm certainly interested, although I'm not a member of these organisations, I'm certainly interested to see how other people cope with it. I know you have not finished your research but is there something that you have picked up along the way, that you could just tell me, oh God, if only you didn't do that?! Everybody else that I have spoken to just doesn't do that! (Gemma)

The importance of this function was highlighted for the two black participants, who found it hard to establish a reference group:

With me being black, my body is different to somebody that's white. I'm not saying that nastily, because it's not nasty. I think that we are physically different, with what we eat, with what we drink, our make-ups are different, so when they section me off and they say I had Cushing's, I understood what they were saying, but I thought that, they need to find out how many black people have it.. and how it affects us differently, because I think it does. [...]And honestly, I ended up not speaking to anybody because I felt as if -- there wasn't anyone to speak to. (Courtney)

Is there any greater incidence in people from ethnic minorities, or is it random? (Have you considered that?) Well -- I don't know any other -- Africans or.. Afro-Caribbean people with the illness, it is certainly something I had not come across until I got it. So I don't know if it's something.. (Gemma)

However, several other participants, though white, also found it hard to establish adequate reference groups. Either simply because of its rarity, or through believing that the variability in sources and symptom patterns made any direct comparisons and conclusions problematic:

So I phoned a couple of people who had had these things for years but they were all, a little bit different, they had their pituitary totally removed, so they were totally reliant on hydrocortisone. Or they had adrenals removed, so a little bit more serious than I was, or they had slightly different stories to tell. (Katy)
But then I don't know anybody else who has had Cushing's disease to compare. I mean, if I knew someone else that had it, I would ask, did you use to smoke, did you use the gym, did you work long hours, did you get enough sleep, did you eat too much, blah-blah. But if you don't know anybody else who has had it, you can't compare. (Pamela)

Strain of Conflicting Beliefs

The value of information and ideas that others provide was often experienced by participants as often very helpful, as well as motivating. However, strain and difficulty was also apparent when the ideas offered are very divergent from the received medical wisdom:

So, my aunt said, alternative medicine! I said oh, that will be good. I said I don't mind taking herbs and bushes and what have you, so I said, I'd do that, whatever. I'll do it, I'll be the guinea pig! I went on -- a regime where, I took- this was so expensive- my mum paid for it- I had Aloe Vera, fish tablets, vitamin B complex, I can't remember, I must have been taking about 20 tablets, vitamins, herbs and bushes -- I don't know what, I just used to take all of them and I thought, it can only make the better, it can't make me any worse. I'm not actually sure what I think I did, I think I actually overdosed myself with all these things. (Courtney)

So I felt, when my mother took me to the doctors, and she was talking to the receptionists saying oh, she might have anorexia, I thought mothers! how can I have anorexia, you know, when I weight so much. I was very embarrassed, other people sitting in the surgery, they should be looking at her thinking oh my, she is completely mad! Thinking that this fat child had anorexia! But that was her interpretation of what you would have if you ate no food. (Gemma)

The beliefs of other people about the illness, and their attitudes to treatment can create a strain on the person’s relationships, create unrealistic expectations or have a significant impact on the person’s relationship to the medical system:

I get a nice pep talks from my sister-in-law, or my in-laws, you know, no, we are not pressuring you, but we really think you should think about it, think about your lifestyle, your choices. So I say yeah yeah yeah, I'm thinking, I have to do
something, you know, they say, don't just say that, have a plan! We are not coming to blows or anything, but they keep... you know... (Gemma)

I think they -- I think I suppose they think that once you have had the operation that's it. (Sheila)

All my friends- all my friends and all my family, they think [click] it's got to work, it's got to happen. [...] Because -- some people, my mother and some of my friends, they take it... not personally, but as if the doctors don't really know, and we've got to, we've got to do something else! We've got to go somewhere else! We will send you to the specialists! We will send you to California, because that's where... ! (Courtney)

Perhaps if I went to the appointment with A [husband], perhaps he would ask questions, but obviously he works so he can't come with me to those. But of course then he starts rows with doctors anyway... (Sheila)

People with CS are exposed to different opinions and sources of information, each influencing the person's own understanding. Relatives, friends and other patients routinely offered ideas and suggestions, valuable feedback, and concrete models for comparison. The complex, uncertain nature of CS means that a wide range of opinions and assertions regarding causes, outcomes and expectations were put forward. Though participants did not automatically accept these ideas and act on them, they certainly influenced their understanding and reasoning, even simply because of their availability and accessibility. Understanding was clearly not simply an individual, internal process, but one that occurred within families and social relationships.

If Doctors Don’t Listen to Me, How Can I Trust Their Advice?

Relationships with doctors strongly influenced the confidence that participants felt in the opinions and recommendations of their doctors. Participants sooner or later became patients, turning to doctors to explain their problems, hoping to get advice and solutions.
Some participants told of their experience of being dismissed and blamed, and as a consequence losing confidence and trust; others spoke of how valuable the doctor's care and listening was in terms of helping them feel understood, confident in the doctor's ideas, and positive in their expectations. Several talked of the need to manage relationships very carefully, and pay attention to the 'politics' of the medical system, in order to get the care they felt they needed. Overall, the core theme is one of relationships acting as a significant moderator on the influence of doctors' advice on patients' models and adaptation.

**Dismissive Doctors**

Participants talked in strong terms about feeling dismissed, infantilised and rejected by their doctors, who did not take their concerns and feelings seriously:

She really got to me because she didn't treat me in any -- she dismissed me, totally, she dismissed my concerns, and even though not a medically qualified person, I'm not stupid, and I do know that there was a reason. (Corrine)

So there was a lot of stress there but that's all that they kept putting it down to. Just stress and thyroid and you just felt no there is- and everything came back OK and you think, no there is something wrong. Are you looking only for one thing or are you looking for something else, and doctors would say well we're the doctors you're not the one to tell us what to do, so you just have to keep going. And you think if I keep going on about it people might listen to me. (Melanie)

It was the next time when the blood tests have proven that I was OK, then he felt that it was probably just me, run down, heavy workload, he did ask me questions like, had I had a holiday in a while, and I hadn't so.. Stress from work, and he just sort of felt, you just take care of your diet and get a life, sort of attitude. (Katy)

I went to every GP I could go to, they told me I was wasting their time. (Glenda)

I just couldn't get the damn doctors to listen to me, they just, they just wanted to give me these happy pills, to go away sort of thing! That was a bit later on, I
exaggerate, but it was, it was difficult to get through to the GP that I got troubles, you know. It might have been that he knew me [chuckles] because I had been going to the doctors very often. (Peter)

And other symptoms like very dry skin, but those seem to be very-certainly for the patient, many of these are annoying, distressing, but I don't think the medical staff really have much time for that sort of thing. But I think these have to do with the Cushing's, the aches and pains. But they never really -- take much notice of you when you are saying, I've got this really bad backache, my muscles just ache everywhere, that sort of thing, nobody really is taking that much notice. (Gemma)

For Courtney, her experience of feeling on a 'conveyor belt' in hospital meant that she could not trust her doctors to be careful, considerate of her own long-term needs and not inflict further pain on her:

And I was really, I was really- when I had the first operation, the one I should have had my adrenal gland out, which I stopped because they were going to give me a blood transfusion. I felt really really upset with the way that they actually treated me because I was supposed to have my operation at 7 o'clock or something, and I heard them talking, seven o'clock for A, 12 o'clock for B, and 2.30 for C. I felt as if I was on a conveyor belt. I felt as if the next shopping trolley would be mine, to be emptied, I thought there is no way that I can put myself through that, because what they do is that they open me up, stick a bit of blood in me, everything is hunky-dory for maybe, it might be 6 months, 10 years, I don't know, but then I find out that I've got something else wrong with me? So I just thought, I just really don't need that. Because I've been through and enough with this Cushing's.

Quite how powerful this need for 'her facts' to be properly considered, not shoehorned into a 'textbook' account, is illustrated by her striking decision to decline treatment:

[Dr T ] didn't give me any leeway at all, he just gave me, as I said, the textbook stuff, you take it or you leave it. Which I thought I'm going to have to leave this, because I don't like it. Which, I'm not sure if I should have taken that.

Did that make a difference to how you approached the illness?

Yes, it did make a difference, because I thought that, at the end of the day, you don't know what I'm going through, so I'm going to find out what I'm going through. I know what I'm going through, and I'm going to find a solution to this, whether with you or without you. I thought these are your textbook facts, and these are my facts, what I'm going through.
Trusted Doctors

Conversely, the trust that participants placed in their doctors, and their doctors’ advice, often emerged from a feeling that they had been taken seriously, and that their doctor would be looking out for their best interests:

She actually turned around to me and said.. I will have you playing football again if you want to. I said no, I feel like an 80-yr-old, I can hardly walk upstairs. She said no, you will be playing football. If you want to play football, you will be fit enough to play football. She was, she was right! [...] I believed what Dr T. told me, I believed it, and I found in it the will to want to beat it is well.. (Nick)

There are two doctors at my general practice, they are the only ones I take any notice of. They are very good, and if they don’t understand anything and think it’s related to the hypo-pituitarism, they will get in touch with O Hospital, so they are very good. The staff at O [Hospital] I feel more safe with. I don’t trust anyone at K Hospital. (Melanie).

Dr A, she seems to be more down-to-earth, and more of a people’s person. She is more.. more -- human, not this -- person that thinks she knows it all about Cushing’s, or she is high-and-mighty, what have you, she just deals with people on an individual basis, which I prefer. People that are natural and just-- not laid-back, but just can take my opinion, and not kick it under the carpet. She does ask, what’s going on with you? And how do you feel, what’s happening with your family, what’s happening with your children- she asks questions that are real, not this -- I don’t know, I think Dr T had this, it was almost as if he was reading it out of a textbook. Which I just felt was not for me. It might be formal etc for other people, but it was not for me. (Courtney)

This cluster highlights the unique value that participants placed in trusting relationships with doctors who would listed, rather than judge and dismiss them.

Evolving Understanding: Overview

Overall, the theme of Evolving Understanding captures the change and adaptation of understanding that strongly emerged from participants’ accounts. None of the
participants had any prior knowledge of CS before noticing their symptoms and being given the diagnosis. Yet they all clearly expressed a strong wish to understand and placed great value on learning about their illness, its treatment and outcomes. This theme reflects their reports of pursuing understanding through individual reasoning and experimentation, as well as feedback and information from others and trusted doctors.

Transmutation of Self

Above I illustrated how participants’ understanding of CS continually adjusted through the course of their experiences. But change and adjustment did not restrict itself to a cognitive domain of understanding; rather, it strongly emerged that participants experienced themselves as changed, and changing:

It's very - Cushing's is an awful illness really because it does so much to you, it completely changes everything, and changes you. There's no way I am the person I was ten years ago. And all the dreams I had that I wanted to do just went up in a puff of smoke. There is no way I can do—have those dreams. My life is not what I thought it was going to be- (Melanie)

Because of the shape that you are, I had these skinny arms and skinny legs, with this fat belly, these big breasts and this horrible round face. And these shoulders that were not mine, and the neck, that was- I've never seen anything like it, I was like a monster! I looked in the mirror and I couldn't actually see myself, I didn't know who it was. (Courtney)

I think [my role in the family has] changed, but not of my volition, or not of any natural force, it happened because of the damned illness.. (Peter)

So, I think that the psychological side of it is extreme, but I can only talk from personal experience, yes, it can mess you up.. entirely. (Nick).

This second broad theme brings together rich and complex descriptions of participants observing, resisting, dreading or even welcoming changes in their sense of identity.
Dissolution, the first component of this theme, collects participant’s experiences of loss of Self; Reconfiguring, the second component, refers to experiences of affirmation, protection and readjustment of Self.

Dissolution

Losing Positive Selves

The illness did not, in participants’ experiences, restrict itself to damaging the body. It took away valued roles, faculties, qualities and Self-images – one’s identity in the broadest sense - in an insidious and inexorable manner.

Losing Body Integrity

Losses of Self were often rooted in the body’s failure. Participants spoke evocatively of losing a positive body image, and its past effortless strength and stamina:

Everything seemed to be sort of slipping away, from being quite fit and well, I was finding was getting tired, I was bruising, I was putting on weight for no reason, so. Your whole self image is slipping away anyway. And mine certainly, I felt so embarrassed because I was looking so horrible anyway. If I hadn't felt so embarrassed I might have gone into the doctors a bit sooner. (Fiona).

Maybe I was a vain person in the past, I don't know [chuckles], but I think certainly that my self-confidence has been a bit dented by it. I mean, I didn't buy any clothes for ages, because I just couldn't bring myself in to the shop to buy bigger sizes. (Gemma).

My abdomen and my torso and my face were all swollen in. I felt like-- it looks like somebody had blown me up with a bicycle pump, actually. And as a result of that, I think my esteem, self-esteem had dropped tremendously. It was probably at an all-time low. And the respect for myself -- just because of my visual appearance -- I found very hard to come to terms with it, the very hard to
come to terms with it. And it's not something I'm proud of, even to look at the photographs now. (Nick)

I was- the type of person that if anything, anyone was to go out, I would be the first one on the dance floor, I'd be the first one to do whatever. I probably was the clown then, I was always the one who was always out and about. But now, I'd just prefer to stay in and prefer not to be seen. Because I am, when I looked at all the photos that I've got, and when I do look back, I just think I can't believe that person has gone, the outwards, and I have actually lost the personality inwards as well. (Courtney)

Well, my sex life was becoming nonexistent. I was very conscious of my body, so I used to hide it. The other day we actually went to the doctors and I had taken off my clothes to show the doctor. My husband was shocked because I had always covered myself up before going to bed, which I never used to do. And it changed a lot from the last time and he'd seen me. So it did affect me. (Melanie)

I wanted to be a happy person again, and I wasn't at that stage, with the way I looked, how I felt about it (Joanne).

**Losing the In-Control Self**

The ‘in-control’ Self was a particular, central aspect of identity that was severely challenged for many:

I wasn't the person I was, and I- I just felt there was nothing I can do about it. It was just sort of rolling along, and I wasn't driving the train any more. It didn't feel right, I didn't like that at all... a little part of me.. is a bit of a control freak.. I like to be doing things.. the way I want them.. but I thought that as I wasn't, it was all just washing over me.. (Peter)

I don't know, I think, I was thinking I was a failure. Because I was thinking, oh gosh, I've had the baby, how many months old, my relationship was rocky.. (Courtney)

Because I had got into such confused state, and I had become a very negative person, as opposed to a positive person, and not- I was allowing other people to run my life, as opposed to me running my life. That was in the very very late stages. The very late stages. (Nick)

All these symptoms were happening, and I was battling away and nothing was happening, so I thought well, it's something that is beyond my control. We are all in control of our bodies, see, if we eat a lot of chocolate, we get pimples, if we eat too much we get fat, if we eat too little we get thin, if we drink too much
we get hangovers, but suddenly all these things were happening to my body and
I was not in control! (Katy)

The metaphor of being robbed, or taken over, was used by other participants to convey
their sense of loss of control:

I do see it as.. the spectre at the feast, as it were. (Sorry?) The spectre at the
feast, like somebody standing over you saying, and no you won't have a good
life, you will have a bad life, and I will be here with you throughout it. And I
know that is extreme, the logical part of my brain is very active, and I can
separate the two, but emotionally, I am suffering, as much as I try to argue with
myself logically.. I do.. I see this as a burglar who has come in and taken every
one of your personal possessions from your home.

Which were the most prized possessions that it took away?
My mind…and my, and my stamina. (Corrine)

I just became so low and so stressed, that other goals just went out the window,
totally. They just take over totally, the medical and the emotional side took over
so much that you couldn't focus on anything else. It took over totally. That's how
I would describe it, like having tunnel vision, you can't think of anything else.
You can't think of anything else in your whole life, you couldn't even sit down
and watch the telly, nothing. It totally took over. Because of the emotional side
of it- not the medical side of it. The medical side of it was easy, I felt. (Glenda)

I got to a point when I wasn't.. wasn't really fighting… (Fighting..?) My
condition, this, whatever it was, which was sort of trying to take me over. It felt
like that. (Peter)

Losing the Active Self

Previously achievable activities, from working to sex, which sustained and promoted
positive views of oneself as active, energetic and productive were no longer possible to
sustain:

Has it changed me? Yes, it has changed me… I still look over my shoulder
before I go down the street... I still get annoyed when I can't do things- I had a
job to do over the weekend in the garden, which meant working on my knees for
about an hour or so, and every 15 minutes or so, I had to take a break because
my back was hurting, you know.. And as it was doing that, I was getting
annoyed. And that annoyance is not just a my back, it's about the whole thing.
Five or six years ago, I would have eaten the job, and done it, because I was
always a hands-on person. I spend my working life telling people how to build things, and from early on I decided that it was not right, it was not morally right for me to condemn a person's handiwork, what he was trained to do, if I could not to do it myself. (Peter)

I found I wasn't, I wasn't doing my job to the best of my capability. I have always been totally focused, totally driven, whereas this.. I thought no, I'm not feeling right I don't want to do this, I just wanted to shut shop, I wanted to win the lottery! (Katy)

And if I physically cannot get up to get to work, I cannot earn any money to provide [my kids] with what I want, it's just like a downward spiral. I can see that this thing can-- I feel that I can cope with what's happening now, because there was sometimes where, as I said, it just as seemed.. it was black, I couldn't.. I just didn't. It wasn't that I couldn't, I didn't want to come out of it, I just thought I've got this illness, and I'm going to be doomed for the rest of my life. (Courtney)

**Losing the Independent Self**

The sense of oneself as an independent, strong and reliable person on whom others can depend was often lost:

But, in a partnership- a marriage is a partnership- if I wasn't being a good partner in relation to not being the strong character which I am- I'm a strongly character!- but I wasn't then, and basically not having any will in any way whatsoever to really just carry on for the next day. (Nick)

Again I suppose, it's also because it has totally changed me: before I was always the rock. If anybody had problems they could always talk to me, and I would console them, but now if anyone's upset it'll waste of time talking to me because I will be in a worst state than them by the time they're finished! (Pamela)

Well before I was in control, I was the one that people came to --people still do to a certain degree but- before they would come to me and I would sort the problems out and that. But now, because I physically can't do things, people are more worried, they don't want upset me, they want to keep me.. cocooned. (Melanie)

I think the effects it's had on my relationships would have been identical if it had actually been a mental illness, I think Cushing's just happened to be thing that prompted it. I think the role problems would have occurred from any illness that made me dependent, that's where my problems start, I'm not a person who likes to be dependent, and I didn't come easily to that. (Peter)
Losing Parenthood

Dashed hopes and plans for having (or extending) a family were another significant loss:

Just, just awful -- awful. Just frustration, we had always been a great planners, we will do this, we will have 2.4 children, you know, will do -- and that wasn't in our diary, I just didn't- that wasn't what we had dreamed for, what we had hoped for. (Katy).

I have lost a certain amount of my independence. I wanted children but I can't have them now, only if I have IVF and an egg donation as well- so it wouldn't be mine, so that's hard. (Melanie)

And then I thought, well.. a big issue, the main issue was that my husband and I wanted another baby. We had a four -year-old and we wanted to.. expand on the family. And when I approached the doctor early in the year and said can we go ahead, he said no! He said not until, let's try another two months, sort of thing, let's see what the next test says. The next test found that my cortisol was rising. So he said no, hold fire, hold fire! (Katy)

Well when I first got diagnosed .. I thought about children, having children.. (Clare)

It wasn't really worth pursuing [having kids] anyway, for various other reasons, you know. We had had enough, we were getting a bit old, we had enough, but it was also the fear.. A lot of my friends who had children since the Cushing's, had difficult children. So, whether it's the children are difficult, or the parents can't cope, well, we thought, do we really want to be in that position? (Fiona)

Gaining Negative Selves

Indeed, at the same time as losing valued selves, unwanted and negative selves were piling on. Participants referred to not only *feeling as* but *taking on* damaged, devalued figures –'the cripple’, ‘the madman’, ‘the fat whale’:

So, yes... I suppose there is quite a lot of self image in it, as well. But one time I was seeing myself as a cripple, and I didn't like that.. I don't really like to see myself as a cripple...(Peter).
When you look in the mirror, you get in the bath, it's awful to see this -- my friends think I'm mad, I say 'the killer whale will be getting in the bath soon'. (Courtney)

[I considered] that I was going mad, yes, I did consider that. If I hadn't had thought that I was, I would not have taken the pills that they prescribed, despite the family pressure, if I thought it would be a total waste of time, I wouldn't have taken them. But I did, so.. It follows that I thought I was.

