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Down's syndrome: cost, quality and value of life.
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Abstract

Routine prenatal screening is based on the assumption that it is reasonable for prospective parents to choose to prevent a life with Down's syndrome. This paper questions whether Down's syndrome necessarily involves the costs, limitations and suffering which are assumed in the prenatal literature, and examines the lack of evidence about the value and quality of life with Down's syndrome. Tensions between the aims of prenatal screening policies to support women's personal choices, prevent distress, and reduce the suffering and costs of disability, versus the inadvertent effects of screening which can undermine these aims, are considered. Strengths and weaknesses in medically and socially based models of research about disability, and their validity and reliability are reviewed. From exploratory qualitative research with 40 adults who have congenital conditions which are tested for prenatally, interviews with five adults with Down's syndrome are reported. Interviewees discuss their relationships, education and employment, leisure interests, hopes, aspects of themselves and of society they would like to change, and their views on prenatal screening. They show how some people with Down's syndrome live creative, rewarding and fairly independent lives, and are not inevitably non-contributing dependents. Like the other 35 interviewees, they illustrate the importance of social supports, and their problems with excluding attitudes and barriers. Much more social research with people who have congenital conditions is required, if prenatal screening policies and counselling are to be evidence based.

Key words

prenatal screening, quality of life, Down's syndrome,
disability, learning difficulty, mental retardation

Introduction

Although Down's syndrome affects relatively few families directly, internationally each year it is discussed with millions of women and couples when they are offered prenatal screening. They need evidence based information, if they are to be able to make informed personal choices, and if these large numbers of people are to have realistic, unprejudiced knowledge about people with Down's syndrome. Prenatal screening policies illustrate tensions, firstly, between clinical support for women's personal freely made choices (Royal College of Physicians, 1989) versus public health programmes designed to reduce disability. Secondly, there are conflicts between intentions to reduce distress versus raising high levels of anxiety (Green and Statham, 1996) and offering, as the only treatment, termination of desired pregnancies (Santalahti, 1998), "when science helps to cause, define and propose solutions to risks" (Beck, 1986:156). Thirdly, screening services intended to promote public health may also undermine health and increase suffering. One clinical geneticist commented, screening "may actually promote the stigmatization and intolerance that is a major cause of the suffering experienced by many affected individuals and their families" (Clarke, 1997:123). Fourthly, efforts to reduce "the costs of life-long care" of people with Down's syndrome through prenatal screening (Wald *et al.*, 1998) may inadvertently increase these costs by propagating attitudes which restrict these people's independence and employment opportunities.

This paper examines these tensions in several ways: by briefly reviewing the background literature which influences screening policies and counselling; by comparing medical and/or social models of disability for their realism and their different views of the origins and nature of the suffering which screening is intended to prevent; and by reporting and discussing an exploratory study of the views of people who have Down's syndrome about prenatal screening, and cost, quality and value of their lives.

The literature relating to screening for Down's syndrome

The medical, nursing, psychological and ethical literature tend to present negative reports of Down's syndrome. It is "not treatable", "the most common form of severe mental retardation" linked to "precocious dementia" (Takashima 1997), associated by mid-adulthood with high levels of (frequently undiagnosed) severe sight and hearing loss, heart and lung disease (often following untreated infections and heart defects), loss of cognitive abilities, epilepsy, serious behaviour problems and "poor communication or confusion due to Alzheimer disease" (van Allen *et al.*, 1999). Some paediatric texts mention Down's syndrome only in reference to prenatal screening (Cade *et al.*, 1995), or emphasise severe pathology as if everyone with Down's is very adversely affected (Taylor, 1995). One expert calculated a net gain to society, because of the assumed severity of Down's syndrome, of screening 100,000 pregnancies, involving 3,000 amniocentesis (2,960 with negative results) incurring the inadvertent miscarriage of 30 unaffected fetuses, in order to reduce the incidence of Down's syndrome from 100 to 60 live births (Painton, 1997). Other experts, however, question such cost-benefit calculations (for example, Fletcher *et al.*, 1995).

The midwifery press tends to be wary of "the prospect of having a child whose prognosis [with Down's syndrome] is deemed very bleak by conventional medical opinion" (Grayson 1996). Psychometric research is valuable in assessing needs and services, but it concentrates on negative issues, such as anxiety, depression, stress and blame (for example, Hall *et al.*, 2000) thus tending to present negative reports about Down's syndrome. The many surveys of raised anxiety among pregnant women during screening implicitly reinforce assumptions that Down's syndrome is something to be very anxious about. One psychological study reported an IQ range from 10 up to 92 among people with Down's syndrome (Lorenz, 1984), and

Noble (1998) found a range up to only 67 but literacy levels of 40 per cent. Today, their educational achievements and expectations are rising considerably (Alderson and Goodey, 1998), showing that prenatal prognoses are complicated by uncertainty about the severity of each case, and the unpredictable effects of future life style.

When it is questioned whether Down's syndrome itself entails suffering, one response is to speak of the relatives' suffering: "women clearly regard a baby with Down's syndrome as an infinitely worse outcome than losing a normal baby from a prenatal diagnostic procedure" (Lilford, 1990). Another response is to redefine suffering "as shorthand for a whole range of disadvantageous conditions. I do not literally mean being in pain or being in discomfort" (Harris, 1990:172). Some philosophers assume that serious intellectual impairment is incompatible with being able to value one's life, or with being a person (Kuhse and Singer, 1985; Harris, 1985). Even theological ethicists also emphasise individuality instead of the moral webs of human relationships (Strathern, 1992). Taken to extremes, the important ethical principles of respect for autonomy and cost-effective justice can be negative rather than neutral towards disabled people, and towards intimate interdependent relationships such as those between mothers and babies (Mendus, 1987).

This is a brief summary of very complex, varied debates, which highlights dominant views that appear to have greatest influence on current screening policies, and which mainly associate cost, suffering and dependence with Down's syndrome. For a comprehensive literature review, see Alderson (forthcoming).

Medical and social models of disability and suffering

How realistic are these predominant data and debates about Down's syndrome? They draw on research mainly conducted within the medical model of disability, which attributes people's suffering chiefly or solely to their diagnosed disorder. In contrast, the social model attributes morbidity mainly to disabling barriers and attitudes which unnecessarily exclude people from mainstream society (Oliver, 1996). Some medical researchers take account of the social model. A study of 280 babies with Down's syndrome attributed their poor health to lack of "necessary care" by parents and health professionals. Denying effective treatment to children because of their shorter life expectancy, thereby contributes to reducing their life span. The authors added that prenatal diagnosis may adversely influence "the social acceptance of Down's syndrome" and their sub-standard health care (Julian-Reynier *et al.*, 1995).

Whereas the medical model attributes each problem - cognitive and behavioural difficulties, sensory loss - to the syndrome (see van Allen *et al.*, 1999 quoted above), medical research which acknowledges the social model links the problems to one another and to social influences. Sensory loss, which follows untreated infections that may be linked to deficiencies in diet and exercise, influences behaviour and confusion which are also greatly affected by relationships and attitudes, as discussed by interviewees who have Down's syndrome later. Clinical and social conditions constantly interact, positively and negatively.

The medical and social models thus differ in their views about the origins of suffering, as either biological/genetic, or arising socially such as through substandard life styles and health care. The models also differ in their views on the nature of suffering. The medical model stresses the pain and misery of physical and intellectual impairments for affected people and for their families who share some of their suffering and restrictions. The social model is less concerned with bodily limitations than with the emotional pain, loneliness and unfulfillment which follow the unjust prejudices, discriminations, barriers and exclusions that unnecessarily disable impaired people. Disability rights authors argue that disabled people can live challenging and fulfilling lives when they have adequate support (Bailey, 1996;

Oliver, 1996; Asch, 1999). Prenatal medical prevention is the logical solution to non-treatable genetic/biological causes of suffering, whereas social/emotional suffering is resolved and prevented by changes in social attitudes and structures towards making societies more inclusive, reforms which are undermined by national prenatal screening programmes.

Shakespeare (1999) questions whether the social model of disability is too dismissive of bodily problems, just as the medical model too readily ignores social conditions, and he calls for more research with disabled people about their diverse experiences. These points are perhaps still more relevant to people with learning difficulties, whose views are so rarely researched and who have no thinking aid that is equivalent to the mobility aids which liberate physically disabled people.

Research methods in medical and social models of disability

Qualitative and quantitative methods overlap, and are useful in both medical and social models. Yet as this section reviews, they can reflect and reinforce the medical or the social model of disability. Psycho-medical assessments of quality of life tend to use questionnaires with closed questions administered in a rather impersonal standard manner, which can confuse some people and talk down to others. High numbers of respondents count for more than spending time on gaining detailed responses from fewer people. When they are discouraged from speaking beyond the main point, people may nervously try to give brief “correct” answers, and then seem dull and lacking in individuality. Questions like “How does your problem affect your life?” can exaggerate the effect of the syndrome by excluding other strong influences like income or transport services. Terms such as “patient, disease, suffering” turn “Down’s” into an illness. Disabled people who are used to being questioned in this way tend to be cautious and distanced, warn Gillman *et al.* (1997), who analyse how these approaches objectify people, measuring them against assumed norms, and contributing to negative records which silence and oppress them. Problems are recorded far more than successes and abilities. “Challenging behaviour” may mean not conforming to rigid routines, or even not being passive (Gillman *et al.*, 1997).