What did that mean for you?
... Loss of control. Visions of being locked up in a psychiatric home.

Visions?
Hm-mm [softly; nods]. (Peter)

Negative identities also emerged when symptoms were attributed to one's own weakness or failure, rather than being seen as symptoms of illness. As described above, this was particularly the case with the 'mental' symptoms of CS. Sheila, for example, attributed her emotional outbursts to 'just me', rather than the illness:

I mean, he [my husband] takes a lot from me. I'm forever shouting at him, and trying to kick him out, with my moods and everything. He says something, and that will be it, I just burst into tears. And I'll go and have a strop somewhere. This is just me.. I just.. take everything he says the wrong way at times.

Reconfiguring

To this process of erosion is contrasted a process of reconfiguring the Self. Participants talked of activities that affirmed and protected the Self and compensated for lost attributes. They reframed restrictions, renegotiated life priorities and goals and new equilibriums of roles were sought.
Affirmation and Fighting Back

Participants experienced a strong motivation to protect their identity from any further incursion by illness. They spoke of their efforts to 'fight on', to maintain a belief that they remained unchanged, the 'same person', and had not been 'taken over' by the illness:

I think for me, certainly, where I am now, the way I am now, I think is- rightly or wrongly- that it comes down to the fact that I have not let this thing takeover my life, I think there is a big- not opportunity, a big chance that something like this can, because there are probably days, times when I don't feel well for months, but I just, I always say, I'm just going to live my life. And not let this thing be the main focus. Now I think this has really helped my outlook. [...] And it just happens to be an incidental part of me, just something that has happened to me, but I don't want it to be me. That is really the main part of the experience, I have not let it rule. (Gemma)

And do you think it has changed you at all as a person, this experience? ... Probably, but I try not to... I don't think I... I don't want to become a medical bore, like people who keep talking about their illness, I have felt the need to do that, but I have resisted it. (Peter)

What is keeping me going, the fact.. well, I'm stubborn now aren't I! (Katy)

Because unless you-- even though I'm weak sometimes, I always have that kick, I sit and think no, this isn't going to take over my life, I'm going to fight, and I am, I think I am a fighter, and I think I will fight right through the end. (Courtney)

I always used to do lots of things but I didn't have to think about it. Now I do have to think but I often just do it anyway, as much as I can, but even going on holidays people say are you sure you're OK, are other hospitals nearby and things, and I say yeah, well I have got do it. I'm one of those who believe that if it's your time to go you are gonna go anyway no matter what you do. (Melanie)

It's Not Me, It's A Condition

Attributing some symptoms to the illness, rather than personal weakness or failure, means that the Self can be absolved:
If you understand some of the things- like losing your temper all the time, you
realise it's not you that's just losing your temper, it's part of a problem, perhaps
you'll learn to control yourself a bit better. (Fiona)

Did it make a difference to you that the weight was an effect of Cushing’s?

Yes, yes.. It made it easier, knowing that I’m overweight because of my
condition, I don’t eat too much, it’s because of my condition.. (Clare)

**What did it mean to you to connect the symptoms with Cushing’s?** I’m not
as low as I thought I was. Maybe I’m not such a bad personality, it’s just a
medical problem. [...] I think it might have helped me cope with it, if I knew that
it was all a medical problem. That I wasn’t going back to that heavy black hole
of depression I had when I was young. That I wasn’t going back to losing my
mind again. That there was a reason for it. (Glenda)

My self-esteem came back because I was ill and it wasn’t my fault. I knew I
wasn’t just bad (Joanne).

And in relative terms, I can look at people who are very obese, although
clinically I probably am obese, I can look at people who, what I’d term.. as
naturally fat, if you like, because they have that look about them. I don’t know
whether this is very simplistic, but to me they have this look about them, they
are very fat because they are eating, they are gorging themselves to death.
(Corrine)

**Accommodation and Re-Evaluation**

In contrast to fighting back, participants also talked about trying to achieve a balance in
one’s mind, trying to accept and live with the restrictions and vulnerabilities without
being overwhelmed:

I felt as probably it had been a learning process, maybe I had abused myself,
maybe I had worked too hard, maybe- I had a stressful life, maybe I wasn’t –
like- I had given birth to wonderful little boy, and I wasn’t doing well by him, I
was taking too much on board with work, and trying to run a household. So I
thought, well, I’ve got to take stock of life, I won’t go that- maybe it's been a
good learning curve- I won’t go that route- I will just take more care of myself.
(Katy)

My life is not what I thought it was going to be- but it's a good life, I have a
good life and I have some really good friends, and I do things, and I can get out,
I’m not in a wheelchair, and I have got all my limbs. And as long as I can keep
taking my medication, I have a good quality of life. I can keep on working, I can
still go out in the evening, I can still do things like that. But you've just got to be that extra careful, and you are different. And no matter how much you try to hide it, it's still there. (Melanie).

And then I remember kind of recently, just recently, I thought I'm going to go out, I'm going to buy something, I went out shopping with my husband and I bought some size 18 skirts. And he said to me, you're not a big person, no, no that's my size, that's my size now, I'm a size 18. Size 18 was too big!.. but I suppose that's how I am seeing myself now.. (Gemma)

I, yeah, I hide behind age as well when I excuse it to myself, that it's not just my physical condition it's also I'm getting too old to do that sort of thing, better to pay someone else to do it. So, that salvages myself self-respect. It massages it. (Peter)

**Positive Reframing**

There was also a strong emerging sense of participants reconsidering values and priorities, their outlook on life, work and other people:

More.. more relaxed, more sensitive, more in tune with other people's illnesses... and I think I am a better wife and mother, in that.. because I had been kicked in the teeth.. I believe that life is too short. OK, I've got Cushing's, my mother was diagnosed the breast cancer, there is people with a worse things and I have. I have a good friend of mine die last year, so you realise lives too short, embrace what you have, just embrace what you have. That's a positive thing that has come out of this. (Katy)

At 35 I was sort of thinking of picking up my career -in the office I used to do executorships of wills and things like that- but at this age I'm not thinking about it. I'm just sort of thinking, ah we'll do what we have to do, and enjoy yourselves, make up for some of the years we've lost really. (Fiona)

I suppose to a degree when you look back you take things for granted. You take your relationship for granted, but I think it's made a very strong relationship, to be honest. And it makes you appreciate, it has certainly made me appreciate, anyway. (Nick)

I think your personal life experiences make you aware of when other people have illnesses, of the fact that no one wants to be ill, whereas.. No one wants to be ill, and if they are ill, it's important that perhaps they want to be offered two weeks, if they need that extra week, to fully recover, then they ought to be offered that extra week. It makes you have more of an understanding, I think, personally that's how I think. (Nick)
I used to be very patient, I don't have patience any more. I see things differently now, I just want to get on and do things now, I can't be bothered to hang about, life's too short! (Melanie)

Suddenly, you get hit in the face by this... I mean.. I do- I am a Christian, I do go to church, I do believe this is maybe God saying, hold on, maybe this is him.. giving me a kick in the ass. To take stock of life.. you have all these things.. (Katy)

Overall, the theme of Reconfiguring brings together those strands of participants' narratives in which they present a sense of actively managing changes in their identity, values and priorities. Courtney evocatively illustrates the enormous, continual struggle to assert one's sense of identity and future and extract it from an all-enveloping illness:

I want to live a life, not the Cushing's life. The Cushing's life is someone that is always ill, or feeling depressed, it's always a downward thing. And I'm not a downward person. So I'm always struggling to get out of this- I want to open my flower, almost. And I'm always struggling: my head gets up, and then I'm thinking, I can't do it, because I'm not feeling 100% today, mentally am not feeling 100%, physically I am not feeling 100%. Sometimes literally, I am indoors, in bed, my mind is doing overtime, I'm never ever going to get out of this rut, because every time I try, I always fail. But I think that it is, it is that flower opening, it opens half and then it closes, and then all of a sudden -- I know that one day, I am going to get it opened, and it's going to be lovely, but I don't know how long it's going to take. But that is my aim in life, and just to get that out of the way, and then start to lead a normal life, or something half normal. At the moment, as I said, I'm living the Cushing's life. Which isn't, it isn't fun at all. (Courtney)

Social Framing of Identity

Participants framed their changes in identity within their particular social contexts, in a similar way to how social information shaped their understanding. They recognised that their own particular thoughts and efforts to shape their identity were influenced by how they were seen by others, and the roles and relationships they were in. Participants
talked about how various *relationships* –mother, husband, daughter, patient– and *contexts* –home, work, hospital– supported or questioned valued identities, thus reinforcing or weakening the person’s own sense of him or herself.

**Identity-Affirming Contexts**

Valued identities, which as illustrated above can come under threat by the illness and the demands of treatment, can be protected, affirmed and ‘shored up’ by other people. This particular form of support was expressed in a variety of ways: participants talked of containing, supportive relationships in which they felt safe, understood, validated and remembered for the person they ‘really are’:

My husband has been on this joyride with me, he has been very supportive, and he fights the obstacles with me. (Katy)

Our relationship, it didn’t waver probably because she is -- a very loving person anyway, but when I think of what she had put up with, me being in such a bad way, and emotional state, she showed her true strength there. (Nick)

I don’t think it is easy for him, he gets very worried as well when I approach these sort of surgeries that I’ve got, so.. I -- I sometimes get concerned that he is having to be very accommodating of me, I’m not taking him for granted, but he always seems to say okay, it’s all right, don’t mind me, and he seems to understand that there is something, that is -- not me! That’s the way I want it to be. (Gemma)

The first Christmas after I had the adrenals out, in the new year we all went away together, and T [friend] said it’s absolutely fantastic seeing you and J [husband] mucking about together and enjoying yourself we haven’t seen that for years. Because, I think it got to the stage where you forget how to laugh really. Everything.. is too serious really. (Fiona)
Friends and relatives were supportive by adapting to the person's circumstances without blame or pressure, by celebrating and reinforcing the person's sense of achievement and strength, or simply by remaining available and keeping connections:

Well I had some very very good friends who have always been there for me. They were very- very- considerate., some of them just came round and did things. They just accepted the way things are and worked round it. That's the way they've done it. (Melanie)

I think most of [my friends] are supportive, I think they reflect on my positive, my determination, my outlook. There are times, when yes, I have really low moments, where I think no, it's- you know, I'm going into a depression, I think no, I'm not getting on top of this. And then people will say, you have come this far are, you have got over the biggest hurdle, it's the support of my family and my husband, that has kept me through, but it also is my outlook, being determined, they respect that. I enjoy people being interested in my predicament, rather than sympathetic, which they are, and they are just going on with- for the ride with me. (Katy)

It's something you live with, most of my friends have lived through it with me, I don't have a problem there. They know I've got Cushing's, they have lived through it and they accept me for what I am. (Fiona)

Parenthood

Participants also talked of the 'grounding' effect of being a parent, and how this sustained a sense of purpose and the potential for a positive identity in the future:

What is keeping me going, the fact.. well, I'm stubborn now aren't I! ... I've got so much to live for, I've got things to do. I've got a nice husband, a loving husband, I've got a good marriage, I've got happy house, I've got- I've got a lot of out there for me, I have things to do, I have the world to conquer. I've got happy healthy boy, and I just want to be healthy for him.. (Katy)

It was probably only the fact that our children were young, and the other one was coming through, being born in April.. probably kept me going, to a degree. (Nick)

But they [kids] just don't let me be get away with being an invalid, or anything of the sort, so, I have to get up and get involved and get on with being- and my
husband as well, they don't treat me as an ill person, I don't get the chance! to
wallow in self-pity! (Gemma)

**Doctors who Listen**

Nick also credited his doctor with validating significant aspects of his identity (as a
worker, a father and husband) by asking about and showing sensitivity to what was
significant to him as a person, not just a patient:

[The doctor] was aware that my business was very very important, and that the
longer I was off work, then the business financially would be drained [...] So,
that was a factor, my enjoying my children was another factor, because -- and
experiencing contentment in my marriage, my wife not having to suffer because
of the mental state and the physical state that I was in.

**Was it important to you that she understood those factors?**

Yes, very important! And she did understand them as well. It was important, but
probably because I explained it to her. But she asked for questions as well, she
did ask for questions. (Nick)

**Identity-Damaging Contexts**

The crucial role of social context in moderating identity challenges in CS was also
illustrated when support and containment is absent or does not match the person’s needs.
Participants talked of having to face personal criticism and blame when others attributed
the illness, or the lack of rapid recovery to personal failure, greed, laziness or weakness:

And people at work said that I it looked like I was giving up, I said I wasn't
giving up, it had become worse, for some reason I was feeling worse. (Melanie)

People assume that if you are fat it's because you're greedy... (tearful) so if you
go to parties or receptions or whatever, you must only have a little plate of food
because if you've got a big plate you will know that other people will be
thinking, no wonder she's fat! So continually, all the time, you're thinking what
all the people are thinking of you. (Pamela)
So I haven't eaten this week and I'm still putting on weight, well not quite as bad as that, but you know. People saying, oh you're eating too much, that's why your putting on weight, but I wasn't. I was also terribly bruised, you would get the odd comment, does your husband knock you about, something like that. It was not so much the comments but the inference. (Fiona)

Okay, with the Cushing you have problems with your periods, which is probably more important, but most people who don't know your medical condition, just people who look at you as a particular human being, and they take an initial assessment of you, you know. Gemma)

Yeah.. he [husband] used to keep saying, shouting. Go and see the doctor! or Take your pills! Go and take something!. So finally I went and got some pills. [..] No one understood, no one else was going through it, it was only what I would tell them. And it was just like, oh well, there is nothing really wrong, just get on with it. (Sheila)

An Invisible Disability

Being judged on appearance can also result in being ignored, or blamed, because the illness –and its treatment- are invisible to the untrained eye:

And- I think it's a horrible illness, people look at me now and they say- they don't understand why people help me and stuff, they say she looks perfectly all right, you just don't want to turn around all the time to people and say I've got this that and the other, how I have to use these drugs to make me work and keep me alive. So it's very like.. ehmm.. I don't know what the word is.. It's like a disability. You look perfectly all right but you've got a disability. (Melanie)

People see you different, people expect different things for you, and when I actually look at people now.. [...] The other day I was watching that thing with Michael Jackson, we don't know what's in people's past, and what has happened to people in their lives, so when you see people, when people see me, they probably think, oh God that girl has let herself go, when she was younger she was a tiny person! And they really don't know what has happened in your life, or what is happening with your body. I've actually learnt that with having Cushing's. That you take people for face value first, and not how they really are or how they used to be. (Courtney)

It's a very insidious disease, to the world. Nobody can understand at work how ill I am, because I present in this fashion, I get up, I had some make up on, I get dressed, and they can't see how I am feeling inside, the complete exhaustion, and the fatigue, the aches and my joints and my muscles, and the dizziness and my head, of the inability to focus properly on something visually, they can't see any
about, all they see is this [gestures]. If I had a broken leg, the perception would be, oh, it's a broken leg, you know, they can relate to that. Than they can't relate to my mental.. Say, the depression, they can't relate to about. So I feel in many ways, that I am perceived as a malingerer- I'm not, but that is my.. but that- is my perception of how I am perceived. So that has affected me greatly. And just the fact that it is so serious, and I can't really get any sympathy, I suppose..

(Corrine)

Lost In The Eyes Of Others

What others -quite literally- see, or fail to see, when they look at the person's altered body, can have a profound impact. Previous sections illustrated how others’ perception could have a feedback function, notifying the person that an illness was present. But they also were experienced as implying a sense of lost identity and failure:

Because my dad is really skinny, my mum is really skinny, all my dad's family and mum's family are really skinny, so it's just like, where did you- who are you? It was that literally, because my nan came to see me from America, and she came into the room -she doesn't wear glasses or anything [...] she looked at me and just walked off. So I said, hi Grandma! And she just knew my voice, and looked at me and thought, who the hell are you? It was so shocking, she was in tears. I said, it's me. She said, I knew you were pregnant, and -- but like, who are you? It was literally, who are you. She was in tears, and I was in tears, you know. What was happening? [...] I felt so… at first, I think I shrugged it off. But when I really thought about it, I was a really upset, because I was really close to her. (Courtney)

I changed jobs in that year, when I went for my interview, which was about a October, I was fine, but when I went back in January, when I started the job, I know that I looked completely different. I remember saying to my husband, I bet they will be wondering if the if it's the same person! (Gemma)

People being concerned, saying you don't look right, and me trying and trying, and people still saying.. when I thought I was looking good, when I was heavily made up and everything, people were saying no-no... what's wrong with you? So, in that respect all of those would be conducive to me having low morale, and losing- all my confidence was sucked out. (Katy)

Even well-intentioned support offered by others may be experienced as invalidating, if it does not fit with the person's own Self-concept:
I suppose you know, because you can’t do the things that you used to do... I mean I have got to have someone to do most of my washing for me... although I supposed I should be pleased, someone is helping me out that way, but.. you know, you’d rather do these things yourself. (Lorraine)

When I look at all the photos that I’ve got, and when I do look back, I just think I can’t believe that person has gone, the outwards, and I have actually lost the personality inwards as well. Which I’ve found, which I find really upsetting because the person I am now is not who I want to be. But everyone says oh, you are lovely how you are and blah-blah-blah. And people start getting used to how I look now. Which I understand, because- well I say I understand, but I don’t understand, because I don’t want to be looking like this! And so I find it really upsetting. (Courtney)

Here, Courtney interpreted her friends’ attitude as entrenching a Self that she was trying hard to banish, forget and reject. Thus, it appears that validation is not simply a function of saying the right things, but actually fitting with the way the ill person themselves sees their identity.

**Becoming a Dependent**

As described above, the onset and exacerbation of CS often meant an interruption in activities and lifestyle, which had implications for identity. In addition, it was experienced by participants as additionally undermining if these restrictions were maintained, and almost enforced, by others’ attributions of the person as fragile, damaged, sick and needing protection:

The roles changed, they did change. [...] Before, I was in control, I was the one that people came to – people still do to a certain degree but- before they would come to me and I would sort the problems out and that. But now, because I physically can’t do things, people are more worried, they don’t want upset me, they want to keep me.. cocooned. Friends I used to look after, they don’t want to come to me because they don’t want to upset me, they want to make sure that I’m OK and safe. So they don’t come. I say that you doing that’s making me feel worse. Because you know that I’m there. (Melanie)
So, sometimes they try to take over, and I've just got to put my foot down, which I do- when it comes to that, I stick to my guns, and I try to, because I feel that people, I don't know...(What do you think they are trying to do?) Sometimes I feel that they are taking over. And I don't like people taking over. As I said, in my past experience with my partner, I felt that he was trying to take me over as well. (Courtney)

You mentioned dependency, why did you think it happened in the first place? Because by.. Either my judgment was impaired, or it was preceded to be impaired, it probably was impaired. Uhm.. And if you are making the wrong decisions, you don't make too many before people take over. Which. is an irrecoverable thing.. (Peter)

Transmutation of Self: Overview

The overall theme of Transmutation intends to capture participants' fundamental experience of having (been) changed as people -not simply as bodies- through the illness experience. The weight gain and its effects on body image, the fatigue and weakness and its effects on activity, the loss of control and independence, the loss of future, were presented by participants as having direct implications for their identity. The degree to which they experienced control over this process was variable amongst participants, since to a significant degree it depended on the physical course and outcome of the illness. However, all highlighted relationships and the social context, including relationships with doctors and the medical system, as shaping and moderating, buffering or exacerbating, the identity pressures of the illness, regardless of its course.
Chapter 4: Discussion

Participant’s stories conveyed, in a vivid and uncompromisingly honest fashion, how intrusive and often traumatic the experience of Cushing’s can be. They described their experiences of being confused, feeling damaged, and being dismissed, but also their experiences of receiving kindness, understanding, companionship and help. Using the metaphor of a dive, some spoke of emerging from the deep, grateful for the breath of air, and swimming back to shore, while others spoke of still remaining in a deep, cold, dark place of shadows. All spoke of the lasting changes that the experience of Cushing’s forced upon them – whether it was a positive new outlook on life and the future, or a wound that wouldn’t heal.
In-depth, qualitative analysis of nearly 24 hours of interviews with 15 participants revealed two broad clusters of meaning in the experience of people with Cushing’s: the *Evolution of Understanding* and the *Transmutation of Self*. In the previous section, a broad and descriptive first analysis of the material was given, which illustrated the main emerging themes and their constituting meanings. This section will return to the research questions, present the implications from the data, and discuss in the context of key theoretical and research literature. Implications for theory, future research and clinical practice will be drawn out. Finally, a critical evaluation of the strengths and limitations of this study will be followed by personal reflections on the broader process.

**How Do Patients With Cushing’s Understand Their Illness?**

Understanding Cushing’s has no clear beginning or set end-point. Throughout the illness, participants revised, refined and retested their understanding. Understanding also is something that happened between people, as much as within the individual patient. Doctors, fellow patients and the patient’s social network were responsible for varying degrees of influence on patients’ understanding. These two broad aspects will be discussed below.

**An Evolving Understanding**

No single model or idea sufficed to guide any patient through the course of CS. Instead, understanding evolved dynamically, influenced by symptoms, tests, coping, information
and advice. Within this continuous process, however, several different qualities of understanding were discerned. Although they do not form a universal or linear progression through CS, understanding did proceed and change over time, which the division into clusters seeks to convey.

The earliest understanding that participants talked about involved commonsense, everyday ailments, minor deviations from normal bodily function, or just routine 'wear and tear'. In the beginning, it wasn't even an illness for most people. Weight and fatigue, the most common initial symptoms, were usually taken at face value – 'I'm eating too much, I'm working too hard, I'm stressed'. On the whole, symptoms were considered individually, in isolation to each other. On the basis of these beliefs, some assigned the various problems low priority, others employed familiar techniques (e.g. dieting) to address these issues, including consulting doctors about these complaints.

If there was no noticeable worsening of symptoms, this state of affairs could last for a very long time. For some, this low-level management of symptoms became routine, but not without its consequences. Managing appearance through dieting and skin treatments became a big uphill struggle, particularly for women. But its stable nature made them think it was something about them – 'it's just how I was born, I thought I had big bones, it runs in the family, I just deal with it'.

When problems progressed, and the ideas and solutions applied produced no benefit, understanding needed to change. Not understanding, being confused, frustrated and uncertain, was an extremely aversive state for people. They redoubled efforts, looked
anew and further afield and asked others, including doctors. Other diagnoses were suggested, but on the whole treatments failed, leaving participants in an even heightened confusion, with no answers forthcoming. There was a sense of the problems picking up speed, but understanding getting nowhere, which was interpreted as evidence of a very dangerous and hidden illness, which would ultimately be deadly. These fears sometimes had a name—cancer, diabetes, MS—and sometimes not, but they certainly created vivid images of infirmity, madness and death.

Cognitive—‘making crass decisions’—and emotional symptoms—‘exploding like a volcano’—became entwined with the struggle to understand. At the most basic level, they interrupted the operation of one’s cognitive faculties when they were most needed: to make sense of very confusing, contradictory information. But additionally, they also added themselves to the confusion. The idea of a bodily illness causing mental as well as physical changes did not seem to crop up, except on reflection: ‘illnesses are either mental or physical’, seemed to be the implicit rule. Thus, participants struggled with the idea that, since they had ‘mental symptoms’, it could be a ‘mental illness’, or they dismissed any ‘mental’ symptoms as resulting from frustration and poor medical care, and concentrated only on the physical aspects.

This lasted for variable periods. Yet at some point something different happened—someone looked at past pictures, saw a locum doctor, got a new hunch, a test result, a second opinion. This broke the deadlock, and a diagnosis of Cushing’s was given. Participants’ descriptions of that moment of shock, delight and hope are very striking. Their understanding changed instantly: nobody knew what this Cushing’s thing was, but
everyone now knew it was something. This vindicated their past beliefs in something, and released a flood of relief and hope.

Two particular features of this new understanding stand out: that treatment necessarily existed, if a diagnosis existed; and that the treatment would be quick and compact. Looking back, some participants joked about their naïve hopes, but at the time, these beliefs were real. Control could be given over to doctors, this could be done quickly, it would be over soon, things would go back to normal – the power of this image of total restitution and restoration was great.

Participants’ first understanding of CS centred on the tumour. They believed the illness was deriving from a tumour which needed surgical removal, and they imagined restoration to take as long as it would take to recover from any surgery- ‘a couple of months’. The majority did not understand or consider that the symptoms were linked to widespread, hormonal changes of multiple systems in the body and that restoration would be in the order of a medium-term timeline -‘years, not months’. If they were told, they didn’t pick it up, or dismissed the information. Confirming evidence –the ‘zero’ cortisol the morning after treatment, the medical encyclopaedia text, the surgeon’s good spirits- was noted, but potentially disconfirming evidence –leaflets and meeting fellow patients– was shunned. This understanding was too good to spoil.

This is perhaps why understanding emerged as such an important component of the experience of CS: it was the valuable grail that was sought through so much confusion and adversity. The experience of CS is defined by the struggle to understand because it
is so difficult and elusive to do. With such a rare, confusing illness, participants were at a loss, with little existing experience and relevant concepts to apply in understanding this strange state of their bodies. When people had a real illness in their sights, they simply did not want this certainty eroded, lest it collapse again as it had so many times for them previously.

What happened to participants next after diagnosis was very variable. Everybody had a diagnosis at some point, but treatment for some was immediate, for others dragged on, needed to be repeated, was unsuccessful, caused its own problems, etc. The majority, however, at some point experienced another radical, but negative, shift in understanding: that this illness was not going to disappear as they had wished. At some point, all had to face the reality of further struggles, tests, ongoing medication, disrupted lives, and various levels of enduring disability.

Disappointment was experienced variably. For some, the disappointment and disillusion was brief and easily overcome by the feeling of progress itself, of functionality slowly returning, of things developing as doctors had said they would. For others, it was more substantial, but emerged slowly and gradually, and participants had the time to process its implications piecemeal. The trigger was sometimes the slow rate of felt progress, which contrasted with expectations. Perhaps very specific goals with specific timescales—having another child before 40, wearing a size 12 to the June wedding—were not met, prompting the whole understanding to be questioned. It was very evocative when Fiona said she had to be told by her husband and other people that she was making progress—in her eyes, it was too slow and laborious compared to what she thought and hoped
would be achieved. In some way, it was only after experiencing disappointment that CS really began to be understood for what it is.

For a few, however, the disappointment came as sudden realisation, a 'bolt from the blue', typically when talking to a doctor about test results or failed treatment, and was experienced as a crushing defeat. This sudden impact was much harder to cope with than the slow, grudging realisation that 'this was going to take longer than I thought'. At a stroke, it eliminated many valued goals and plans, and put paid to hopes. This appeared to force a return to a state of uncertainty and despair, rather than lead them to a gradual reworking of the representation of CS into a more long-term, complex problem.

When participants talked of their present and future expectations, they conveyed what can be distinguished into two distinct clusters of understanding. The first was CS as a medium- or long-term problem, with lasting but controllable effects. Participants appreciated its complexity, how it affected many bodily functions, and how it comprised mental as well as bodily symptoms. They conveyed sophisticated medical knowledge in a more-or-less coherent way, and were clear on the need for ongoing medical supervision. Some felt they needed to fight on and apply their own efforts to the problems, while others took a step back, and let the drugs and doctors do the work. Overall, they felt their knowledge and experience had prepared them well for the future, they had action plans in place, and felt prepared to face events. The future was uncertain and sometimes tinged with anxiety, but no longer terrifying.
The second cluster was CS as a vague, menacing presence. There was no solid understanding of what was happening, or why. There was much more emotion than information: a sense of being stuck, being haunted, despairing, with nowhere to go but down. CS was no longer an illness, but a gaoler. There was little motivation for action, and less hope of success. Negative cycles of failure, withdrawal and self-blame seemed established. This understanding appeared linked with a state of depression, which participants identified in themselves as a 'black hole' that had a powerful hold on them. It was clearly also apparent in their pattern of thinking, talking and relating in the interview. When they talked of their understanding of CS, these participants talked in the globally negative, hopeless language of depression.

**Evaluation, Links and Implications**

To a significant degree, these findings match and illuminate many of the central concepts of the CSM, however there are significant aspects which require further clarification.

Several key aspects of the CSM were mirrored vividly in the data. The motivation to understand health threats per se was a major aspect of the overall experience. The symmetry rule — matching labels to symptoms, and finding symptoms for labels — was both explicitly mentioned by participants, and implicitly observed. Bilevel representations (abstract and concrete) were pointed out, with participants using facts — 'it will take six months' — as well as vivid pictures of different illness scenarios — 'I will
be well by the time of the wedding'- to reason about their illness. The five dimensions of illness representations -though not explicitly extracted from participants’ accounts to avoid fragmenting the overall experience into small components- were easily apparent in participants’ reasoning, and seemed to indeed capture the intuitive way of commonsense reasoning.

The concept of coherence also appeared important in how participants understood CS. The lack of coherence in understanding prompted participants to revise their ideas and look further for answers. For example, participants talked of needing to have ideas that ‘made sense’ to them, and were unsatisfied with tests and medical treatments which in their opinion did not ‘add up’. Also, achieving coherence of understanding appeared to be linked with acceptance and adaptation, and some participants talked of ‘needing to know’ in order to ‘come to terms’ with the situation. This suggests a broader role for coherence, mirroring the suggestion by Horowitz et al. (2003) that coherence is ‘a property of the entire self-regulation system ...[which] should not be treated as a single construct that can be assessed by a single scale.’ (p.10). These basic observations support the potential for the CSM to be useful in formulating future research into adaptation in CS.

The Dynamic Nature of Representations

A particular property of the CSM that has received less research attention, the dynamic nature of representations, was highlighted by the data. Leventhal et al. (2001) suggest that ‘representations change as disease episodes unfold’ (p.29). As would be predicted
by the CSM, participants worked through their understanding of CS by moving between representation, coping, evaluation and revised representation. When their ideas failed to result in effective coping and symptom control, participants were motivated to revisit their ideas to achieve a coherent, useful understanding. Because of being such a rare, complex disorder, with no existing social representations, it required even more revision of understanding than most illnesses. Although limited by being cross-sectional and retrospective, these data goes some way in demonstrating how representations can develop radically over time, within the course of the illness.

Existing studies from a CSM perspective have generally adopted a methodology of examining associations between beliefs, coping and outcome at a specific time-point, and have not aimed to chart how this understanding develops. For example, a study on Addison's Disease (Heijmans, 1999) demonstrated two distinct clusters of illness representations, and linked each to distinct functional outcomes, but did not examine what might lead to people developing such distinct beliefs. Similarly, in examining representations of people with MS, an illness which is by definition variable and cyclical, Jopson & Moss-Morris (2003) used a cross-sectional method to examine links between representations at one time-point to social, affective and behavioural aspects of adaptation. Even in a prospective study of outcome in osteoarthritis (Orbell, Johnston, Rowly, Espley & Davey, 1998), representations were assessed only as predictor variables of post-discharge functional outcome. On the basis of this study's results it could be argued that, in the context of any illness (but especially progressive and variable ones) it is problematic to assume any fundamental stability in representations.
In effect, the dynamic aspect of illness representations has been present as a central theoretical assumption, but has not received as much research attention and elaboration.

An exception is a study by Horowitz et al. (Horowitz, Rein & Leventhal, 2003), which performed a qualitative analysis of the representations of patients with congestive heart disease (CHD). Though primarily aiming to account for the poor observed outcomes in CHD by comparing patient models to medical models, it describes several factors in how patients developed their maladaptive beliefs. In particular, it suggests that repeated experiences of controlling the illness by seeking Emergency care will reinforce an episodic, acute schema of CHD, leading the patients to adopt this strategy in future. It also suggested larger contextual factors, such as the tendency by poorer people to recourse to Emergency care in the US as a short-cut through complex medical insurance systems and expensive preventative care.

Because of its idiographic focus, the present study can highlight how understanding developed, for each person but also for the group of participants as a whole. Though the course of CS is anything but typical, a tentative developmental sequence emerged. Working in parallel with the illness course, this was a psychological sequence, with turning points defined by (sudden or gradual) changes in understanding, from ‘Everyday Causes’ through to ‘Cushing’s Is For Life’ or ‘A Vague Menace’.

The clusters that emerged do not capture a single, discrete understanding, but a similar set of representations of cause with roughly equivalent consequences, courses and timelines. So, for example, within ‘Everyday Causes’, life events, stress, minor illness
and diet imbalance were considered as causes, with roughly equivalent expectations of timeline and causes, but different implications for coping. Each cluster also had distinct emotional consequences, and implications for care-seeking, which also changed as understanding changed. For example, anxiety was lower in the earlier parts of the sequence, rising and peaking with uncertainty, brought right down by diagnosis and a compact understanding of the illness, but rising again to low levels (long-term view) or high levels (vague menace). Doctors were peripherally or circumstantially involved when participants attributed their symptoms to ‘Everyday Causes’, but played a crucial role when participants experienced rising uncertainty, and remained in an important monitoring role when participants believed in a long-term outlook.

Further investigation into the process and development of these parameters is very important to test and develop the CSM. In addition to further research, further analysis of this dataset from this perspective could bring additional aspects to light. Petrie & Weinman (Petrie & Weinman, 2003) have called for similar attention to the dynamics of how people reach decisions to seek medical care at different points in an illness episode.

Multiple Understandings

The data brought up additional questions about the CSM which require further elaboration. Patients’ shifting understanding of CS highlighted the potential for people to shift back and forth between models, and hold multiple understandings. This was illustrated by participants who, at certain points in the interview talked in a certain way
about, yet at others appeared to hold different views. Such subtle but noticeable shifts could not possibly be captured by questionnaire data but could only be observed in a qualitative study, and thus pose interesting new questions. For one, is a view of representations as stable cognitive structures in memory simply a useful shorthand for a much more dynamic, responsive, moment-to-moment variation in beliefs?

This view would be supported by the radically distinct viewpoint of Self-Categorisation Theory (SCT; Turner, Hogg, Oakes, Reicher & Wetherell, 1987) on how people reason about health and illness. SCT suggests that when a given identity (i.e. as a man, a lawyer, a wife) becomes salient, the individual will think and act in terms of the beliefs which are relevant to the particular identity. Thus, beliefs are constantly re-shaped in context, and it is not meaningful to talk of ‘enduring’ cognitive structures. Applied to health, this would imply that symptoms are evaluated not against a ‘stored’ memory, but by reference to what identities are salient at the time. Support for this viewpoint as applied to health was provided in a series of studies (Levine, 1999; Levine & Reicher, 1996). In one, men in a rugby team rated a disfiguring injury differently when they thought that their responses were being compared to women’s responses, than when the comparison was with another male group (Levine & Reicher, 1996).

On the other hand, instead of assuming that participants with CS had different, distinct sets of beliefs depending on their moment-to-moment context, one could see these data as suggesting that participants had one model which was internally incoherent. Leventhal et al. (1999) suggest that it is possible for the representational system to contain conflicting information, by maintaining an abstract (conceptual —'I know it
is...’) representation that is different from the concrete (imagery – ‘I see it as..’) component. It would also follow from schema theory that once established, a certain representation could be re-triggered in the right context, thus giving the impression of having been re-created. Finally, the Illness Perception Questionnaire, which covers the main dimensions of illness representations, shows good levels of temporal stability and reliability (Moss-Morris et al., 2002), indicating that there is some enduring component to these beliefs.

A theoretical question, then, is the extent to which illness beliefs are constructed versus stored. Does it matter where, when and to whom patients talk about their understanding, or is possible to assume that the same views will be held in different contexts? In terms of measurement, are statistically derived clusters of beliefs (as those employed by Heijmans, 1999) meaningful, or simply an artefact of the measuring instrument? In terms of clinical practice in CS, is it possible to assume that beliefs about the illness, once discussed and agreed between doctor and patient, will be retained and acted on consistently, or will they change noticeably when discussed with a spouse, or another doctor? These would be important implications for future research work to explore.

Social Shaping and Misshaping

Understanding CS was not a solitary preoccupation for participants. Through the entire course of the illness, other people’s opinions shaped patients’ understanding. Spouses, families and friends had beliefs of their own, and used them to observe and label the symptoms, and direct participants towards the care they thought was warranted –the
gym, the doctor, the dietician. What others thought became increasingly influential to participants as their own efforts and ideas wore thin and failed. Following diagnosis, doctors’ opinions and suggestions became more important, but the presence of a spouse or partner to learn with helped participants hold onto and process information. None of the participants told of anybody in their immediate circle who knew of Cushing’s, let alone anything about it, thus it was also a learning process for the people close to them. Participants spoke of spouses coming along to interviews, meeting the doctors, learning with the patient and sometimes for the patient when it was all too overwhelming. In contrast, the absence of others to share in this task left some participants feeling isolated and ‘alone with the illness’.

On the whole, other people deferred to medical opinion, and looked for their understanding to medical textbooks, support groups and doctors. As a consequence, the understanding they obtained fitted well with what doctors thought, what patients were told and what is written in the textbooks. But CS being a complex disorder with no determined cause leaves open considerable space for interpretation according to other schemas –alternative or folk medicine, religious ideas or other idiosyncratic concepts. In the group of participants, Courtney was the one to talk most extensively about the influence of such ideas on her understanding and her choices. Though essentially an exception, her account is valuable in highlighting the key role of fit between the understanding of different parties involved, and is worth a closer discussion.

Regardless of truth or utility, it emerged that when ideas promoted by different parties in Courtney’s life did not fit with each other, issues of trust, respect and collaboration
become more problematic. Who knows best? Who can be trusted? Who is on my side? Whose beliefs will I believe, and act on? Courtney was in a bind between trusting her own beliefs and practices, taking on board the opinions of her family and friends, or believing that doctors had her well-being in mind, and taking their advice. Compare:

As I said, if I’m ill, I try to do my own medication, and I do it with my children, I’m not sure if it’s right, but instead of me taking some cough medicine, I boil up some ginger and garlic. And it works, it works for me, it works for them! So I think why go to the doctors if I can do it naturally? I don’t know, I don’t –

You struggle with this.
I do now.. because I’ve actually have to put myself in the hands of the surgeons, who say they know best. And I think, do I know best? I don’t, and I don’t, I know nothing about Cushing’s. I say I don’t know, well, I know how Cushing affects me, but I don’t know what will cure me, because if I knew the cure, I would be doing it!

Here, she tries to figure out whether she can trust her doctors to know well enough. In the following illustration, she placed implicit trust in her aunt and mother, because her beliefs (not surprisingly) fitted with their suggestions:

Yes, my mother is really really good. She is quite sceptical, and I’ve got my aunt and she is a nurse as well. What they suggested -- I’m not a tablet person, I hate taking even paracetamol. So when [the doctors] said to me about living on tablets, or when I read in the books that you could be on medication for the rest of your life, I was like, I don’t think so! I said, I can’t be taking medication for the rest of my life! If I had to do it, I’d do it, but it was not something that I want to do. So, my aunt said, alternative medicine! I said oh, that will be good. I said I don’t mind taking herbs and bushes and what have you, so I said, I’d do that, whatever. I’ll do it, I’ll be the guinea pig!

Yet later on she complains about having to resist other people imposing their ideas, and affirms her trust in her doctors:

Because -- some people, my mother and some of my friends, they take it.. not personally, but as if the doctors don’t really know, and we’ve got to- we’ve got to do something else! We’ve got to go somewhere else! We will send you to the
specialists! We will send you to California, because that's where..! No! At the moment I think that it's just best if we plod on with what's happening, and just find out.. I say, the same way in those other places, I would have to go through the same regime, and the same people would have to deal with me.