The contrasting social research methods used for the exploratory research are described below, as used before, during and after the interviews, followed by a report of the main findings and then a discussion about the validity, reliability and policy implications of the study.

Before the interviews

The project was approved by the Institute of Education ethics committee. Forty adults who had a condition which is tested for prenatally were contacted, mainly in south-east England; five had Down’s syndrome. The other conditions were cystic fibrosis, thalassaemia, sickle cell and spina bifida. Information leaflets were sent through informal networks and self-help organisations with reply envelopes asking people to opt in to the research. Contacts were not made through health or social services, in order to emphasise confidentiality, and to encourage a person-to-person approach rather than a practitioner-patient/client one between interviewer and interviewee. The informal contacts and respectful opt in methods made it harder to contact interviewees, and people with Down’s syndrome were the hardest to find, perhaps partly because carers assumed that the interviews would be too complicated or distressing for them, and because many people with Down’s live secluded lives. To enable everyone to give informed consent or refusal, the leaflet explained that the interview topics were:

about your views about your daily life, your family and friends, your school or

college, your work - or being out of work. We would like to know about your aims and hopes, the things you enjoy doing, and parts of your life you might like to change. The research also asks what you think about the new genetics, gene tests and antenatal screening. If you don't know much about genetics and screening, we still hope that you will talk to us. We are not looking for expert answers, it is your own views that would help our research.

These topic themes, and not set questions, were the basis of the interviews, and also covered what interviewees might like to change about themselves (if anything) and about society, and their views on being or becoming a partner and a parent, which they often introduced when talking earlier about their hopes. If they seemed willing, they were asked about screening for Down's syndrome. The leaflet continued:

“What is the research for?”

1. To report a wide range of views from around Europe;
2. To increase informed public debate about screening;
3. To make sure that the views of people who are most directly concerned are more widely known;
4. To inform those who plan and provide health services around Europe about the kinds of services people want to have.

What does helping with the research involve?

We invite you to take part in an interview or a small group session. We would meet for about an hour, at your home or somewhere else that suits you. If you agree, we would tape-record the session. We will 'phone each person one week after their interview, to see if there is anything more they wish to talk about. We will send a short report about the research later this year to everyone who helps us.

Research and your rights

- * It is for you to decide if you want to talk to us. You do not have to say 'yes'.
- * If you do say 'yes', you do not have to do the whole interview.
- * We could stop when you want to, or have a break.
- * If you do not want to answer some of the questions you can just say 'pass'.
- * Before you decide if you will help us, you might like to talk about this project with your parents or with a friend.
- * We will keep tapes and notes of the interviews in a safe, locked place.
- * When we talk about the research and write reports, we always change people's names, to keep their views anonymous.
- * We would not talk to anyone you know about what you have said, unless you talk about the risk of someone being harmed. If so, we would talk with you first about what could be done to help.”

There were details about the sponsor and the researchers and how to contact us. Page 4 added:

“Lots of research is done with people with Down's syndrome.

What is new about this project?

1. Most research about people with Down's sees them as patients and is about their health treatment. This research is about you as a person and about your own views on your life.

2. We want to do this research as partners with people who have Down's.
3. A lot is known about doctors' and nurses' views about screening and life with a disability. Little research has been done about the views of the people they want to help.
4. The planners need to know more about the views of the people they want to help, if they are to plan useful health and screening services.”

I interviewed three people, a colleague interviewed John who wrote to us in response to our article in a self-help journal asking for interviewees, and a speech therapist interviewed Colin, whose mother contacted us through the journal and who lived a long distance away. All the other groups in the study were easier to contact and were each interviewed by one researcher.

During the interviews

With their consent, four of the people with Down's syndrome were tape-recorded and notes were made of the other interview. Three people were interviewed at home and two at their college, from between 30 to 150 minutes. To encourage them to talk freely, and to share control over the topics, pace and style, open questions were used, such as: “Will you tell me about the schools you went to?” Prompting questions elicited varied individual narratives in an informal conversational style. We hoped that they felt confident that their views were respected and not judged for their correctness. We aimed to establish mutual trust and rapport (Oakley, 1981; Smith, 1994; Booth and Booth, 1996; Ward, 1996; Rodgers, 1999), and to move from the formal to the “private” voice when people express more personal views (Cornwell, 1984). Some people with Down's syndrome speak slowly and rather indistinctly, so that the whole pace of the interview is slower; I repeated some words to help with later transcribing of the tape recording, hoping that this sounded respectful.

I felt anxious about asking people, in effect: “What are your views about the value and quality of your life? Do you feel your life is worth living? Did/do your parents want you and what might they have decided if prenatal tests had been available to them before you were born?” These sensitive questions were not asked directly, although several interviewees, including one with Down's syndrome, raised the later ones. Most of the 40 interviewees talked openly about screening, but those with Down's syndrome and spina bifida were most likely to express or imply distress about this topic, so that direct questions were more limited with them. Instead, through enquiring about their everyday and past lives, and their hopes and concerns, we aimed to create a picture with each person of their positive and negative experiences, of the value and quality of their life to them, and of potential lives with their condition.