Overall, this brings out the essential concept of *fit* in how social information influences the patients' own understanding. As an emerging hypothesis, this suggests that ideas which fit will be effortlessly and invisibly adopted; ones that don't will strain understanding and relationships as well.

The attitudes of others towards symptoms such as weight gain and fatigue could also weigh heavily in a less helpful direction. When others did not understand, they *mis*understood, and patients had to struggle with the illness, as well as with the blaming or confounding attitudes of others. When others believed the illness was 'all in the head', the patient's own fault or 'nothing to worry about', they forced additional confusion and doubt in the minds of the participants who were already struggling to understand what was happening. Families and spouses may even have instigated treatments –like Peter's antidepressant medication- on the basis of their beliefs. One could speculate that, if these opinions were just information about the illness, they might be far less potent in causing confusion. Yet they also contained potent judgments and attitudes towards the patient as a person, which are global judgments that are much harder to resist. This will be discussed more in a section on the social influences on identity.
Peers & Comparisons

The role of fellow patients reserved special mention, and was experienced as very valuable in shaping participants' understanding throughout the process of treatment and rehabilitation. Whether as part of a support group or individually, other patients were fellow travellers or models of how things might develop—for better or worse. The concrete, vivid nature of the information, but also the fact that it came with first-hand knowledge and experience gave it high legitimacy and implicit trust. Identification with each-other through common experiences and a common purpose, and the absence of any wider social representation or information about the disorder made this circle of 'buddies' feel like a small oasis of support and companionship.

At the same time, establishing whose experiences and advice to listen to was particularly problematic for some. Is the Syndrome the same as the Disorder? Is CS in a black person the same as CS in a white person? The higher the divergence of one's symptoms from the 'textbook', the more uncertain the comparisons appeared. Both black participants wondered whether race was a factor, but no such information was forthcoming in the textbooks. It is possible that this uncertainty reflects a deeper, enduring uncertainty amongst some participants about what is significant about this illness, and what is peripheral.

Doctors & Trust

How about the impact of doctors? Clearly, it is doctors who have the understanding, and whose opinions and beliefs count most in the context of most illnesses. It would not be at all surprising to expect patients to listen to their doctors; yet, what emerged clearly in
participants' discussion was the reasons and circumstances in which they did not listen to their doctors, because they felt dismissed and patronised. There was vocal, angry complaint expressed by participants directed against those doctors who refused to treat their initial complaints seriously. Instead, participants felt they pursued only cursory investigations, and became angry and patronising when challenged- 'we are the doctors, you won't tell us what to do'. In effect, participants felt dismissed by the doctors who made moral rather than medical judgments.

As a consequence, participants could not trust that doctor's advice, and could not incorporate it into their own understanding, while searching for a cause for this illness. Some, after several times of being told to 'get a life', gave up on pursuing doctors and just 'let things happen', which allowed the disease to progress unchecked. Others redoubled their efforts to get a proper assessment, because they realised they needed doctors, and 'there had to be an answer' to this. Though they queried their own senses sometimes -am I indeed mad, or weak, as they keep on saying?- they kept up their efforts to get properly examined and reach some -any- conclusion. However, they were still cautious and apprehensive about doctors' advice, and very careful to hold back with the way they presented themselves and their symptoms, to avoid further criticism.

This is contrasted to doctors who 'asked the questions' and listened to the answers, investigated things seriously, were proactive and became actively, personally involved. Such interactions with doctors were experienced as tremendously valuable, not only for the reassurance and containment this offered, but also because participants could now learn from their doctors, build coherence and be prepared for dealing with the illness.
constructively and collaboratively. It would appear that the two functions were linked—feeling contained promoted new learning and understanding, whereas tension and conflict maintained incoherent, defensive, maladaptive views.

**Evaluation, Links and Implications**

Three particular concepts are suggested by the data: the functions of model fit between patients and relatives, the use of social comparisons and the effects of the doctor-patient relationship in shaping beliefs.

**Belief Fit With Significant Others**

Firstly, the issue of fit between the beliefs of different parties, and its influence on representations will be discussed. The CSM assumes that an individual, from social experience and exposure to cultural stories about illness, would have available a set of available schemas to classify and understand illness. Consequently, these will be more-or-less shared by others in his or her context, ensuring some degree of fit. Indeed the fact that most participants, and the people close to them, did not clash in terms of beliefs, but supported and provided congruent information for one-another suggests that there is substantial overlap in beliefs about illness. Yet this would qualify as fit only at the most general level— for example, a basic belief in physical, rather than moral, causes of illness— and we need to examine fit at a more interpersonal, moment-to-moment level. Indeed, compared to the large volume of research into individual cognitive
representations and their outcomes, the CSM has produced less research into the social and contextual determinants and consequences of unshared representations.

Figueiras & Weinman (2003) explicitly refer to the gap in this area of knowledge about the CSM. Their study prospectively followed patients in recovery from MI, and assessed congruence between spouse and patient beliefs. They found that between 15-30% of couples had conflicting beliefs about the different aspects of the illness representation. Couples with non-conflicting, positive beliefs (i.e. more control, fewer consequences, etc) were more likely to report better adaptation and marital functioning than couples with non-conflicting, negative beliefs. Unexpectedly, couples with conflicting beliefs, did similarly well in terms of outcome. The authors argue this might be the effect of at least one partner having more positive beliefs which were the ones influencing the process. They suggest that better measurement of congruence than the very simple categorical classification they used would help clarify these issues.

This study intended to build on previous research which had indeed shown that belief conflict between spouses was detrimental to outcome. For example, Bar-On & Dreman (1987) found that the degree of congruence between patients' and spouses' causal attributions about the causes of myocardial infarction was related to better convalescence in the short term. Equivalently, Heijmans, deRidder & Besing (1999) found that in both Addison's Disease and CFS, incongruence between beliefs of spouses was associated with less adaptive coping and worse functional outcome for the patient.

Overall, there is some evidence that poor fit of beliefs between spouse and patient results in poorer outcomes. However, much less is known about the dynamics of how the illness
beliefs of one person may influence the other’s, who and under what circumstances will change their beliefs, and what helps in this process. This study suggests that relationships, trust and control all influence this process. Though only a couple of participants identified conflict, several talked of spouses and family being actively involved in adaptive efforts, not simply as sources of ideas, and thus having more substantial impact. In addition, there is the parallel issue of how carers and spouses come to form their beliefs, while living through this experience along with the patient. Several participants spoke of ‘us’ coping, acting, getting better, which indicates that adjustment happened within couples, as well as within individuals.

Clearly, the picture is much more complex than at present reflected in the literature, and this might be a fruitful area for cross-theoretical integration. There are large bodies of knowledge about social influence, belief change and decision-making, and dynamics in families, amongst others, which could be usefully brought in to shed additional light. For example, in a qualitative investigation adopting a family systems perspective, Patterson (1989) found that following cardiac surgery, patients tended to change their beliefs more in the direction of their wives, leading to increasing congruence, and also found that conflicting beliefs were connected to pre-existing relationship dynamics.

Social Comparisons

The second issue these data highlight is social comparisons. Generally, a good deal is known about the use of social comparison as a very broad coping strategy, providing both information and emotional support in a variety of contexts (Taylor, 2003). Within
the CSM, social comparisons are seen as effortful coping procedures linked to particular self-regulatory goals. Leventhal et al. (2001) suggest that ‘motivation for social comparison is powerful when somatic experience is ambiguous’ (p.36). In addition, accounts by others can provide vivid, concrete (as opposed to abstract, conceptual) evidence on all aspects of the representation.

These aspects were indeed closely paralleled in the present data. Participants sought others as evidence about how long it would last, what could be done, and for advice and suggestions about the many ‘little niggles’ that the doctors appeared to ignore. Participants who experienced the fear of ongoing diagnostic uncertainty and had little information socially available to them, prized the input of others. However, the present data draw out some further issues: how do people reason about the selection of comparison targets? For example, when the goal is general self-esteem enhancement, then downwards comparisons are made – ‘I am better off than others’. Though this was also observed, illness identity appeared to be a more salient dimension of comparison. The data would suggest that common targets are selected on the basis of perceptions of common illness identity (pattern of symptoms) rather than common label (diagnosis), or with reference to how well or poorly someone else was doing. It also follows that the less the coherence of one’s own representation, the less the potential to identify a suitable target for comparison, since it is not even clear to the person what to look for. In effect, this could mean the people who most need information could be the least likely to know what they need, let alone know where to find it. This issue would require further investigation, and is particularly pertinent for clinical practice.
Secondly, what about those instances of people not wishing to join an obviously relevant group of fellow patients? Given how rare this condition is, an external observer might consider this a very useful resource, and indeed most participants' doctors had recommended it. The data suggest there were good reasons for this, if not immediately apparent ones. As will be argued in more detail in a later section, this aversion appeared to be linked to issues of preserving one's identity from being engulfed by illness, and joining 'the ranks of the ill'. However, it may also be linked to a paradoxical self-regulatory goal: to not know about the illness, to not revise the representation that fits well with other life goals, identities and motivations. It is possible to speculate that if coherence is still fragile, and past representations very aversive, new sources of information may be avoided because of their potential to cause disruption.

Leventhal et al. (2001) indeed suggest that the longer and more strenuous the past experience with the illness, the more 'infused' with emotion (effectively, meaning) the representations could be. They suggest that 'in situations in which the exposure to a disease threat provokes very high levels of fear, and procedures to control the fear and threat are unavailable. disease schemata become catastrophic, as they integrate ultimate threats, loss of control and powerful affective expectations' (p.31). They refer to such representations as 'hot cognitions', i.e. representations laden with 'motivating potential' (p.31). It is possible to speculate that participants' aversion for new information may derive from a motivation to hold onto a new schema of the illness as 'compact and quick', and maintain maximum distance from such 'catastrophic' schemata. Again, these ideas are very important for clinical practice and will be discussed more extensively in a later section.
The Doctor-Patient Relationship

The quality of the doctor-patient relationship appeared to moderate the way participants adapted their understanding. This again is a very broad area that has been researched extensively (Weinman, 1997; Bissel, May & Noyce, 2003). Not being listened to, infantilised and depersonalised are all too common experiences of patients, with a clear and significant impact on delaying treatment, adherence, and overall quality of life (Taylor, 2003). Patients who present with vague symptoms such as gradual weight gain, emotional reactivity and fatigue, which are most often associated with stress and lifestyle, also tend to attract labels and stereotypes – ‘neurotic’, ‘hypochondriac’ – from doctors as much as from lay people. On the other hand, it is also commonly observed that patients who feel understood and empathised with may have more confidence in the advice which is given by the professional. (Weinman, 1997)

The accounts of participants tally with these findings, and shed some further light on the experience of being dismissed by doctors, and its effects on understanding the illness and coping with it. Firstly, most participants experienced self-doubt when faced with negative opinions. This could have very damaging effects on self-esteem, and confidence in one’s own ability to make sense of the situation: ‘If the doctor doubts my conclusions, how can I have confidence in myself?’ For some, this led to not seeking care, which clearly allowed the disease to progress unchecked and in turn damage long-term prospects for remission (Reitmeyer et al., 2002).
Most participants, however, talked of redoubling efforts to ‘get the bloody doctors to listen’. To do this, they looked for other opinions, but also amplified the message – ‘look, this is serious, this feels awful’. When already struggling to make sense of things and evaluate the illness and the threat it poses, having to convince doctors you are ‘really really ill’ may lead to biasing one’s own evaluations of symptoms, inflating and perpetuating anxiety, and effectively increasing the threat. If perhaps medical interactions had been less conflictual, representations may have also been less catastrophic. In addition, having to defend one’s dignity and self-esteem in the face of apparent dismissal also makes doctor-patient relationships more polarised, and undermines good communication and collaboration.

Similar links have been suggested in several studies of functional disorders, such as CFS and chronic back pain. From a social-constructionist perspective and using a Q-sort methodology, Eccleston, Williams & Stainton-Rogers (1997) examined the beliefs of patients and professionals about chronic pain. ‘Contested reality’ emerged as the strongest theme, with patients asserting the physical status of their disorder and their lack of responsibility. In contrast, psychologists emphasised habit and behaviour and sidelined physical causes, while alternative healing practitioners stressed lifestyle and indeed patient responsibility. The authors argue that, perhaps more than reflecting established beliefs, these views represent efforts to manage the apportioning of blame and responsibility. It follows that illness representations can be shaped by social motivations and concerns of the individual that have little to do with forming a ‘true’ representation. The authors conclude by asserting the importance of considering relationships in order to understand construals of illness: ‘we strive for meaning and
demand that those who are the keepers of the truth for our physical being should freely offer sense for senseless experience.’ (p.706). There appears to be a strong case for further research considering the impact of such self-presentation and relationship motives on how patients develop their understandings of the illness.

Depression, Self-Regulation and Adaptation

To what extent is the overall process of understanding CS -dynamic, framed by context-typical of that in any serious illness, and not particularly specific to CS? In general, there is much confluence between the present data and the process of understanding that the CSM proposes. As I have demonstrated above, the qualitative nature of the data particularly highlights dynamic aspects of the CSM, as well as its fundamental premises. This implies that much of the existing knowledge can be useful in developing a psychosocial framework for CS, which is the overall purpose of this study.

There is at least one element, however, which emerged distinctly in the data and is infrequently encountered in chronic illness: the challenge of depression. Though depression is often observed in chronic illness, it is characterised as secondary, resulting from the stresses and losses due to the primary illness. In the case of CS, no such clear distinction can be made. As a symptom, depression is considered (at least in the neuroendocrine literature) as a direct consequence of hypercortisolism in much same way that central obesity or osteoporosis are. Its effects are additional, but not secondary, to all other bodily symptoms, and can be readily combined with cognitive deficits (Forget et al., 2002) to create a very complex psychological experience.
There is a large body of literature linking stress, cortisol and depression (Chrousos & Gold, 1998; Steptoe, 1997; Steptoe, 1998; Vedhara et al., 2003) in complex patterns, and this issue is currently very topical in the neuroendocrine literature on CS (Sonino & Fava, 2002; Sonino & Fava, 1996). Though this study clearly cannot provide data relevant to biochemical processes, it can raise some new questions about how these may be linked to psychosocial factors. Firstly, are people who understand the emotional components to be part of the illness identity likely to be more effective in overcoming it, by means of escaping the negative withdrawal/self-blame spiral? It appeared that, of the many participants who experienced emotional symptoms, the ones who more clearly attributed them to the illness, and were not giving themselves 'a hard time', were less troubled. On the other hand, participants who had previous experiences of depression, related it to their enduring disposition and could 'see this black hole coming' were more likely to be struggling. Thus, how patients understand their symptoms may make a palpable difference to their responses, and minimise the impact of any depressive symptoms.

Secondly, relating to the CSM, what is the impact of depression on the self-regulation process, when it is a primary rather than a secondary symptom? Participants talked of 'making stupid decisions', indicating that the capacity to reason was itself impaired. How does regulation proceed under such pressure, and how does it change? Can we talk of 'self-regulation' in that state, and what are the implications for clinical practice in terms of informed consent, understanding the illness and giving responsibility to the patient to engage in rehabilitation? Leventhal et al. (2001) suggest that negative mood
affects cognitive processes in two ways: it increases vigilance and prompts attribution of ambiguous symptoms to the illness. Participant talked of becoming very frustrated with the remaining symptoms, and the lack of progress, and felt easily disappointed. This might suggest that they had inaccurate timeline representations, but also that low mood affected symptom perception and evaluation. Overall, the accounts emerging here provide very vivid demonstrations of the complex routes through which depression may affect adaptation in CS, and pose several questions for research.

Understanding: Overview and Further Questions

Patients' understanding of CS emerged as a dynamic, malleable process, with one tentative sequence described, but also subject to many moderation factors, not least the context of relationships with friends, family and doctors. The congruence of the data with existing accounts of illness representation is indeed helpful and forms a useful basis for further research into the psychosocial aspects of adaptation. Drawing on the richness and immediacy of the qualitative data, several issues pertinent to understanding were examined in detail, and several further questions emerged:

- What factors (including disease progression) influence how a patient with CS will react to the disappointment of an ongoing illness, whether with a ‘fighting spirit’ for the long term, or with despair and hopelessness -personality, mood, past understanding, social support?
o Do patients who understand emotional symptoms *qua* symptoms, rather than as 'personal failure' or 'impending madness' suffer less, and adapt better?

o Is a motivation to maintain a 'compact' understanding and minimise imagined consequences a factor in maintaining and entrenching vivid representations of quick post-surgical recovery? Conversely, what are the factors that help patients overcome any initial motivation to 'not know'?

o To what degree do illness representations remain stable, and what contextual factors (including, but not restricted to, disease progression) may influence its moment-to-moment adjustments –mood, relationship dynamics, trust, self-presentation?

o With what concerns and motivations do patients with CS approach their encounters with doctors, given their past experiences of dismissal, and what is the impact of this on their understanding and collaboration?

o What types of doctor-patient interactions are more effective in helping patients with CS manage the complexity of issues involved, and develop more adaptive beliefs?

o What ideas and beliefs do patients with CS encounter in others, and what accounts for the differential impact of those beliefs on the understanding, mood and adaptive action of patients?

o Is understanding the illness in couples just an extension of the individual’s reasoning, or does it have significantly different dimensions and processes?

o Do patients with incoherent representations find it harder to select and draw conclusions from social comparisons, and is that indeed a factor that maintains incoherence?
Several of these questions can possibly be addressed in more detail with further, more targeted analysis of the present data, and more comparisons within the sample. For example, further analysis could compare participants who were ‘crushed’ by disappointment, versus those who reacted less dramatically, and come up with more specific psychological hypotheses.

How Does Cushing’s Syndrome -and Patients’ Understanding of it- Affect Identity? How Does Identity Influence Representations?

In the previous section, fundamental questions about patients’ understanding of CS were considered. This section will examine issues of identity, and the impact of both the illness and representations on how identity changes during the course of CS. Findings will be discussed in terms of the Possible Selves Model and other theoretical accounts and empirical data. Finally, links between identity and illness representations are explored, and some emerging hypotheses proposed.

A Changed Self

Patients’ experiences of CS involved deep and lasting effects on identity, through a variety of internal and external mechanisms. Progressively during the course of the illness, valued, established aspects of the Self were gradually eroded, but participants

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12 For present purposes, unless otherwise noted, I shall continue to use the terms identity, Self, self-concept and representation of the Self interchangeably.
brought forth their own efforts to resist losses, protect the Self, and affirm its vitality. By living through the experience, participants felt they had reconfigured their outlooks and perspectives, and defined their identities in new ways. Throughout, they spoke of others who supported and accompanied them, but also of spoke of experiencing undermining social responses, not least because of the ‘invisibility’ of CS.

Dissolution of Self

Participants described the impact of CS as slowly but inexorably eroding and dissolving their identity, starting from the damage to the body and spreading to all domains of life through which they defined themselves.

Aspects of identity that were rooted in the body suffered the first wave of the assault. The obese, asymmetrical, ugly body brought confusion, fear and disgust. The face that stared back from the mirror was distorted and unfamiliar. Failing strength and stamina showed themselves in the supermarket, the tennis courts, the housework, the workplace. For the weak and fragile body, the world became a dangerous and difficult place: bones snapped like twigs and a few steps felt like a steep slope. The sense of bewilderment was palpable –what was once familiar and faithful now became alien and subversive.

For women, CS brought to the fore a particularly poignant and bitterly contested issue: having a socially desirable appearance. No self-respecting woman, as the implicit social rule goes, can ‘let herself go’. The inevitable failure to reach the accepted social standards of appearance was condemned by others, but crucially also by oneself. The
men in the group also experienced shame and embarrassment about their bodily appearance, and their loss of strength and stamina. But the women struggled much more with the changes, and their own inability to put them right. It is possible to speculate that for the women with CS, the implications of a damaged and unattractive body went deeper, and caused deeper rifts in identity.