After the interviews

When I phoned a week later, everyone said they felt all right about the interview and had nothing to add. I sent transcripts and a short end of project report, asking if they wished to have details of published reports. No one with Down's syndrome replied. I met two of them later at a performance of their play. I analysed transcripts by hand, and reread them for the overt themes, reported here in the order they were discussed during interviews, and for underlying themes: quality and value of life, costs, dependence, contributions to family and society, discrimination, replies which fitted mainly the medical or social model of disability.

Interviews about living with Down's syndrome

John and Colin both aged 20 lived with their parents. Martha aged 35 lived mainly on her own in her large flat where her boy friend sometimes stayed, Peter aged 43 and Philip aged 40 lived in a house with two women with learning difficulties which their parents bought for all of them 14 years previously. They were very conscious of being taped, and of making sense of interactions. For example, when Peter coughed he looked concerned about the noise this would make on the tape, and later Philip gently corrected my mistake.

Peter: We do drama now you see, that's why, well Id, that's a short title.

Interviewer: That's the one you're doing now, It?

Peter: Yes.

Philip [politely spells] I. D.

Int: Oh, id, like in Latin?

Peter: That's right.

The three older ones cooked and did their own housekeeping. Martha also cooked for her boyfriend, who has diabetes; she laughed when she said his diet helped to keep her weight down. Martha had a notice "please take off your shoes" outside her flat, "because why should I clean up after people? I've got better things to do." Like Peter and Philip, she checked the spy hole before opening the front door.

Education and employment

They were all literate and numerate. Colin was pleased to "be the first one" who attended a special learning unit at his local mainstream school and Martha's mother "fought" for her to be able to leave special school and attend mainstream school, where she was very happy until the head teacher she liked left.

Int: So your mum had a row?

Martha: It wasn't a row, it was a fight. She had to fight for my rights you see. [The new head teacher had said] AWhat is this mongol person doing in my school?@ I was there for five years, *five years*, and she said that!

Int: So what did your mum say?

Martha: Well, what *did* she say to her? My mum went to the high court straight away. Of course. The high court of justice! [The fight was to be able to stay in a mainstream school] Because the special school didn't do me any good *at all* basically.

Martha had GCSEs (school leaving exams) in French, drama and housecraft. In inclusive mainstream schools it is usual for pupils with Down's syndrome to take GCSE courses, and some are bilingual when their families are (Alderson and Goodey, 1998). With his brother who was at university, John used "to run a local youth club, we had to do the tidying up, 50p to get in. We used to play games, pool, snooker, and we played music on grandad's radio. They didn't have music, so we took the radio over to the youth club." John was at further education college.

This year I'll be on NVQ level. I've taken six computer exams....I want to go into management. I'm on work placement two days a week, folding leaflets, typing, photocopying, sending post. I am trying to broaden out a bit, into the computer programme, computer analyst stage. I'll look in the local paper and the high street usually, to see what [jobs] they've got.

Colin worked two full days a week in a hospital kitchen, and went to a learning support centre at college on three days. His courses included horse management, pottery, communication, computing, geography, motor power, health and safety and catering. Martha also went to further education college travelling across London by underground by herself; she learned office skills.

Then they said, “okay, you can do the community care course,” and I thought “what are you talking about? I don’t want to do that. You should ask me what I want, not what you want,” so I looked down the page and I saw “food” and I thought “Yes, food! Yes I’m going to do that one.” So in the end I did catering for two years.

Then she worked in an office for 13 years, and later taught at workshops for people with learning difficulties on independent living and safe sex. Like Peter and Philip, she taught health professionals about learning difficulty and empowerment. Martha has lectured abroad, was a free lance artist and wanted to be a college art teacher. She wished she could still find paid work.

Peter and Philip arrived late back from work looking tired and ate their supper which their house-sharers had cooked. Then they began explaining their theatre work; for some months every year for the past seventeen years they had worked as actors. The cast, who all have learning difficulties, talk about ideas with two professional directors who then write the script which the cast memorise using typed copies and tape recordings. The plays about disability are presented on television and in provincial and London theatres. *A change of heart* was about a girl who was refused a heart transplant because she has Down’s syndrome, and *Breaking the mould* was about genes. In their current play, *Mongol Boy*, a Victorian industrialist rejected his son who had Down’s syndrome and who joined a circus of freaks. Peter and Philip distinguished between the denigratory language in the play and their own views, raising a main theme of all 40 interviews: discrimination against disabled people.

Int: So do you like that kind of drama, that you’re really involved in?

Peter: Oh yes, it’s amazing.

Philip: It is very important, and when you learn to say the part you can. But it’s not very nice to say this, and I’m not a very rude person, but I think that a learning disability part - I don’t want to be rude -

Int: You seem to me to be a very polite person.

Philip: I don’t like to take the mickey of people because I know they’ve got rights, they’re a human being like us you see, but they are “freaks” very small people and it’s wrong to take the mickey out of themand make them upset because they’ve got rights as well.

Int: Yes, very important.

Philip: It is important.

Int: Yes, and do you think - with people with learning difficulties - that other people think enough about your rights?

Philip: Some people don’t understand. They want us to keep quiet. Because they think, you’ve got a learning disability - like when I came out [of the house], somebody pushed me, just out here, pushed me and didn’t apologise and I didn’t like that at all.

Int: Did they push you by accident or on purpose?

Philip: I think they did it on purpose. They’re very strange people.

Peter: And that happened at [names another place]-

Philip: Yes, I know Peter, and it happened here too, a man and a woman tried to push me.

Int: And what did you do?

Philip: Well, the point is this, we are not allowed to talk to strange people.

I asked what they did, apart from the annual play, and their expressions changed from intent enthusiasm to resignation. They talked about having grown out of the day centre.

Philip: [quietly as if he doesn't want to be critical] I don't like the centre myself, but I say stop.

Int: Stop going to the centre?

Philip: No, I'm saying stop, it's too personal.

Int: Oh you don't want to talk about it?

Philip: No, no. Peter as well.

Peter: No is the answer

Int: Right, thank you And is there anything you'd like to change about your lives? Make anything different?

Philip: Well I would like to change....

Philip spoke of his ambition to share in creating and directing a play about the twenty year history of one of their theatre companies, and to take it on an international tour, partly to raise awareness about the abilities of people with learning difficulties.

Changing society

Colin who also attended a special centre was depressed about it, and implied that his problems, at least partly, arise through interactions and other people's attitudes. He missed his friends.

Colin: I sometimes feel a bit lonely at home especially here at the centre sometimes there's no one to talk to.

Int: No one who thinks like you do?

Colin: No not really. I don't have a life here really with no one to see, you know. I used to have a girl friend once, at one time, but unfortunately that didn't work out properly.

Int: How about the people you work with? What do you think of them?

Colin: Well fairly good [laughs rather sadly].

Int: How do they treat you?

Colin: I can't really say, I'm afraid.

Int: Because you can't remember, or you don't want to say?

Colin: I don't want to say. Some are quite difficult.

Int: How do they make you feel?

Colin: Well a bit sad you know, a bit happy, a bit of both, a bit miserable sometimes.

Int: Do they ever get angry?

Colin: Other way round.

Int: You get angry?

Colin: I get - it's them making me, it's the other way round.

Int: Oh, they make you angry?

Colin: Yes, sometimes.

Int: Oh, how do they manage to do that?

Colin: I really don't know, I, I don't understand that myself, I don't know I'm afraid.

On changes in society, Colin said that he did not watch the “boring” news, and when asked “what if you could change the world, what would you do, change anything at all?” he replied “me”. He said he wanted to be “a married man I think I would say, and a snooker player. Yes I wish I was, I’m not now, maybe I will be soon, with luck.” John said “I don’t think I’d like to change a thing about myself”, and later added, “I’ve got no weaknesses, I’ve got certain strengths.” “In the world outside” John was concerned about “ the environment...and pollution and oil, and car exhausts....and pollution of rivers....and wild life really.”

Int: What about the way society is run?

John: Put it this way, the ones that are controlling the world are MPs and they’re trying to do their job. They’ve got a picture, they need to focus, they can’t get a perfect picture, but a better picture, and take some action. They’ll probably face new problems, like council tax, new problems, and look towards a better world, rather than sticking to the old one.

Int: Are there some people you don’t like?

John: [thinks hard] I don’t think so, no.

Int: Are there some people who don’t like you?

John: In the first year they tend to boss you around and tell you what you’re better off doing, and it goes from there - till *they* feel stupid rather than you. You’ve got to get used to it, their attitudes, they change from time to time, they started by bossing me around and then they realised they shouldn’t and they stopped and *they* felt stupid. The message comes back to them....You’ve got to speak up for yourself. [Referring to Princess Diana’s recent funeral] Earl Spencer was speaking out for himself, he had to make people understand him and how he felt. The newspapers and paparazzi, they didn’t know what else was going to happen. They didn’t realise that the message is coming back to them. Like twisting someone’s arm, they end up twisting their own arm.

Int: What do you find most difficult?

John: [smiles] Put it this way, I’ve got no weaknesses. Hopefully next year I might get a job, it all depends what standard they come up with.