Changes in the body’s appearance and function set off a chain of consequences that rippled through all that was important and meaningful in patients’ lives. Sexuality was inhibited through hormonal changes as well as one’s own shame and fear of self-exposure. Parenting became fraught and unfulfilling, and working was compromised and often abandoned outright. Activities and functions consequent on a positive body image—dating, socialising, going to the beach, even getting a medical examination—were avoided, and as a result the identities they sustained dwindled. Plans, hopes and prospects—to have children, an active life, career success, a mortgage, an intimate relationship—appeared more and more remote and had to be abandoned or fundamentally modified.

The slow but steady progression of the illness, while still unidentified, also eroded participants’ sense of self-control. They employed the usual self-management routines only to find that nothing was improving and problems were mounting. This inverse correlation between action and outcome formed a powerful argument against the ‘in-control’ Self—in effect, learnt helplessness. The body that was once the puppet of the will now became its master, and control was reversed. Participants were no longer in ‘the driving seat’, and felt like ‘passengers’. Independence and self-assertion soon turned into dependence and passivity. From being ‘the rock’ of their family, many felt they had
become the object of others' pity and concern. Later on in the experience, as doctors and treatments objectified the body and took over its control, this loss of control was less frightening but still estranging, and potentially causing dependency. Equally, when accommodation extended into overprotection by others, it also damaged control and independence.

The net effect of these changes was that participants felt robbed, damaged, and fundamentally changed as people. Their bodies were damaged, ugly and unfamiliar. What they did with their bodies - the lives, roles and goals they invested their energy in - was severely interrupted, forcing an enormous impact on identity. They seemed to ask themselves: If I no longer look like myself, I can no longer work as I used to, no longer be the mother I want, no longer look forward to the things I did, who am I?

While positive aspects of identity were being eroded, negative identities were creeping in through the back door. By implication, but no less powerfully, when you no longer are beautiful or capable, you are a monster and a cripple. And most terrifyingly: when you are no longer sane, you are going mad. The feeling of going mad was real and palpable for people who didn’t know quite how else to understand the experience of feeling awful, but getting negative tests from doctors and ‘funny looks’ from others. For some, this challenge had to be kept at bay for much longer than it took to diagnose and treat the illness, because the unexpectedly slow recovery again prompted question-marks about capacity, determination, and ‘being a fighter’ vs. ‘succumbing to weakness’.
Reformation of Self

While on the whole blaming the illness for the losses, participants promoted their own responsibility for fighting back, effecting positive changes and making gains. It is important to note that there did not appear to be any specific point in the course of CS where dissolution was halted, and reformation started. Pressures on identity were sustained well beyond treatment, and fighting back and resisting were amongst the first response to the first identity-damaging symptoms. However, as soon as the illness was recognised *qua* illness, identity could start to be disentangled in earnest from the shame, blame and failure: there is a real reason, and the reason isn't 'me'. On the whole, reformation of Self was presented as an active, voluntary process, that balanced and counteracted the involuntary, forcible dissolution of identity.

Participants talked in two distinct but complementary ways about how they pursued a new balance of identity. On the one hand, there was a strong push to protect identity against being taken over by illness, and affirm control and meaning. Cushing's was not to be allowed to rule, and engulf the Self, and had to be pushed away. This attitude did not deny that limitations were present, but sought to minimise changes and reverse existing damage. Living with the illness was portrayed as a constant, unrelenting fight, and participants prodded themselves to remain on a state of alert against further incursion. On the other hand, they conveyed an accepting, accommodating style. They acknowledged the impact of the illness on themselves as people and accepted that it imposed limitations on their lives. However, they saw themselves as willing and able to accommodate to these changes, and work around them. Living with the illness acquired
a sense of normality and ordinariness, in which the Self could heal. These two attitudes, though distinct, appeared complementary to each other. It appeared that participants engaged in behaviour informed by both attitudes at different points—fighting on some fronts, accepting and living with some other changes.

For some participants, this hated illness was nevertheless the catalyst for a valuable process of reconsideration and reflection on one’s identity and approach to life. Meaning was found in the whole experience as a force for positive change. Participants talked of drawing valuable lessons about one’s strength in adversity. They took away powerful examples of their own capacity to cope with adversity and survive. The assessment of impairment due to illness-imposed disability was reconsidered in a positive light, and non-impaired aspects were highlighted and reaffirmed. The value of connectedness was underlined, and new ways of relating to others were potentiated: being more empathic, more understanding, less judgmental. Participants looked anew at their close relationships, re-evaluated their positions and reaffirmed their commitments and links. They reconsidered their priorities, and reached new balances: to be more easygoing, play more, enjoy life, enjoy the moment. For some, there was even the glimmer of purpose in the whole experience, a 'message from above' about how to live life, but even people who believed in the illness as a chance event could draw lessons from it about themselves and their relationships to others. Overall, these were not simply new behaviours that participants had learnt, but effectively formed new ideologies, new possibilities and new identities.
Social Framing of Self

These twin experiences of dissolution and reformation were embedded in the social forces that support and protect, or undermine and perpetuate injury to Self. When participants talked of CS and their identity, the impact of others was as vividly present as the impact of the illness itself, or one's own ideas. Firstly, supportive influences were strong and prominent. Others who 'came along for the ride' were companions to coping and adaptation but also contained anxieties. Others would temporarily take responsibility on themselves to keep existing relationships and roles alive on behalf of the patient. Others noted the problems, but accommodated in a way that fit the person's preferences and requirements—not too protective, or not too demanding. Doctors who showed compassion not only helped understand, as argued before, but by attempting to connect the treatment to the life circumstances and concerns of the person, also sustained significant aspects of identity. The experience of a compassionate doctor promised restoration of life, as well as body. Overall, the sense was of identity being buffered, propped up and sustained by the quality of others' presence.

Secondly, two different but related social reactions were felt to be undermining: when others saw *too little* change, but also when others saw *too much* change. On the one hand, because of the invisibility of the disorder, but the very prominent dysfunction and incapacity, others (who were unfamiliar with the person's past appearance) could *see* little visible reason for it, and could only attribute it to personal weakness, lack of ability. This was, interestingly, something that several participants admitted in themselves, prior to their own experience of being on the receiving end, but they clearly
named the struggle to keep these implications at bay. On the other hand, people who
knew the person well could see too much evidence of change, and their alarm and
sadness only served to reinforce the person’s own. The experience was of the ‘true’ Self
being hidden behind ‘a mountain of fat’. It felt like one was even further lost when lost
in the eyes of others. It was clear that each individual had their own preferences about
what balance between ‘ill’ and ‘normal’ they would prefer, but overall talked vividly of
the social pressures for too much of both.

**Evaluation, Links and Implications**

The accounts of the experience of people with CS strongly mirror the ideas and findings
suggested in the extensive medical-sociology literature on lay accounts of illness
(Lawton, 2003). In a seminal article, Bury (1982; 2003) presented a view of serious
illness as a biographical disruption that forces one to face failure and death, and prompts
reconsideration of the ‘script’ of one’s life. Eccleston et al. (1997) claim that ‘central to
the experience of prolonged suffering is a fundamental challenge and threat to identity.
What is at stake is ‘self’” (p.707). Charmaz, another influential writer, derived the notion
of ‘loss of self’ from her extensive qualitative work (Charmaz, 1983). She notes that
‘serious illness forces re-evaluation and, often, redefinition of self’ (Charmaz, 2000,
p.230). Her work has explored the ways in which stigma can be injurious to the Self
directly, but also through the absence of opportunities for self-validation through work
and relationships.
Clarke & James (2003) also used a qualitative approach to explore the experiences of people with CFS. The first themes that emerged suggested that participants experienced the loss of the former Self, body functioning and relationships. They related this loss to the contested diagnosis, failure in everyday functions which gave structure and meaning to identity and goals, and the abandonment of social relations. Later themes suggested the formation of a radical new Self — better in every way, with changed values, more relationship-oriented, more balance between work and leisure, more confidence and renewed faith. The authors suggest these themes describe a process of change, from an initial overwhelming assault on the for-granted Self, people moved to reject the old Self and its values, and create a new Self. They contrast these findings with research that suggests that on the whole, it is a 'restoration' of Self that is often sought. They argue that in CFS, the lack of legitimisation through a 'serious' diagnosis damages the old Self, prompting patients to adopt a new organization and discredit past ones.

A qualitative investigation of changes in Self through the experience of chronic pain (Hellstrom, 2001) highlighted similar themes, such as the process of dynamic change, the impact of others in shaping the Self, and the Self entrapped and isolated in time and space. However, there was one significant disjunction with the findings of Clarke & James (2003) in CFS. Patients with chronic pain (who were interviewed as inpatients and thus had achieved a level of 'legitimacy') spoke strongly of 'maintaining the consistency of the past Self'. Views of past identities were positive, changes were resisted, and losses were mourned. Change was not welcome, which is in striking contrast to CFS, where patients were motivated to gain distance from their past selves.
The emerging themes from both studies are strikingly resonant with the findings in this study, particularly the process of loss, followed by reconsideration and reworking of Self. They also allow the present data to be situated between the two, with respect to the motivation vs. resistance to rework the Self. Patients with CS talked about both losing valuable past selves and looking forward to a new identity. The eventual diagnosis of a somatic illness in CS -which is what CFS patients keep hoping for- relieves much of the stigma. In comparison to CFS, this is possibly a factor that accounts for how patients with CS are less rejecting of the 'old Self', which was, ultimately, proved 'right'. But they are also different to patients with chronic pain in having a different future outlook, with significantly less entrapment and chronicity, and thus looking forward and welcoming change and reconfiguration of the Self.

The emerging themes are also very congruent with the theoretical account proposed by the Possible Selves Model (Markus & Nurius, 1986) and its elaboration in the study by Hellstrom (2001). From this perspective, the experience of the early stages of CS is one where the working self-concept becomes constituted less and less by familiar, positive selves, accrues new or reactivated feared selves, and radically curtails future selves. Diagnosis prompts a rapid reorganisation, with the many feared outcomes rejected instantly and vivid, valued selves re-engaged, but disillusion involves further changes. For patients facing uncontrollable CS, uncertainty and depression, versions of earlier feared selves can re-emerge. Overall, the shifting balance of identity emerging in the data would be captured well by the PSM, and makes a substantial contribution to the application of the PSM to issues of illness.
Research on benefit-finding in illness (e.g. Sodergren & Hyland, 2000; Thornton, 2002; Siegel & Schrimshaw, 2000; Folkman, 1997) and disability (e.g. Albrecht & Devlieger, 1999) has brought a distinct perspective and an increasing appreciation of how such difficult experiences can have positive outcomes for a significant percentage of people. The emerging themes in this study are very congruent indeed with this literature, and the overall classifications of benefits that have been proposed. For example, a study by Collins, Taylor & Skokan (1990) with patients with a diagnosis of cancer found more positive than negative effects on self-esteem (including a concept of oneself as strong and self-assured), relationships and empathy, life goals and priorities and views of the world. Similarly, Petrie, Buick, Weinman & Booth (1999) asked patients with breast cancer and patients who had suffered a heart attack to answer an open-ended question on benefits. They assigned answers to pre-determined categories, and found that up to 20-30% of patients in both groups identified positive effects in relationships, outlook on life and personal priorities.

This study usefully adds to the existing research that has demonstrated positive effects by tentatively suggesting why and how this might happen. The data suggest that positive changes can be seen as tools used in the process of adaptation, rather than just outcomes. For example, participants who understood CS and cortisol as somehow relating to ‘stress’ sometimes concluded they had to ‘keep positive’ in order to ‘not stress the body more’ and get better quicker. This draws on a commonsense belief about coping with disease by effortful mental control and is not entirely idiosyncratic to CS as has been shown in studies that highlight the ‘stress-illness’ rule: ‘I must avoid stress because it leads to illness’ (Leventhal et al., 1997) or conversely: ‘I must be positive in order to
help my body fight the illness'. It is possible that, at least in part, such changes were brought about in an effort to cope with illness, and were congruent with a commonsense understanding of the links between CS and stress. This idea is mirrored in the study by Petrie et al. (1999) who found that the most frequently cited category by cardiac patients was 'healthy new lifestyle' (68%), which is congruent with the representation of the illness (as brought on by lifestyle factors) and its future prevention (to adopt lifestyle changes, e.g. diet, work patterns and pressure, etc).

Identity: Overview and Further Questions

How patients with CS see themselves is subject to radical change throughout the process of the illness. Both external (e.g. weight, fatigue) and internal (e.g. infertility, lability of mood) changes to the body set off a process of erosion of key aspects of identity. These challenges are met by patients’ own efforts to protect, reform and reconstruct various aspects of identity, as well as the affordances and pressures placed by the social context. Existing theoretical accounts of the Self fit well with this data, and can be drawn on to understand the illness experience of CS. In turn, the data point to further questions that can be asked about CS:

- What factors determine which aspects of identity are most affected by the illness—the particular symptoms of CS, the combination of symptoms and social stereotypes of gender, race, class, previous personality?
- What is the impact of CS on female and male identity? Is it different, and is it responded to differently?
o Is the motivation to restore one’s damaged identity, and to minimise engulfment by the illness, a factor in maintaining and entrenching vivid representations of quick post-surgical recovery?

o What factors determine whether patients ‘fight’ vs. ‘accept’ the illness -or better, who uses which approach at what time, and with what ends?

o Do patients’ beliefs about CS (its causes, consequences, timeline, treatments etc) make a difference to what aspects of their identity they will reconsider and reconfigure?

o Does having a ‘fund’ of positive past selves protect against ‘loss of Self’ -are patients with more diverse, well-elaborated and positive past Selves less likely to experience engulfment and loss?

o Do patients vary in the optimum balance between being seen as ‘too ill’ vs. ‘too normal’-and which factors determine this balance?

o Which processes in medical organisations, and which types of medical interaction support identity, and which undermine it? Would changing any of these aspects make a difference?

As argued previously, many of these questions can be further elaborated by using the present material, with additional analyses, links and comparisons. The level of analysis presented here was oriented towards a broad, preliminary description, upon which these questions could be used to elaborate further.
Connections Between Illness Representations And Identity

The present data suggest that there is potential to further develop a cogent psychosocial approach to CS. In particular, this study has described changes in understanding, as well as changes in identity, as key components of the experience of CS. It has tentatively proposed potential dynamic sequences of change and drawn possible links to adaptation.

Are the present data, in turn, theoretically informative? The accounts of illness self-regulation and identity appear to have less explanatory power alone than in conjunction. At least in the context of CS, it appears difficult not to connect cognitive regulation of the illness to the parallel regulation of the Self. Yet these two substantial lines of inquiry have not been effectively combined. As argued in a previous section, the CSM places the Self 'above' and 'around' the self-regulation process, and claims its oversight, but makes no specific connections. Theoretical papers on the CSM consider the potential utility of the concept of Possible Selves (Brownlee et al., 2000) or of 'hot cognitions' (i.e., cognitions imbued with meaning) but these questions have not been particularly pursued.

Research in the medical sociology field has similarly not connected with parallel accounts proposed by the CSM and PSM. Charmaz (1987; 1995) introduces the concept of 'identity goals' as 'preferred identities that people assumed, desire, hope or plan for' (p.659). She describes how the ill person will be motivated to realise future identities, or forced to accept ones imposed by the illness in the present, an account remarkably similar to that of the PSM. More generally, in an overview of the use of narrative
research in illness, Bury (2003) distinguishes three types of possible research foci: on ‘contingent narratives’, which ‘address beliefs about the origins of disease, the proximate causes of an illness episode, and the immediate effects of illness on everyday life’ (p.263); on ‘moral narratives’ which explore the social valuations of illness, and finally on ‘core narratives’, which explore how the individual’s illness concerns draw on and reflect broader issues within society. Clearly, the CSM and much related research surveyed here would be clear examples of ‘contingent narratives’, at least in purpose if not necessarily in (narrative) form. Though Bury calls for research across various levels, from more psychological to more social, he make no reference to the literature on self-regulation.

These two broad research routes share similar goals and common concepts, but have yet to intersect. The present data can suggest such a meeting point. At its most broadest statement, I suggest that IRs are reciprocally related to the Self and particularly Future Selves (i.e., Identity Goals). In one direction, the specific way an illness is understood will determine its impact on the Self. Without some assessment of the illness, the consequences for the Self cannot be determined. There would be no fight against the illness, or any attempt to protect identity, if it was not predicted to produce negative consequences. It was clearly demonstrated that when participants attributed symptoms of CS to lifestyle stress, they appeared more likely to question their own lifestyles and give in to the pressure from others to change; yet when they attributed symptoms to ‘a tumour’, they appeared under much less pressure to modify their behaviour and approach, but rather reaffirmed their identity, asserted it to others, and protected its independence against illness engulfment. Put another way, an illness representation
(rather than just the illness itself) will affect identity to the extent that it includes representations of consequences to identity-maintaining roles, capacities and functions.

In the other direction, Identity Goals/ Possible Selves may determine (bias) the reasoning and evaluation of illness threats, selecting representations that cause less discrepancy between consequences and desired future Selves. The Self in effect sets the goals and determines constraints for the process of reasoning: ‘don’t come up with something too heavy and scary’. For the Working Self, maintaining a positive affective balance by avoiding the activation of dreaded Selves is a valid motivation, and biasing illness cognition in certain directions is an acceptable method. For example, the ‘Gone-and-Forgotten’ cluster of beliefs was clearly formed under the influence of a particular motivation: to reject, as quickly as possible, blamed, shamed, mad, crippled, dead future Selves. Overall, the motivation to maintain a positive identity equilibrium will constrain cognitive illness representations.

These are simple hypotheses and can easily be tested empirically. As one example, it would be predicted that patients with strong and well-elaborated ‘in-control’ Selves would be more likely to develop and maintain positive control representations and fewer consequence representations relating to failure and dependency, than patients with identical symptoms but less prominent ‘control’ Selves. Equally, it would be expected that amongst patients with the same level of symptoms, those with stronger negative representations of consequence, and stronger attributions to personal causation would be more likely to report higher levels of ‘loss of Self’.
It is possible that the above links were only made apparent because of the uncommon features of CS: an apparently 'functional' onset, an 'acute' diagnosis and treatment stage, and adaptation as a medium- to long-term 'chronic' illness. To paraphrase Ecclestone et al. (1997), patients with CS are eventually handed a warrant for serious scrutiny by doctors, which evades those suffering from CFS or idiopathic pain. The ensuing dynamic and radical change in understandings placed consequent identity changes into relief, and conversely it was observable how motivation deriving from Self was influencing understanding. Thus, CS may be unique and instrumental in exploring significant dimensions of the illness experience.

Implications for Clinical Practice

Though the main aims of this study were exploratory rather than applied, several specific suggestions for clinical practice can be drawn from the data. Participants spoke at length, and in detail, about their interactions with the medical system. These interactions, as argued above, have a crucial function in shaping the understanding and identity of patients, and by extension would be expected to be influential in their outcomes. It is therefore possible to draw out suggestions, grounded in the data provided by patients' accounts, about issues of service organisation and delivery, which could be useful in clinical practice.

Clearly, these do not claim to be evidence-based clinical guidelines in the sense of being based on reproducible evidence of benefit to patients. Also, because of the non-random,
local selection of participants, and the broad range of presentations and stages of the illness, no fundamental claim is made about representativeness or generalisability to other groups. Finally, the fact that issues of clinical practice were not specifically raised in the interview but only occurred spontaneously, or by extrapolation from interview data and not verified with participants, places further limits on the scope of these suggestions. Nevertheless, despite these limitations, to the extent of my knowledge this study represents the first attempt to explore, from a phenomenological perspective, patients’ experiences of Cushing’s. It is therefore in a position to offer, even speculatively, suggestions for clinical practice from a very distinct, novel perspective than other research has not previously offered. Several broad areas are considered in turn.