Martha, who replied to questions rapidly, paused when asked what she might like to change in society.

Martha: Well when my mum took me away from special school and the local authority came round and said “why isn’t she at special school?” my mum said, “not over my dead body, sorry” [laughs].

Int: So you’d like all that changed - education?

Martha: Yes.

She added that better employment opportunities were needed for people with learning difficulties.

Enjoyments and hopes

They all talked about things they enjoyed doing, being with friends and relatives, watching television, music, playing snooker or going out for a meal, doing art and drama. Asked about hopes and aims, Colin wanted to be a world champion snooker player. His immediate aim was to live in a group home but in this he seemed to feel helplessly reliant on his social worker. He was very anxious because his mother

planned to move away from the area and he wanted to stay but, like many people his age, could not afford to rent anywhere. Martha wanted to discuss her work with a famous French artist. She enjoyed yoga and being with her respite carer who was also an artist. Martha's address book of friends was crowded with names. John smiled when he was asked if he would like to get married.

John: Having a girl friend, which doesn't mean you have to get married. I've been to a genetics professor....But the main question is, will I have any children? The answer was yes, but she talked to me about getting married and that leads to responsibilities and coming to terms with the risks my children will take.

Int: What risks?

John: Electrical things can be dangerous, electrical appliances, plugs, radios, kettles.

Int: Would you like to have children?

John: That all depends on picking someone, no, that's not the right word, picking, but if the perfect girl comes up. I don't know if she will. Children - it all depends on her career and my career. The important things come first. We've got to discuss these matters at the time.

Colin was asked if he had any hopes.

Colin: If I had a lot of money I would stay here [in this town]. I don't know really, I might get married even one day.

Int: Would you hope to have children?

Colin: I haven't thought about that yet, I would have children but it is too early to say, a bit early for me, anyway.

He talked of a former friend who was now too busy with wife and children to see Colin. Martha thought she might get married one day but "it's nice having independence too, I like that. Eventually, you never know, in some years' time...and it takes a long time to arrange a marriage, not getting married, but doing things before you get married [laughs] yes." Martha said it was harder for people with learning difficulties to behave normally because of their circumstances "they don't get a chance", and she mentioned how two people behaved at her party.

Martha: It's all very well, and there I am talking about safe sex and that up and down the country, well I mean....normal people wouldn't do that because they're normal people, but people with learning difficulties they don't get a chance.

Int: When you say normal people, do you think of yourself as quite a normal person in many ways?

Martha: Yes I am I think, well my dad said to me he was so proud of me being on the television he said you're not Down's syndrome, you're up syndrome [laughs]. [Martha enjoys travelling but] The trouble is getting the money together, we're on benefits, you can't afford it on benefits.

Int: No, but you do manage to live quite a - good life?

Martha: I do, yes.

Int: You have to be very good at housekeeping?

Martha: Yes, I do manage my own money, that's fine but if it's big money my sister comes in and sorts out for me.

When asked what they might like to change in themselves or their life, if anything,

Martha said she was happy as she was, and Colin replied, "I'm trying to be independent." Peter and Philip said they did not have much time for the interview, as they had a lot of lines to learn, so they omitted some topics and moved to the final theme of screening

Prenatal screening

Martha spoke about the chances of her children having Down's or diabetes.

Int: If you were going to have a baby and they said this baby's going to have Down's syndrome, what would you do?

Martha: I know what to do, you know, it's quite easy [laughs] everything, you know, the baby, it's a human being, feed it, look after, give it lots of love, everything.

Int: And can I just ask you about testing and screening? Do you think it is a good idea or does it make more problems?

Martha: It makes more problems really I think, because [describes problems her mother had with previous pregnancies] I'm not really sure what really happened, I wasn't around at that time [laughs]. I do know when I came I was a big shine to my mother, when I was born. She was pleased that she had me, and yes it was a pleasure for her, for me too, and people need to see that, like in [she described children with Down's syndrome in soap operas]. They should show more like that.

Int: If you talked to counsellors who talk to women who have had the test and the baby has Down's what would you tell them to say?

Martha: [pause] Well you see, there wasn't a test when I was born [pause]. Well if they want to think about having it, it's not my problem, but I really think they shouldn't have it [the test].

[We talk about how counsellors may not know anyone with Down's, and Martha recalls working in an office with a telephone help line about prenatal testing.]

Martha: I was the only one in the office with Down's syndrome, all the rest are normal, right? Well it's all very well someone who's normal talking about it. I could have been on that telephone and I know the history of Down's syndrome and I could easily say to anyone who might have a Down's syndrome baby, "Look at me then! Ha ha!" [Said triumphantly.]

Int: Do you think your mother fighting for you was so helpful, and saying, "Let's go and do this and try that"?

Martha: Yes, exactly.

Int: Where did she get her wonderful ideas from?

Martha: Well she just had it, and when she died it was like a bomb dropping on us, it was very sudden, it's been 8 years, and after that I had bereavement counselling....

Int: Did you want to say anything else about screening and what Down's syndrome is like?

Martha: Er, it's 27 cells we have I think, I can't remember. We have one extra cell, so that's a good thing, and we also tend to be quite floppy, the babies have that, but they don't scream and cry much. [They showed Down's syndrome dolls on television] they're all right to play....but I don't think the dolls are really like us because they're plastic and we're real, we're alive, we're very very alive I might say.

Int: Do you know anyone with Down's who does as much as you do?

Martha: No I don't know anyone who does as much as I do. I think it's quite sad that most people don't really know what to do [or] yes, they would have done a lot more, it's a shame [and she describes friends who she thinks are over-protected and thereby restricted.]

John was similarly accepting of babies with Down's syndrome. He described his conversations with his mother about birthing difficulties, then said: "You've got to look for signs if he or she is a Down's syndrome baby. They gave me a reflexology tap across the knee" and he goes on to describe his exercises for his dislocated knee. When asked what he might say to someone who was expecting a baby with Down's syndrome, John said:

John: It all depends how they feel. Let's say me, for example, if I were to discuss abortion, I would. But you've got to contact someone who *wants* a baby. I would discuss it with my "wife" as you put it [he had said he would not necessarily get married] and see how she feels. What I'm trying to say is, you could discuss fostering and they [prospective foster parents] would probably discuss each statement and further and further in each statement.

Int: Does it make you feel bad that someone might not want a baby with Down's?

John: I wouldn't say that entirely. Say that someone doesn't *want* -they're going to get further advice, housing advice, council advice to actually get things going again before they take any action. [He described someone in a soap opera who was going to have her baby adopted then changed her mind.] The mother can't say that the baby has Down's syndrome, it's up to the doctor or nurse. She'd probably think, "Oh, what should I do?" And she should ask her mum and dad for help and advice and support, because of the risks.

Int: Do you think doctors should be testing for Down's syndrome?

John: That all depends on how he or she feels, and what they have to say, before they take any other action.

John's views are more like those of the interviewees with sickle cell, cystic fibrosis and thalassaemia, who tended to say that they respected people's rights to information and choice. Colin was uncertain about what he would say to a woman told that her baby has Down's syndrome, he began by saying, "I have that, unfortunately." Then he said "I don't know," he sighed and after a long pause said, "I haven't a clue."

Int: If your wife knew she was expecting a baby with Down's, how would you feel?

Colin: [Sighs and pauses] Good, I think, [he laughs uncertainly].

Int: Would it be good?

Colin: Mmm, I'm not quite sure.

Int: Whatever you think is fine.

Colin: I don't know anything really.

Peter and Philip eagerly described their play *Mongol boy*, and then were asked what they might say to a woman expecting a baby with Down's syndrome. Their expressions suddenly changed and they looked very sad.

Peter: That is actually what we are doing in this play, for heaven=s sake. [A pause] I=m speechless. The productions we have done are just fantastic. [They look as if their reply is too profound to put into words.] Carry on [to the next question]. Pass.

Int: Yes. You just think it is a completely out-of-order idea? It's not worth talking about, it's so dreadful?

Peter That's, that is what I was thinking of.

Int: Yes, thank you. I am sorry to ask you about these things. The reason is, people around the world are asking doctors, nurses, parents, experts, but they don't ask people with Down's syndrome, and surely you're the people who really know what you're talking about, that your lives are interesting and really worth living and good lives.

Peter and Philip: Yes.

Int: Yes, it's very good of you to talk to me. Was there anything else you wanted to say?

Philip: Well it's just about some of these things, they are too personal, I don't want to talk about them to you or anyone.

Discussion

As an exception, despite Peter saying "pass", I continued a little with the theme, to avoid leaving their response simply silent and unrecorded, or misinterpreted. It felt strange to visit a house where four adults had lived amicably together for so long, and receive these deep responses, as if they could not express their concern in words, because words are inadequate, too definite, finite, compared with their sighs and looks of great sadness and sense of overwhelming enormity. They had wrestled with these ideas for years in their dramas, and Martha had in her teaching, as well as in their daily relationships. Colin seemed troubled by these questions, as if he needed someone with their insights to talk to about his mixed feelings on whether having a child with Down's was "good". John's involvement with his family and friends in enjoyable, responsible activities seem to leave him less worried about discrimination. Possibly his confidence and good humour helped him to cope until "they" realised that they were being stupid, whereas Colin seemed to have no one to help him with his anger about similar treatment.