**Supporting the Emerging of Understanding**

The central theme of *Emerging Understanding* described the strong, ubiquitous motivation of participants to develop a coherent, useful understanding of their illness. From the point of view of the patient, understanding brings hope, control, relief and meaning to the experience. How to best support and positively influence this process is an important issue that is routinely addressed in clinical practice. The following is a list of suggestions:

Revisit Explanations. Understanding developed and progressed throughout the course of the illness. Very few, if any, participants remained static in their understanding
from diagnosis to treatment and beyond. Some participants spoke of not ‘taking in’ a lot of the information that was given to them at first, which is a common occurrence when any diagnosis is given. In addition, in CS there is demonstrable cognitive impairment of memory, concentration and planning, which would additionally limit the volume of information advisable to convey at any one time. But as the complexity and depth of their understanding progressed, people developed the capacity to learn detailed information which at first may have initially appeared too complex. At later stages, patients would also be developing their own ideas, and be keen to share them. By hearing them out, the clinician can understand how they see the illness, and assess whether any additional information is required. Overall, respecting that understanding is a dynamic, long-term process should form the basis of all attempts at educating patients. It should not be assumed that, once explained, no further discussions are needed. Instead, it would be advisable to revisit patients’ understanding, expanding and adding more detail, qualifying previous statements, and refining expectations.

Encourage Use Of Peer Support. Participants gained new information through information packs, discussions and diagrams, online resources, test results and many other sources. Yet the most consistently powerful role was accorded to the real-life models of fellow patients. By virtue of being first-hand, the information was more immediate, vivid and concrete, as well as more trustworthy for many patients. They sought out other patients to compare notes, learn new methods of dealing with problems, and ask questions. Communicating with other patients also appears to have many functions beyond simply providing information: the relief of being understood, the
respite from a world that doesn't know and may not care, a relationship free of judgments and imbalances of dependence. These fundamental functions of peer support groups are well documented (Taylor, 2003) and appear to be very powerful for patients with CS both in terms of understanding and maintaining a positive identity.

Name The Ambivalence Of Joining. Equally, however, some participants spoke of their ambivalence in joining support groups and meeting other patients. They felt they didn't have the time for it, or would not need it, drawing on their beliefs about their illness being brief. They were also quite conscious of being unwilling to join 'the ranks of the sick', thus further endangering the struggle to maintain feared identities at bay. It is probably this dimension of feared engulfment that clinical staff could usefully address. Patients could be advised that it is common for people in a similar position to be rather averse to meeting other sick people; but, as they are likely to discover, most people experience an enormous amount of support, validation and strength from others. In this way, the implicit fear of joining is brought into the open, examined, and positively reframed in a way that could make it more likely for patients to consider joining peer support groups at earlier stages.

Shape Meaningful Comparisons. Although they prized information from others, given the complexity of the disorder, many patients talked about not knowing who they can compare themselves to- is adrenal the same as pituitary? Is black the same as white? Does age matter? As well as encouraging patients to seek peers as sources of information, clinical staff could be helpful by explaining which dimensions are thought
relevant in these comparisons (e.g. age, type of treatment, need for maintenance medication, previous illness, etc) and which are not.

Using ‘Chemical’ Rather Than ‘Surgical’ Language. The strong motivation of participants to consider Cushing’s as a short-term problem that can be dealt with cleanly and quickly, emerged distinctly from participants’ accounts. It appears that participants form such an understanding partly out of a strong wish to see an end to the creeping, damaging illness process they had been experiencing for so long. Though there is clearly an important self-protective function in this, as discussed above, it also appears that for some, this understanding is formed with reference to a commonsense ‘surgical’ model, which leads them to believe and expect that removal of the tumour will lead to rapid recovery.

Explicitly explaining the distinction between tumour excision and systemic restoration may not be enough. The CSM suggests that images are frequently more powerful than facts in determining which models are adopted. Thus, clinical staff could help patients by using language that avoids implicitly reinforcing an acute ‘surgical’ model of recovery and instead use language that evokes, primes and reinforces conceptions of a ‘chemical’ model. For example, it might be useful to minimise talk of ‘getting rid of’ the tumour, and make more of the need to gradually restore the chemical balance in the body over the longer term. Here, use of metaphor can be intuitively appealing in a way

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13 I realise that by making this assertion I am also suggesting that a ‘chemical’ model is a better ‘fit’ to the medical facts, and more adaptive for patients to have. Clearly, this is not a medical opinion, but an opinion I have formed from a combination of participants’ reports as well as medical textbooks, and may not be fully accurate.
that information is not. Communicating in such a way may help patients avoid what participants frequently described as the disappointment of unrealised expectations, and losing faith in medical treatments.

**Depression As Not Just A Symptom**

Depression is a frequently observed symptom of CS. The majority of participants talked of a past experience of depression, but several participants identified themselves as depressed at the time. It was striking how these participants held much more vague, negative and hopeless beliefs about their illness and its outcome. Patients with CS who were depressed not only had biased expectations about the illness and foreshortened futures, but were often ensnared in dependent roles, had withdrawn from valued social activities, were alienated from others, and made little effort to rehabilitate. There is well-established evidence that mood affects information processing, as seems to be occurring in patients with CS. This way of thinking and experiencing the world could bring on limitations in everyday functioning above and beyond those imposed by CS itself.

Once engaged, vicious cycles are hard for an individual alone to break out of. Even when mood improves, these changes in the social fabric of their lives may maintain patients at lower levels of functioning. Thus, it is not enough to think of depression as just a symptom, to be addressed by medication alone. It is important to take into account how mood, and negative thinking, may have much wider implications in various domains and levels of the person's experience- relationships, social activity, capacity to
work, etc, which may reinforce and maintain the symptoms. Additionally, as well as medication, other actions could involve talking to families together and addressing these issues, integrating medical with psychological consultation (i.e. joint working) and framing psychotherapy as part of the package of care for CS.

**Relationship Management**

Few patients were fortunate to have a quick diagnosis from their GP, and even fewer not to encounter negative attitudes from professionals. Participants conveyed in strong terms their experiences of being dismissed and ignored by some of their doctors. This related mainly to interactions prior to the diagnosis of CS, as patients struggled to be believed, rather than labelled, by their doctors. It is likely that such polarised past relationship will affect expectations, and consequently the approach to future relationships. Asking a new patient about previous medical experiences, what was helpful and what was not, is likely to help bring up and address these issues. Accepting and validating patients’ past experiences can be very powerful in starting off a new relationship in a less polarised manner, which as argued above could have a distinct impact on how medical advice is accepted, understood and followed.

However, the feeling of being dismissed was often also experienced post-diagnosis, when doctors only cared for the ‘big’ things, and appeared not to listen or care about the daily hassles and ‘niggles’ that small problems appeared to cause to patients. Given the often very negative expectations that patients may carry about being believed and heard,
it appeared important to participants that professionals demonstrate a caring attitude by inquiring about and attempting to address these small problems\textsuperscript{14}. Even if the problems are indeed transient or uncontrollable, and even if they ultimately fail to be controlled, the attention \textit{itself} is a concrete marker of the doctor's understanding and personal attention, and could form the basis for a relationship of trust and safety.

### Whole-Person Care

Participants found it valuable to have the time, space and help to put together the physical and mental components of the illness, their thoughts about it, and reflect on the whole experience. This is distinct to just talking about symptoms, or just talking about thoughts or feelings – it is about linking the whole experience together. Corinne wished \textit{somebody} could have put things together for her:

> Everybody needs different things, I need to ramble! But to know that I am rambling, and that underneath, that I am rambling to somebody who can make proper sense of what I'm saying, and because they are listening with the different ear, and are able to pick up things that stick out like beacons to you, but which I can't see. So maybe if I'd had this chance to just talk about it, talk about the disease and how I felt about it, what it was-not medically- and just in terms of how you visualise it and feel about it, and not necessarily to have any sort of conclusion, and just the opportunity of having the somebody taking you seriously from the side of the feelings, rather than the medical point of view. And clearly the doctors don't have the time or the expertise to do that, and it's very important.

\textsuperscript{14} But also see Michie, Miles & Weinman (2003) who review the evidence for 'patient-centred' care and suggest that studies which define 'patient-centred' as activating and motivating the patient \textit{as well as} listening and understanding appear to demonstrate the better outcomes.
Professionals who related to the patient as a whole person, with a personality, life, family, hopes and goals, got glowing endorsements. Patients felt safe and understood in these relationships, and in turn were reassured more easily, and accepted their doctors’ directions more readily. Significantly, being related to as a person distinct from one’s illness is invaluable to patients who are at all times struggling not to be taken over by the illness and the negative identities it forces. When the doctor relates to you as a person, this is a powerful ally against encroaching devalued identities; conversely, when the doctor talks to you ‘out of a textbook’, it is much more likely you will reject his or her advice wholesale.

**Considering The Context**

How patients understand their illness, and how they manage to fend off identity dissolution appears to depend to a significant extent on their social context and close relationships. The beliefs and attitudes of spouses, extended family, friends and co-workers all form a powerful context that frames adaptation. Given how little is known about Cushing’s, and how powerfully people may feel about ‘a tumour in your brain’, or equally how quickly others might forget an ‘invisible disability’, this particular illness holds the potential for other people to develop a wide range of beliefs, from helpful to downright abusive.

Consequently, it is both the person and their context which is interacting with the clinician at any moment. It would be advisable for the clinician to bring this implicit but palpable third presence into the consulting room: What does your family think? What
does your boss think? Do they understand? How do they react to you being ill? Who
pushes you to keep going, and who wraps you in cotton wool? Whose ideas do you most
agree with? What other ideas have you come across? These (and many more) are
questions that can open up and bring to the fore possible tensions, and allow the
clinician to correct misunderstandings, give more tailored advice, and even get involved
in educating family and friends. The accounts from patients suggest that many will be
relieved that their wider context is taken into account, and will use the opportunity to air
other ideas and beliefs which they might have previously kept to themselves.

Critical Evaluation & Alternative Accounts

This study marks an extension of the literature on CS from more biological or symptom-
focused accounts, and introduces consideration of the utility of psychosocial factors. It
has prioritised an ‘inside perspective’ on CS, and attempted not to prematurely fragment
the experience but look for broad common experiences and concerns. Though
exploratory and idiographic, it has been grounded in theoretical perspectives which
allow further work to build on it constructively, as well as critically. It has demonstrated
essential points of contact with other research on chronic illness. Finally, it has also
generated general and specific implications for theory, further research, and clinical
practice.
A number of limitations can be identified, dealing with conceptual, methodological and analytic issues. An alternative organisation of themes is also suggested and discussed.

**Conceptual Limitations**

Firstly, it is important to acknowledge that the choice of an IPA approach has necessarily committed the study to a certain frame of reference. In terms of its overall approach, this study accepted the premise of IPA that beliefs and opinions exist independently of context, and are relatively stable properties of the person (Smith, 1996). This is in contrast to radical approaches to qualitative research such as Discourse Analysis (DA; Potter & Wetherell, 1995) which suggest that what people say depends more on how they want to be perceived, and much less on any notion of context-free cognition. DA effectively views speech as a *behaviour*, and directs the researcher to perform a functional analysis to uncover the self-presentation goals of the person, and the general social discourses that inform them. This would suggest that within the interview situation, what participants said and how they talked conveyed more about their (implicit) interaction goals –e.g. to validate one’s status as a patient, or appear as a ‘successful, in-control’ patient- and less about personal enduring thoughts and attitudes. Bury (2003) also warns against assuming that all qualitative research will necessarily convey the ‘unalloyed subjective truth of the underdog patient’ (p.281), and suggests that researchers pay attention to self-presentation and social scripts.

As a result, this study has been limited in its capacity to uncover interactive factors and discuss the broader social discourses that impact on the experience of CS. Themes have
been framed in individual, intra-personal terms and related to individual circumstances, rather than linked to wider social concerns. As a particular example, this study (at this level of analysis) presents a relatively gender-free account, thus implicitly claiming a certain degree of commonality between men and women. For example, body image and identity changes were not essentially differentiated for men and women. From a feminist perspective, however, the experience of being fat, being a patient, being dismissed as hypochondriac, need to be placed within wider social attitudes towards female bodies, power and status. As another example, the data that showed that at different points in the interview participants appeared to present different understandings about their illness, was labelled ‘multiple shifting models’, effectively an individual property, rather than investigated as a property of the interaction between the discussants at the time.

On the other hand, these were predictable, accepted limitations from the study’s outset. I would argue that the cost of adopting a DA approach, for example, would be that the resulting data would link much less easily with existing theory and research, which in fact was a key aim of this study. DA could be more useful in problematising and deconstructing established accounts, than exploring new areas for similarity. Furthermore, although clearly there are strong resonances with wider social discourses, what this study aimed to highlight was the distinctiveness of the experience of CS, and the factors of individual adaptation. Finally, I would draw a distinction between an individual perspective, which seeks to highlight the individual (and idiosyncratic) processing of social influences, as was adopted by this study, and an individualistic perspective, which only deals with intra-individual factors in understanding and adaptation (e.g. personality traits, cognitive efficiency and flexibility).
Methodological Limitations

Three points can be discerned – the appropriateness of the sample, independent verification of the analysis and the use of supporting quantitative measures.

In terms of sampling, what needs to be examined is not sample size power or representativeness, which are not primary concerns of qualitative research, but whether the process of selection was adequate for the study’s specific purpose. Elliott et al. (1999) refer to this criterion as achieving general vs. specific research tasks. The research task of this study was indeed broad and exploratory, looking at experiences of CS across an extended span of time. In this respect, the sampling was likely too conservative and could have usefully been expanded to include patients more than 36 months post-treatment. This would have likely resulted in a wider variety of perspectives, since participants would be looking back on their experiences from different points in the illness course. This argument prompted the inclusion of two pilot participants in the main group, as well using the data to refine the interviews schedule. However, the group of participants actually were widely variable along other parameters. The range in terms of years since onset of the disorder, experiences of treatment, outcomes and prospects, was wide, and indeed offered a both broad and deep descriptions of experiences of the illness.

A second potential methodological weakness is that only a small sample of interviews were analysed independently. This could potentially expose the analysis to personal bias, and an individual agenda. I attempted to address this point by broadening the
consultation with participants, peer researchers and clinicians. In addition, results were judged very congruent and illuminating by the participant to whom I sent extended notes for comment. Finally, the strong parallels with other similar research gives additional confidence in the integrity of the analytic method.

Thirdly, no other data were collected on participants, e.g. on illness status, functioning or coping. Within qualitative research, such quantitative data are often used to form comparisons between participants. Data on mood and social support could have been useful, for instance, allowing more confident comparisons between the representations and identity structures of participants scoring high vs. low on those measures. On the other hand, however, the cost would be the integrity of the phenomenological approach. Using particular scales would be to predetermine and pre-select particular comparisons, and necessarily neglecting others, rather than attending to what emerges from participants’ own accounts. This would in turn blunt claims that this study took the broadest possible view, in order to provide a basis for future research that is responsive to the actual concerns and experiences of patients. Finally, although the combination of qualitative and quantitative methods within a research domain is recommended (Barker, Pistrang & Elliott, 1994) there has been no clear methodological description and guidance on the use of such combined methods within a single study.

Analytic Limitations

It is essential to the process of qualitative analysis that new and alternative accounts can continue to be drawn and redrawn from the data. In this sense, any written account is
partial, and can be expanded or re-categorised according to the interests and needs of the analyst, in order to illuminate certain aspects of the data. This study presents a broad and descriptive set of results which is only a preliminary analysis. As was pointed out previously, several of the questions that arise can indeed be approached by closer reading of the data. Many more comparisons can be made, to suggest and refine hypotheses, for example comparing the accounts of participants who talked of forming trusting relationships with doctors with those who didn't, to suggest factors that could account for this. Additionally, particularly interesting and striking areas could be examined further. One particular example is how patients with CS experience depression, how they understand it, and how they try to deal with it. By working from this broad level and focusing on more particular aspects of the data, the analysis could proceed more deeply.

This leads on a second limitation of the analysis: bringing out individual differences versus creating a single story. Though differences between participants are acknowledged, what is presented above does not go far enough in illuminating and examining individual pathways of experience and adaptation. Clearly, there were many 'stories' of CS, but in this attempt to capture and encompass what was significant across participants, a large degree of individual variation is lost. Furthermore, the analysis at present does not provide further insight into which themes could relate to individual (trait-like) differences versus to having different experiences. For example, it begs the question of who experiences CS as a 'vague menace and who sees it as a long-term condition, and what determines who will develop such beliefs. Further analysis could
focus on bringing out complexities and differences, make comparisons and, and draw an overall picture which is more complex and subtly detailed.

On both issues, it is important to acknowledge that this analysis is the first step in an ongoing process. If more time was available, the next step in the analytic process could be expanded by using the questions presented here as the basis for further analysis and elaboration. Such analysis will indeed continue with a view to publication of several important and novel findings.

**Alternative Accounts**

The analysis presented here has been necessarily informed by my particular readings and interests at this time, and discussions with other parties. As argued above, different analytical methods such as DA, and different priorities, such as adopting a feminist or a social-constructionist perspective, would have resulted in distinct accounts.

In particular, I would like to suggest one alternative categorisation that I considered and discussed at length but finally opted not to follow. The question focuses on how to represent and situate the unmistakably strong role of social processes on both understanding and identity. The option I have followed here has portioned off the social elements relevant to identity, and those relevant to understanding. The alternative option would bring these together as a third distinct factor, with reciprocal links to both identity and understanding. This third factor, labelled ‘Negotiating the Social World’ would have highlighted how the person accepted or resisted information and implications from
others, related to doctors, and obtained support. The advantage of this organisation would have been to keep all themes involving interactions together, and avoid fragmenting what essentially feels seamless to individuals. The dual role of most relationships, informative as well as identity-shaping, could also have been more easily accommodated.

The key disadvantage, however, would be a relative loss of clarification and specificity. For example, the chosen structure allows a finer examination of interactions that centred around information and beliefs about the illness, as distinct to those that centred around social beliefs about the ill person. This allowed more specific conclusions to be drawn, and consequently a clearer assessment of the fit with theoretical accounts. In addition, the collection in a single theme of interactions between the patient and the social world would result in a third factor that would arguably be rather generic to all serious illness.

My priority is clearly to illuminate CS in particular, and choose structures which promote consideration of its unique features. Nevertheless, this highlights the potential for alternative, equally valid accounts in qualitative analysis, and underlines the need for transparency and a clear statement of theoretical and research interests.

Finally, the words used to label themes and categories were mostly my own interpretations but sometimes were used verbatim from participants’ speech. As with the argument about organisation, similarly there is an argument about language. The labels placed on meanings will shape the meaning within, and alternative words can and will have alternative impacts on the analyst as well as the reader. This shaping function of labels on content and meaning has to be recognised as an essential limitation when
seeking reproducible evidence, but when individual perspectives, vividness and resonance are sought, it is indeed an essential tool of the analysis.

There was one striking example of this in this study's process. At a late stage, Corrine contacted me with her comments on the interview transcripts which I had sent her for checking. She said that the interview 'undoubtedly' captured the most significant aspects of her experience, and was felt very positive about her participation. She then named her core experiences as 'love' (towards the people who supported her) and 'anger' (towards the incompetence of the doctors, the illness, and her own weakness). By comparison, my words -social context, support, dismissal, relief, self-blame, depression- appear very anaemic and distant. Without doubt, adopting alternative terms and labels could add different layers to this analysis, and the existing labels are necessarily only one set out of many possibilities.

Summary

This study adopted a phenomenological methodology to examine psychosocial factors in the experience of CS. It is advisedly problematic to draw 'key' conclusions from qualitative data, since as argued above re-analysis and reconsideration of the same data from a different perspective can yield distinct views. However, I would tentatively suggest that two broad and significant themes emerged from a first in-depth analysis of interviews with fifteen patients with CS. First, the Evolution of Understanding captures the strong motivation by participants to understand their illness, charts the dynamic change in their beliefs over the course of their experiences and considers the role of
social context in shaping these beliefs. Second, the Transmutation of Self describes how identity, based in the body, roles and goals, was damaged and restored throughout the experience of the illness and the social supports and pressures. These findings are congruent with existing theoretical accounts, however also pose additional questions. In particular, a hypothesis is proposed about a reciprocal role between illness representations and identity. The richness of the data was further used to draw suggestions for clinical practice.

Final Reflections

The experience of hearing the participants' experiences has been salutary. On one level, I felt very privileged to be trusted to share intimate, powerful and painful stories, and as I discussed previously felt invisibly bound to reflect this in my writing. But issues of gratitude were to develop further. Recently, two people close to me were diagnosed with serious disorders. And suddenly, roles were reversed: participant’s stories were useful in my interpreting my own experiences. Who was interpreting for whom? I had shared in some hard-earned wisdom, and this allowed me a valuable vantage on pressing personal questions. In return, participant’s words were no longer simply concepts and themes on paper, but lived realities to which I could relate. Abstract words on paper like uncertainty, unreality, future selves, coping, support, acquired vivid substance, and this brought new and richer links and connections to mind. In this way, this study has been exploratory and collaborative on more than one levels, and hopefully has achieved in using these connections to highlight a fascinating and complex domain.
References


## Appendices

<table>
<thead>
<tr>
<th>Appendix</th>
<th>Title</th>
<th>Pages</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>The Hypothalamus-Pituitary System</td>
<td>220</td>
</tr>
<tr>
<td>B</td>
<td>Research Ethics Approval Letter</td>
<td>221</td>
</tr>
<tr>
<td>C</td>
<td>Participant Information Form</td>
<td>222</td>
</tr>
<tr>
<td>D</td>
<td>Participant Consent Form</td>
<td>223</td>
</tr>
<tr>
<td>E</td>
<td>Interview Guide</td>
<td>224</td>
</tr>
<tr>
<td>F</td>
<td>Invitation to Potential Participants</td>
<td>225</td>
</tr>
<tr>
<td>G</td>
<td>Demographics Form</td>
<td>226</td>
</tr>
<tr>
<td>H</td>
<td>Participant Vignettes</td>
<td>227-30</td>
</tr>
<tr>
<td>I</td>
<td>Qualitative Analysis Level 1 Themes</td>
<td>231</td>
</tr>
<tr>
<td>J</td>
<td>Qualitative Analysis Level 2 Themes</td>
<td>232</td>
</tr>
<tr>
<td>K</td>
<td>Qualitative Analysis Level 3 Themes</td>
<td>233</td>
</tr>
</tbody>
</table>
Appendix A: The Hypothalamus-Pituitary System

Chapter 7-6: Hypothalamic-Hypophyseal Portal System

The pituitary gland (A), which works closely with the hypothalamus, is responsible for the secretion of a number of hormones that are vital to various organ systems. This small oval lies at the base of the brain, within a pocket of the sphenoid bone known as the sella turcica. The pituitary gland is also known as the hypophysis and consists of two divisions, one anterior and one posterior. The anterior pituitary (B) is also known as the adenohypophysis, and the posterior pituitary (C) is known as the neurohypophysis. The two differ in size, physiological function, and embryologic origin.

The adenohypophysis constitutes approximately 80% of the pituitary gland and originates from Rathke's pouch, which is an agglomeration of pharyngeal epithelium; for this reason, the adenohypophysis contains glandular epithelial cells. The neurohypophysis is derived from an outpouching of the hypothalamus and is of neural origin; it contains axons that arise from neurons of the hypothalamus.

The pituitary gland secretes a number of hormones, all of which are regulated by hypothalamic regulatory hormones secreted in the hypothalamus. These regulatory hormones are hypothalamic releasing hormones and hypothalamic inhibitory hormones. With the exception of prolactin, which is regulated by an inhibitory hormone, and growth hormone (GH), which acts on both inhibitory and releasing hormones, the hormones of the anterior pituitary respond to the hypothalamic releasing hormones. They reach the anterior pituitary via the hypothalamic portal veins (D). In the anterior pituitary, the group of cells that is responsible for the secretion of a certain hormone is stimulated by the appropriate hypothalamic regulatory hormone. Once stimulated, the group secretes the hormone, which acts on a specific organ, known as its target organ. The target organ then secretes its particular hormones.

Feedback system exists that helps regulate the rate of hormone secretion: If there is an excess of a particular hormone in the blood, that hormone will feed back to the hypothalamus and pituitary to decrease their rate of secretion. This process is called negative feedback, and it occurs in all hormonal systems of the body. For example, the hypothalamus secretes thyrotropin-releasing hormone (TRH), which acts on the group of adenohypophyseal cells responsible for secreting the hormone that TRH regulates—thyroid-stimulating hormone (TSH). TSH is then secreted by the anterior pituitary and acts upon its target organ—the thyroid gland. The thyroid gland then secretes its hormone, thyroxine. But say that an excess of thyroxine in the bloodstream occurs. In this case, thyroxine feeds back to the hypothalamus and the anterior pituitary and instructs them to decrease their rate of secretion of TRH and TSH.

The following table lists the major hypothalamic and anterior pituitary releasing and inhibitory hormones.

The adeno- and posterior pituitary are of neural origin, and the anterior pituitary is not. The anterior pituitary is connected to the hypothalamus via the portal circulation, which is a network of arteries and capillaries that drain into the anterior pituitary from the hypothalamus; the portal veins bring hypothalamic releasing and inhibiting hormones from the hypothalamus to the anterior pituitary. Also, the anterior pituitary hormones are manufactured within the anterior pituitary, but the posterior pituitary hormones are manufactured in the hypothalamus, and then transported via the axons, not the blood vessels, of the infundibulum (E) (the pituitary stalk) to the posterior pituitary for storage and subsequent release. Release of these hormones is not regulated by hypothalamic hormones secreted into the portal circulation, but by nerve signals from the hypothalamus that travel to the posterior pituitary via a neuronal tract called the hypothalamic-hypophyseal portal tract (F). The two principal hormones secreted by the posterior pituitary are oxytocin and vasopressin (also called antidiuretic hormone—ADH), which are synthesized by the paraventricular (G) and supraoptic (H) hypothalamic nuclei. Oxytocin stimulates contraction of the uterus during labor and stimulates contractile cells of the breast during milk ejection. Vasopressin acts on the kidney to reabsorb, and thus decrease the excretion of urine, and is therefore also known as antidiuretic hormone (ADH).

{ Color G and H different colors. }

<table>
<thead>
<tr>
<th>Pituitary gland</th>
<th>Anterior pituitary</th>
<th>Posterior pituitary</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>B</td>
<td>C</td>
</tr>
<tr>
<td>Hypophyseal portal veins</td>
<td>Infundibulum</td>
<td>Hypothalamic-hypophyseal portal tract</td>
</tr>
<tr>
<td>D</td>
<td>E</td>
<td>F</td>
</tr>
<tr>
<td>Paraventricular hypothalamic nuclei</td>
<td>Supraoptic hypothalamic nuclei</td>
<td></td>
</tr>
</tbody>
</table>

-88-
HYPOTHALAMIC AND ANTERIOR PITUITARY HORMONES

<table>
<thead>
<tr>
<th>Hypothalamic Releasing and Inhibiting Hormones</th>
<th>Action of Hypothalamic Hormones on Anterior Pituitary</th>
<th>Target Organ and Function of Anterior Pituitary Hormones</th>
</tr>
</thead>
<tbody>
<tr>
<td>Corticotropin-releasing hormone (CRH)</td>
<td>Stimulates release of adrenocorticotropic hormone (ACTH)</td>
<td>ACTH stimulates adrenal gland to secrete glucocorticoids and androgens</td>
</tr>
<tr>
<td>Gonadotropin-releasing hormone (GnRH)</td>
<td>Stimulates release of follicle-stimulating hormone (FSH) and luteinizing hormone (LH)</td>
<td>FSH and LH act on the ovaries and testes to stimulate ovarian follicle development, spermatogenesis, and steroidogenesis</td>
</tr>
<tr>
<td>Thyrotropin-releasing hormone (TRH)</td>
<td>Stimulates release of thyroid-stimulating hormone (TSH)</td>
<td>TSH stimulates the thyroid gland to secrete thyroxine</td>
</tr>
<tr>
<td>Growth hormone-releasing hormone (GHRH)</td>
<td>Stimulates release of growth hormone (GH)</td>
<td>GH has no specific target gland but acts on almost all tissues of the body, promoting growth and influencing metabolism</td>
</tr>
<tr>
<td>Growth hormone-inhibiting hormone (GH-IRH)</td>
<td>Inhibits release of growth hormone (GH)</td>
<td>As above</td>
</tr>
<tr>
<td>Prolactin inhibitory factor (PIF)</td>
<td>Inhibits release of prolactin. This factor is believed to be dopamine</td>
<td>Prolactin is involved with the initiation of milk production by the mammary glands</td>
</tr>
</tbody>
</table>

TSH — Thyroid hormones
ACTH — Adrenocortical hormones
FSH, LH — Estrogen & Progesterone
Prolactin — Milk
GH — Growth hormone
MSH — Melanocytes

The hypothalamic-hypophyseal portal system

Feedback Regulation

Inferior hypophyseal artery

The hypothalamic-hypophyseal portal system

The hypothalamic-hypophyseal portal system

Hypothalamic artery

Superior hypophyseal artery

Anterior pituitary

Posterior pituitary

Arterial supply to hypothalamus

Venous drainage of posterior lobe

Hypothalamic

artery

Corticotropin-releasing hormone (CRH)

Gonadotropin-releasing hormone (GnRH)

Thyrotropin-releasing hormone (TRH)

Growth hormone-releasing hormone (GHRH)

Growth hormone-inhibiting hormone (GH-IRH)

Prolactin inhibitory factor (PIF)
Appendix B: Research Ethics Approval Letter

The confirmation letter for the Ethics Committee.
02 October 2002

Dr L Liao
Sub Dept of Clinical Physiology
Philips House
London
WC1 1PG

Dear Dr Liao

REC Ref No: 02/0220 (please quote in all correspondence)
REC Name: Committee A (please quote in all correspondence)
Study Title: Patients' experiences of Cushing's Syndrome

The Joint UCL/UCLH Committees on the Ethics for Human Research reviewed your application on 26th September 2002. The documents reviewed were as follows:

- REC application form
- Patient information sheet (version and date)
- Patient consent form (version and date)
- Research Protocol

The members of the committee present gave approval for your research on ethical grounds providing you comply with the conditions of approval set out below:

- You do not recruit any research subjects unless you have received a notification of no objections from the R&D office.
- You do not undertake this research until the relevant Trust management approval has been received (via the R&D office).
- You do not deviate from, or make changes to, the protocol without prior written approval of the REC, except where this is necessary to eliminate immediate hazards to research participants or when the change involves only logistical or administrative aspects of the research. In such cases the REC should be informed within seven days of the implementation of the change.
- You complete and return the standard progress report form to the REC one year from the date on this letter and thereafter on an annual basis. This form should also be used to notify the REC when your research is completed and in this case should be sent to this REC within three months of completion.
- If you decide to terminate this research prematurely you send a report to the REC within 15 days, indicating the reason for the early termination.
• You advise the REC of any unusual or unexpected results that raise questions about the safety of the research.

• The project must be started within three years of the date of this letter.

NHS REC is compliant with the International Conference on Harmonisation/Good Clinical Practice (ICH GCP) Guidelines for the conduct of trials involving participation of human subjects.

Your application has been given a unique reference number please use it on all correspondence with the REC.

Yours sincerely

Dr R MacAllister
Chair

Enclosure: REC Response Form
            REC Progress Report
01 April 2003

Dr L Liao
Hunter Street Health Centre,
8 Hunter Street

Dear Dr Liao,

REC Ref. No: 02/0220 (Please quote in any correspondence)
REC Name: Committee A (Please quote in any correspondence)
Study Title: Patients' experiences of Cushing's Syndrome

Thank you for your letter dated 16th January 2003, regarding your protocol amendment. Please accept our apologies for the delay in replying to you.

There are no ethical concerns and I am happy to approve this study amendment by Chair’s Action. Your protocol amendment will be notified to the ethics committee at the next committee meeting on 24th April 2003.

Yours sincerely

Dr R MacAllister
Co-Chair
Appendix C: Participant Information Form

The study information form that was sent out to potential participants.
Information Sheet

We would like to invite you to participate in this study, which looks at the experiences of people who have had a diagnosis of Cushing’s Syndrome or Cushing’s Disease.

Various studies have described Cushing’s from a medical point of view, but few from the patient’s perspective. The aim of this research is to understand more about how this illness has affected you as a person, and what you think and feel about it. This information will help health professionals understand more about what it’s like for people with Cushing’s and may also help improve healthcare services.

This Information Sheet gives you some details about what taking part involves. You may keep this for future reference. Please read and consider it carefully.

What you would have to do

We will ask you to meet with a researcher to complete a simple questionnaire about your views on Cushing’s, and an interview. The interview will be open-ended and flexible, and participants are encouraged to describe their experiences in their own words. The whole process will last about an hour and will be audio-recorded. The meeting will be arranged at a time that suits you (sometime in the next 2-3 months), at your home, at UCL, or wherever is most convenient to you.

What happens to your information

All the information that participants give us will be made anonymous, held securely and in the strictest confidence. It will not be passed on to anyone else, not even the medical team or your consultant. The audio-recording will be transcribed into text, but names and other personal details are deleted from the data so that no one can be identified. After the study is over, the recording is erased, but other data may be kept (securely and anonymously) for future research or teaching. You can have a printed copy of the transcript if you like.

All proposals for research with human participants are reviewed by an ethics committee before they can proceed. This proposal was reviewed by the Joint UCL/UCLH Committees on the Ethics of Human Research.
Information Sheet (continued)

Will it be difficult?

The interview asks about a time that some people find stressful, so you might find some questions upsetting. It is OK not to answer any of the questions, and you don't have to speak about things you don't want to. On the other hand, some people might find that talking about their story is a helpful and enriching experience.

Do I have to take part in this study?

You do not have to take part in this study if you do not want to. If you decide to take part, you may still withdraw at any time without having to give a reason. Your decision whether to take part or not will not be disclosed to anyone else, and will not affect any of your care in any way.

What happens next?

Please complete the enclosed reply slip, and return it by post in the stamped envelope enclosed, so that we know whether or not you wish to participate. A researcher (Alex King) will contact you with a reminder if we do not hear from you within two weeks.

Who to contact for more information

For any further information, discussion or questions, please contact the researcher:

Mr Alex King  
Sub-Dept. Clinical Health Psychology  
University College London  
Gower Street  
London WC1E 6BT  
e-mail: a.king@ucl.ac.uk  
Phone: 07905 943-654 (please leave a message for me with your name and number, and I will get back to you very soon).  
Fax: 0870 167-2909
Appendix D: Participant Consent Form

The consent form signed by participants and researcher prior to the interview.
Patients’ Experiences of Cushing’s Syndrome

Consent Form

Please read and tick the following statements

I confirm that I have read and understood the Information Sheet for this study and have had the opportunity to ask any questions.

I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason and without my medical care being affected.

I understand that all data will be kept strictly confidential to the research, and no information will be passed back to the medical team or put on my medical file.

I agree to take part in the above study.

Please sign and print your name below:

Signed (Participant):

Signed (Researcher):

Name:

Name:

After the study is complete, we can give you feedback if you wish

Would you like a summary of the results to be sent to you? YES NO

Would you like a transcript of the interview discussion to be sent to you? YES NO
Appendix E: Interview Guide

A set of questions and prompts covering the main areas of the interview.
Interview Guide

- Purpose of interviews – as it explains in the information sheet, the purpose of this study is to add to the medical knowledge. I am undertaking this research as part of my doctoral dissertation.
- Confidentiality & Use of Info – The most important thing to say is that all this information is strictly confidential, and none of it gets passed back to the medical staff. I record the interview to remember and analyse it better, but I would delete all identifying information before putting anything in writing in my research.
- Comfort – It is also very important to say that, although I hope you find the questions stimulating, you may find some questions hard, and not want to answer them – this is not a problem, just tell me to move on.

Could you think back to the first time you noticed any symptoms, and can you tell me about your experience of the illness since then?

1. Experiences of illness
   a. What was it like, living with this illness?
      i. What did you notice about yourself? What did you make of any changes?
      ii. What changed in your body?
      iii. What sort of thoughts and feelings were you having?
   b. How did the illness affect your relationships?
      i. How do you think others were towards you?
      ii. What did other people close to you notice? How did others see you?
      iii. Why do you think your relationships were affected?

2. Illness Representations
   a. How did you understand the symptoms at the time?
      i. What symptoms do/ did you have?
      ii. What do you think caused the illness in the first place? (Cause)
         1. Did it remind you of any other illness you had had, or had heard about?
      iii. How long do/did you expect it to last?
         1. How could you tell?
         2. Do you think you will be entirely free of symptoms at some point?
         3. What do you think might happen next?
      iv. What do you think has been the effect of these symptoms on your life?
      v. Does/ Did it feel that things are/ were under control?
   b. What do you expect will happen with this illness in the future?

3. Identity
   a. Has the illness affected how you see yourself as a person? Have you changed as a person?
      i. What about in the future, do you expect to be a ‘different person’ in some way?
      ii. What do you hope for yourself? What do you fear?

4. Thank you very much indeed for sharing your experiences and thoughts with me.
   a. Do you think there is anything important that I didn’t ask about?
      i. Did it bring up anything new for you, something you hadn’t thought about before?
   b. Are there any concerns about the interview?
      i. What did you think of our conversation?
      ii. How did it feel having it?
      iii. Do you have any questions for me?

5. Conclude the interview.
Appendix F: Invitation to Potential Participants

The invitation-to-participate letter that was sent to potential participants identified on the clinical records.
Dear ----

We are writing to invite you to participate in a research project on Cushing's. This study is aimed to help us learn more about patients' experience of living with Cushing's Syndrome (or Cushing's Disease).

An Information Sheet, which tells you about what participation involves, and gives contact details for the researchers, is enclosed.

Whether or not you decide to participate will have no effect on your medical treatment. All the information you give the researchers will be strictly confidential to that project, and no information will be passed back or put on your medical file.

Could you please complete and return the reply slip in the enclosed envelope to let the researchers know whether you are interested in participating or not.

Yours sincerely,

Dr Gerard S Conway
Consultant in Endocrinology

Mr Michael Powell
Consultant Neurosurgeon

Please Note - We are contacting people who have been seen by Dr Conway and/or Mr Powell. Occasionally, when we undertake this kind of project, letters go out in error. If you think this has happened please inform our office.
Appendix G: Demographics Form

The demographic details Form that participants were asked to fill out.
Demographic Information

For the purposes of this study, it is useful to have some basic information on the background of people who take part. This information, as all with any and all information you give, is strictly confidential.

Please answer the following questions:

1. Are you male or female? Male Female

2. What is your age? ___ years

3. Marital status: (circle one)
   - Single
   - Living with partner
   - Married
   - Previously married, now single

4. Do you have children? No Yes, age(s): __________________

5. Do you own or rent a home? Own/Buying Renting/Sharing

6. Does your household own or have the use of a car? Yes No

7. Education: (circle the one which describes your highest qualification)
   - School, no qualifications
   - GCSE or similar
   - GCE A-level or similar
   - Higher Education or similar
   - University Degree or similar
   - Postgraduate degree

8. Current employment: (indicate one)
   - Full-Time
   - Part-Time
   - Off work on sickness/disability benefits for ____________ months
   - Unemployed, for ____________ months

9. How would you describe your ethnic group? (indicate one)

   White
   - British
   - Irish
   - Other White background

   Asian & Asian British
   - Indian
   - Pakistani
   - Bangladeshi
   - Other Asian background

   Black & Black British
   - Carribean
   - African
   - Other Black background

   Mixed
   - Black Carribean/White
   - White/Black African
   - Asian/White
   - Other mixed background

   Other Ethnic Groups
   - Chinese
   - Greek/ Cypriot
   - Any other ethnic group, please specify: __________________

Thank you for completing these questions!