Through specific examples (being pushed in the street, or excluded from mainstream school) they described the frustrations, pains and restrictions of prejudice. The 40 interviewees tended to attribute problems to negative attitudes and social barriers rather than to their congenital condition, and most were frustrated at not having the opportunities, employment, income and social acceptance to enable them to live their lives as fully as they thought they could. The greatest asset for the five people with Down's syndrome appeared to be family and friends, including artists and actors, who helped them to join mainstream education, work, housing and friendship networks. Deliberately or not, the interviewees' views appeared to adhere to social rather than medical models of disability, informed by their experiences.

Generalisability and validity of research about Down's syndrome

It could be argued that people with learning difficulties cannot know their own limitations, and everyone is liable to blame circumstances or other people rather than their own failings. Yet the marked contrast between Peter's and Philip's drama skills, which might easily never have been realised, and their boredom at the day centre, suggests that the potential of people with learning difficulties will not be realised until they have many more opportunities to develop it. Research which enquires beyond morbidity into people's potential and achievements, and the social influences which support or constrain them, is required before

their ability can be assessed realistically. Such research moves on from notions of fixed (dis)ability, static syndromes, expert researchers and inadequate subjects, to acknowledge how the research questions, methods, interactions during interviews, and hidden assumptions about medical or social models of disability, all shape the data.

This small study may involve exceptionally able people, who present unrealistic impressions of Down's syndrome which could dangerously mislead prenatal decisions and policies. The five people were articulate, although research through interactions and observations with people who do not speak about their quality of life can be very informative (Smith, 1993; Booth and Booth, 1996; Alderson and Goodey, 1998). Until far more research is conducted which takes proper account of social contexts, generalisations cannot be formed about the costs of unavoidable dependence, or the value and quality which people with Down's syndrome experience in the everyday details of their lives.

Qualitative studies and associated literature reviews cannot provide such generalisations. They can, however, contribute the following. Reviews of the history, research, policy and ethics of screening programmes question whether Down's syndrome began to be screened because this was technically possible rather than because it was the most serious condition. The reviews reveal the lack of evidence on which fears about Down's syndrome are constructed, and the underlying negative rather than neutral, intellectually elitist assumptions, which bias the relevant literature and research. Very small samples of interviewees can include the exceptions which challenge established assumptions (Secker *et al.*, 1995). They can show the need for broader, more realistic evidence about the range of ability among people with Down's syndrome, and about links between intellectual ability, contentment and self-esteem (the general population shows that there are no clear correlations and that life style may be more salient than intelligence to quality of life). The interviewees highlight tensions between the aims of screening to reduce anxiety, disability, suffering and cost, and the outcomes of inadvertently increasing these, thereby illustrating the need to reassess cost-benefit calculations in screening policies, and to base them on adequate evidence. This reassessment is urgently needed as the numbers of people with Down's syndrome who can contribute to these debates fall through prenatal selection (Sawtell, 1996). Qualitative studies also provide social models of interactive research methods which are useful in quantitative research.

Qualitative interviews and observations are valid, meaning close to the reality they examine, when they elicit authentic insights into people's lives and explore their own meanings, perceptions, feelings and detailed experiences, as well as possible differences in interpretation between the interviewee and researcher (Crabtree and Miller, 1991). The researcher tries to "step into the mind of another person, to see and experience the world as they do themselves" (McCracken, 1988), which can raise unexpected new themes (Britten, 1995), and explore the ambiguities that closed survey questions cannot do. In reports of small studies, there is space for interviewees' "active voice [to be] heard in the account" (Acker *et al.*, 1983) in detail; they appear as people not numbers, and thus enable readers to assess original data.

Critics of research interviews say that interviewees do not give accounts of external reality but simply present themselves as competent, moral members of particular communities, such as being adequate research interviewees (Murphy *et al.* 1998: 120). People with Down's syndrome who achieve this, powerfully demonstrate their social competence, thus demonstrating an ability which cannot be faked to reflect on the value of their lives. Everyone's accounts and moods are partial and changing, influenced by contingent truths, expressed with different emphases according to different contexts, and researchers have to take account of this, while accepting that some lasting realities are discussed too. The validity

of research also depends on valid theories or models through which the data are understood, on how competently data are collected, reported and analysed, and how nearly the report accords with the differing experiences of the interviewees, standards which this project strives to meet.

There has been too little research with people with Down's syndrome to know how unusual these five people may be, or what a typical person might be like. Down's syndrome is not a fixed clinical fetal diagnosis with a clear prognosis, when it is impossible to assess how severely a fetus might be affected or how life style might affect future capacities. Recent changes in attitudes and education, and the growth of movements run for and by people with learning difficulties like People First, are raising expectations and opportunities. These enable people with Down's syndrome to achieve far more than was previously supposed possible. The five interviews are reported in order to stimulate discussion and further research which will help screening policies and counselling to be based on more realistic and wide ranging evidence.

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