CONFIDENTIAL
Appendix H: Participant Vignettes

Though describing the sample through summary statistics allows the reader to judge its composition, homogeneity and potential generalisability, those criteria are less central to this particular study. Given that this is an exploratory, phenomenological investigation of illness experiences and beliefs, it is more important to fully convey the broad range of participants' experiences. This will better allow the reader to situate the sample, have an overview of the types of experiences that participants have had, and evaluate the analysis with reference to individual stories. For these reasons, the following table will present brief vignettes of all participants in a way that, although unsystematic, that attempts to reflect the main events and concerns of participants. All names and identifying details have been changed:

---

As Cushing's is such a rare disorder, individuals could conceivably be identified more readily, but this would still require access to medical records or other confidential information. Nevertheless, all details presented here will be deliberately masked.

15
Melanie is in her late 30ies, and is white British. She noticed her first symptoms, especially fatigue and weight gain, long before the diagnosis and surgery some years ago. She is married, but the illness interrupted any plans for children. She endured many post-surgical complications and takes medication for hypopituitarism. She has returned to part-time work, which she says is ‘as much as she can cope with’.

Fiona is a white British woman in her late 50ies. She experienced major life events prior to the first symptoms of Cushing’s. It took nearly a decade of checkups and tests to get a diagnosis, then establish the source of the hypercortisolism. Adrenal surgery finally helped, and she now feels ‘as well as she will ever be’, though she has given up her work in the family farm. She is married, without children.

Pamela is a white British woman in her late 30ies, who had been working in the catering trade. She ‘had been coping with the symptoms’ such as weight and hairiness since her teens, when she was diagnosed with polycystic ovaries, and had been in employment and in a relationship. The diagnosis of Cushing’s came as a surprise to her, and post-surgically she has experienced many complications which have led her to question the need for the operation. She has stopped working and lives with her parents.

Clare is white British, in her 50ies. She had been hiding her hairiness and ‘despised’ her weight since very young, and was only diagnosed incidentally by the GP. Unclear tests and failed operation meant she was on medication for more than a decade. She had a series of sessions with a CPN some years back, because of depression, but ‘it didn’t help’. She has had several operations more recently, and just the morning of the day of the interview she received a letter from the Consultant saying all her test results now were ‘perfect’. She lives alone and has never been in a relationship.

Katy is a white Irish woman in her late 30ies, ‘happily’ married and previously working in a sales job where she ‘lived in her car’. She noticed her symptoms the same year as giving birth to her first child. Private tests led to diagnosis and surgery ‘the quickest that doctor had ever done it’. She recovered well, and decided to stay at home. She recently has had some test results suggestive of a recurrence, and was due for an appointment with the Consultant the week following the interview.

Courtney is a black British Caribbean woman in her early 30ies, who had been working in an office and living nearby to her extended family. The first symptoms coincided with having a premature baby, a relationship break-up and a bereavement. Multiple initial tests were all negative and ‘falsely reassuring’. She declined having adrenal surgery when Cushing’s was diagnosed some years ago. More recently she had two unsuccessful operations. She was due the following week for an appointment with the surgeon for another go at adrenal surgery.

Sheila is white British, in her 30ies, married with two children. She noticed the first symptoms, especially sweating, weight and fatigue when she had her second child. She got an immediate diagnosis from her GP, and following surgery is on medication, but ‘hates it’ and has noticed little benefit. She said she had been feeling depressed ‘for a long time’ and is currently on antidepressant medication from her GP.
Glenda is in her early 30-ies, white British, and identified herself as ‘an epileptic’ since her teens. She said she had a terrible, unexplained depression in her late teens, for which counselling helped, and she had since been helping manage a business. She felt that witnessing a family member have heart attack brought on diabetes and Cushing’s, but the cocktail of drugs and the multiple confusing symptoms delayed diagnosis. She recently had surgery, and was due for a post-surgical follow-up the week following the interview.

Peter is a white British man in his 60ies. He is married with three adult children, and has worked throughout the illness in a professional job. A few years back he noticed symptoms which were initially put down to ‘craziness’ and he was put on antidepressant medication, but he was very afraid he had cancer. He sought private treatment and following a successful operation has great faith in his doctors.

Joanna is in her 50ies, and is white British. She is married with three adult children. About five years ago she experienced the first symptoms, which she put down to menopause. After deciding to see a doctor for a persistently blocked nose, she underwent various tests and finally got a diagnosis of a large pituitary tumour. This was removed partially 18 months ago, and she underwent further radiotherapy. She was glad to leave her job and be at home with her husband and grandchildren. She is now feeling, and has had tests to show, that the tumour is growing again, and is expecting further investigations.

Ahmed is Afghani, in his late 20ies. He came to the UK with his father seeking asylum, and his appeal is due to be heard soon. His main symptom was weight gain since his early teens, and headaches prompted him to seek treatment in Iran, where he had two operations. The weight remained, so when he came to the UK he asked his GP for a referral, but seeing the Consultant took 1½ years. One year ago he had further surgery and has noticed great improvement. He hasn’t heard from his five siblings and his mother since coming to the UK.

Gemma is black British African, in her late 30ies. She first noticed symptoms when at University, and had a successful trans-frontal surgery. She recovered fully, trained and worked professionally in finance, and got married. She thought the tumour was gone until she had a long series of recurrences and treatments in the past few years. She links these with being pregnant with two of her three children, though doctors have suggested this isn’t the cause. She has had various forms of surgery and radiotherapy, but the problems keep recurring every several months. She was schedule for yet another operation the day following the interview.

Nick is a white British man in his late 40ies. He was remarried with young children and ‘leading the good life’ when symptoms appeared and progressively became very severe, before diagnosis and surgery about a decade ago. He made a good recovery, returned to running his business, but had a recurrence a couple of years ago. He was again treated surgically, and Nick now ‘trusts his doctors completely’. At the time of the interview there were some very early signs of yet another recurrence.
Lorraine is a white British woman in her late 50ies. She said she suffered from depression for ‘a very long time’, and has arthritis pain for some years now. She lives with her husband and their adult son. Cushing’s was noted and investigated in a hospital follow-up a couple of years ago, leading to an operation a few months ago. She has since noticed some improvements in weight, though she is concerned that it is now going up again. The day before the interview she had a dynamic test of her pituitary function.

Corinne is white British, in her early 50ies. She described a long history of relationship problems and personal psychological difficulties. Initially she connected the symptoms to the polycystic ovaries she had for many years, and the weight gain with her eating and mood problems. The diagnosis came as a shock, but ‘the real shock’ was realising, after surgery a couple of years ago, that Cushing’s could be a long term problem. She has since experienced many medical problems and depression, and was soon due to have a first appointment to see a psychologist.
Appendix I: Qualitative Analysis Level 1 Themes

The following shows an example of a 1st level organisation in the analysis. The first six themes are included, this particular interview stretched to 32 1st level themes.
Level 1 Analysis

Each box contains a preliminary 1st level theme. The shaded areas are the 1st level theme labels. Within boxes, quotes sometimes also have labels. Numbers denote transcript line numbers. Only six of the full list of themes are shown here.

### 1 Symptoms
/ Little niggly things

<table>
<thead>
<tr>
<th>Theme</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Major and minor mental and somatic manifestations</td>
<td></td>
</tr>
<tr>
<td>7-12</td>
<td>I became very very tired. I had a miscarriage and my periods became very irregular, so I went to the doctor's saying that I had put on weight so I had to change my diet and. I had been going to the gym three times a week – a very active life doing also to things and finding things more and more difficult to do. I got depressed as well, started getting quite a few spots on my face and I was getting hair down the side of my face.</td>
</tr>
<tr>
<td>29-32</td>
<td>And after that she said you'd been having constipation, feeling unwell and it's just little things- it was a continual thing, little niggly things, it just wouldn't go away</td>
</tr>
<tr>
<td>271-3</td>
<td>They say you have mood swings, and I think well oh no I didn't. I've coped with it really well. But I suppose, I've gone off my handle a few times, which I never used to do.</td>
</tr>
</tbody>
</table>

### 2 Overwhelming body change
/ Depressing fat body

<table>
<thead>
<tr>
<th>Theme</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Extracted from (1) because it appears to be a more salient symptom and central concern</td>
<td></td>
</tr>
<tr>
<td>12</td>
<td>It was mainly really the weight</td>
</tr>
<tr>
<td>53-4</td>
<td>I had noticed humps on my back and my face had gone really round</td>
</tr>
<tr>
<td>182-3</td>
<td>Well the fat really depressed me. I mean I was 36 and I went up to 42. I had a figure before like hourglass then I was going straight up</td>
</tr>
<tr>
<td>188-</td>
<td>My body was really really depressing me. My clothes started to get tight, I couldn't stop eating any of the bad things because I wasn't eating them. So I just didn't know what to do, I was going to the gym more trying to see if that helped. I did try to do everything I could myself and nothing happened.</td>
</tr>
<tr>
<td>223-7</td>
<td>Shocking, shameful body</td>
</tr>
<tr>
<td>Well my sex life was becoming nonexistent. I was very conscious of my body, so I used to hide it. The other day we actually went to the doctors and I had taken off my clothes to show the doctor. My husband was shocked because I had always covered myself up before going to bed, which I never used to do. And it changed a lot from the last time and he'd seen me. So it did affect me.</td>
<td></td>
</tr>
<tr>
<td>241-4</td>
<td>I didn't want to go out as much either. My friends said I didn't go- If a friend said come on, let's go out, I said no I'm too tired. Because none of my clothes fitted right, I just felt very conscious of the way I looked.</td>
</tr>
</tbody>
</table>
Active life hard to maintain
/ insipid deterioration

9-10 I had been going to the gym three times a week - a very active life doing also to things and finding things more and more difficult to do

45-47 I'm living in the bungalow so I didn't notice the stairs so much - I only noticed that when I went to the gym and I found that I was finding it harder and harder to get up them

282-3 I just felt so ill and so lethargic and I felt this isn't normal

183-8 And I became very week, I couldn't pick things up, and very lethargic. Before I was going to work at 6 coming back at 10 then having dinner, then sometimes going out in the weekend or doing the windows or working on the garden and doing the paperwork. I was in a continual go and then all of a sudden all I could do was lie down, go to sleep.

208-9 Forced towards alien new lifestyle
Yes, it was not like the hours I used to do, I would go in for 8.30 and go home, finish at 5, which was very alien to me.

Life's thunderstorms
/ life events

Gives the image of a calm coper in the eye of a thunderstorm

290-7 In the middle of a life thunderstorm
I said it happened five years ago - well within [sic] those five years, I had lost a baby, my brother had gone through a divorce and he was in a financial mess which I had to sort out for him. My mum and dad got divorced so I had to sort her out. Things came out about what my dad try to do me and my brother when we were younger. It was all those things going on. I also took on a new role in the company I was working with, before they went into liquidation. I was the assistant to the MD, so I saw everything that was going on and then I started up a new business. So there was an awful lot happening...

Testing possibilities in vain
/ seeking to understand
/ efforts to control

Very Leventhal: every hypothesis indicates some response, and this process tests the hypothesis. Testing also establishes the limits of one's own control. Could later distinguish different classes of hypothesis, and see what procedures are attached to each.

44-5 Testing the overwork hypothesis
Well I cut down on working and I got another job and I was doing half the work that I used to do and I was still feeling very tired.

137-8 Am I a fake?
But the problem was because you didn't feel particularly ill you thought it was in your mind so you are thinking well am I trying to get attention or something like that

140-2 It could not be a bad lifestyle
I was a very healthy person - I ate the right things and not many of the wrong things, I don't smoke - well I drink but only socially - and I did a lot of exercise. So I really looked after myself

189- Diet and exercise
I couldn't stop eating any of the bad things because I wasn't eating them. So I just didn't know what to do, I was going to the gym more trying to see if that helped. I did try to do everything I could myself and nothing happened.

Taking on advice about stress

So a lot of times I thought it was that. I said to myself calm down. Also the doctor I went to see about ME said I should cut down about my hours so I gave up everything. I gave up my job, my business and that and just went temping. I got a job that was really nothing compared to what I used to do to see if that would help.

Efforts and sacrifices in vain

Well it was quite nice at first because you didn't have the responsibility and I could come home forget about it. But I was still feeling really really tired. And I said to myself I shouldn't be now I should be better now I should have more energy now. I was still going to the gym and things like that, but nothing changed really.

I just felt so ill and so lethargic and I felt this isn't normal. I mean it can't be an age thing, I was only my 30s. I know you slow down but you don't slow down that much. I just couldn't do anything!

Mental stress (madness) hypothesis

No, well part of it, when they kept saying it was stress, I thought well, is it mental, is there something mental that I have get sorted? I even went to see a psychiatrist to see if that would help- I felt fine after I got some things off my chest but nothing was improving, so I said no it can't be that.

6 Confusion/ Fear/ Alone / fearful uncertainty

<table>
<thead>
<tr>
<th>Page</th>
<th>Sentences</th>
</tr>
</thead>
<tbody>
<tr>
<td>190</td>
<td>So I just didn't know what to do,</td>
</tr>
<tr>
<td>135-6</td>
<td>Well most of the time I felt scared and depressed cos I didn't know what it was.</td>
</tr>
<tr>
<td>136</td>
<td>I was lonely because nobody seemed to be taking any notice of me.</td>
</tr>
<tr>
<td>137-4</td>
<td>But the problem was because you didn't feel particularly ill you thought it was in your mind so you are thinking well am I trying to get attention or something like that. I felt alone and confused.</td>
</tr>
<tr>
<td>150-1</td>
<td>Well I'm not a person that cries but I used to cry a lot when I was on my own and I just didn't know what to do, who to go to</td>
</tr>
<tr>
<td>153-4</td>
<td>I just felt very lonely and not knowing what to do- at the time I didn't know what to do.</td>
</tr>
<tr>
<td>310-3</td>
<td>So I don't know, I just—I felt awful and lonely, I didn't know what it was, I just thought something was wrong. I didn't think it was cancer or anything like that, I just knew that something was wrong.</td>
</tr>
<tr>
<td>461-2</td>
<td>?some confusion and gnawing questions remain .if I hadn't gone through all those traumas after I had the operation, would my pituitary gland be OK, you just don't know.</td>
</tr>
</tbody>
</table>
Appendix J: Qualitative Analysis Level 2 Themes

The following is an example of a 2nd level organisation in the analysis. All 2nd level themes for this interview are listed, along with the 1st level themes that cluster underneath them.
Level 2 Analysis

Wider 2nd level themes are shaded, with 1st level themes listed underneath.

<table>
<thead>
<tr>
<th>Health control strategies fail</th>
<th>Devouring &amp; evaluating information</th>
</tr>
</thead>
<tbody>
<tr>
<td>Diet fails</td>
<td>Know all about it</td>
</tr>
<tr>
<td>Timeline exceeded</td>
<td>Devouring information</td>
</tr>
<tr>
<td>Doctor's advice</td>
<td>Calculating odds of cure</td>
</tr>
<tr>
<td>Negative tests</td>
<td>Demystifying doctors</td>
</tr>
<tr>
<td></td>
<td>Take it with a pinch of salt</td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Visions</td>
<td>Enduring the wait</td>
</tr>
<tr>
<td>Cancer eating me away</td>
<td>Urgency</td>
</tr>
<tr>
<td>At a loss</td>
<td>Part of the process</td>
</tr>
<tr>
<td>Anger/frustration</td>
<td></td>
</tr>
<tr>
<td>Fear</td>
<td>Rollback</td>
</tr>
<tr>
<td>Stuck/hopeless</td>
<td>As if by magic</td>
</tr>
<tr>
<td>Passivity/passive coping</td>
<td>Gone and forgotten/ done and dusted</td>
</tr>
<tr>
<td>Taken over</td>
<td>Knowing vs hoping</td>
</tr>
<tr>
<td>Expecting death</td>
<td>Impact of expectations on family</td>
</tr>
<tr>
<td>Downwards spiral</td>
<td>Wish to forget</td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Identity Intrusion</td>
<td>A longer process</td>
</tr>
<tr>
<td>Identity slipping away – taken over</td>
<td>Accepting</td>
</tr>
<tr>
<td>Threatening identities</td>
<td>Waiting</td>
</tr>
<tr>
<td>Cripple - dependent</td>
<td>Ultimate endpoint</td>
</tr>
<tr>
<td>Madman – locked away</td>
<td></td>
</tr>
<tr>
<td>Loss of valued attributes</td>
<td>Future threat</td>
</tr>
<tr>
<td>Temper – stability</td>
<td>Not worried</td>
</tr>
<tr>
<td>Judgment- acumen</td>
<td>Accepting randomness</td>
</tr>
<tr>
<td>Control-freak</td>
<td>Have action plan</td>
</tr>
<tr>
<td>Paterfamilias</td>
<td>Enduring family impact</td>
</tr>
<tr>
<td>Maintenance of valued attributes</td>
<td>Father role/leadership</td>
</tr>
<tr>
<td>Working</td>
<td>Decision-making</td>
</tr>
<tr>
<td>Quick-witted</td>
<td>No change of outlook</td>
</tr>
<tr>
<td>Challenges to temporal continuity</td>
<td>Still opinionated and selfish</td>
</tr>
<tr>
<td>Memory problems</td>
<td></td>
</tr>
<tr>
<td>Negotiating identity in social domain</td>
<td></td>
</tr>
<tr>
<td>Resisting engulfment</td>
<td>Road to Damascus moment</td>
</tr>
<tr>
<td>Safety in numbers</td>
<td>Relief</td>
</tr>
<tr>
<td></td>
<td>Vindication</td>
</tr>
<tr>
<td></td>
<td>Hope</td>
</tr>
<tr>
<td></td>
<td>Anger (lost time, suffering)</td>
</tr>
<tr>
<td></td>
<td>Coherence</td>
</tr>
<tr>
<td></td>
<td>Allows search to begin</td>
</tr>
</tbody>
</table>

**Interview: Peter**

dismissed by doctor
No there's nothing wrong
Labelled
Invalidated

Family labels
Madness
Old codger
Appendix K: Qualitative Analysis Level 3 Themes

The following is one of the (several) 3rd level organisations of themes. Themes are organised hierarchically under the two broadest themes.
Level 3 Analysis

2nd level themes across all interviews have grouped, considered and organised hierarchically under two broader 3rd level themes. This organisation presented here is one of the preliminary ones, which evolved itself into the organisation that is presented in the Results section.

I. Evolving Understanding

A. Clusters of Understanding
   i. Everyday Causes
      a. Testing by coping
      b. Ignoring
   ii. There is Something There
      a. an existing problem
      b. something evil inside
      c. madness and lies
      d. frustration and confusion
      e. failing faculties
      f. dangerous delays
   iii. Gone and Forgotten
      a. paradoxical relief
      b. light at the end of the tunnel/ a breath of fresh air
      c. it wasn't just me being mad/ I told you so
      d. restoration and undoing
      e. cutting it out
      f. a hopeful illusion
   iv. Cushing's is for Life, Not Just for Christmas
      a. when the box burst open
      b. it's something you live with
      c. chemical vs. surgical
      d. keep on fighting
      e. prepared for the future
   v. Vague Menace
      a. it will come back to haunt me
      b. fragility

B. Multiple Shifting Models

C. Social Framing of Understanding
   i. Shaping and feedback
      a. beliefs of others
      b. marking the illness
      c. isolation
      d. buddies
      e. establishing comparisons
      f. strain
   ii. If Doctors Don't Listen to Me, How Can I Trust Their Advice?
      a. Dismissive Doctors
      b. Unresponsive System
      c. Managing interactions
      d. Trust
II. Transmutation of Self

A. Dissolution
   i. Losing Positive Selves
      a. Losing the in-control self
      b. Losing the active self
      c. Losing the independent self
      d. Losing parenthood
   ii. Gaining Negative Selves

B. Reconfiguring
   i. Affirmation and Protection
      a. It's because of my condition
   ii. Accommodation and Re-evaluation
      a. Positive reframing

C. Social Framing of Identity
   i. Identity-Affirming Contexts
      a. Parenthood
      b. Doctors who listen
   ii. Identity-Damaging Contexts
      a. Blame and shame
      b. An invisible disability
      c. Lost in the eyes of others
      d. Becoming a dependent
      e. Becoming a recluse