SHORT REPORT

Parental attitudes: antenatal diagnosis of cystic fibrosis

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In order to understand attitudes to antenatal diagnosis of cystic fibrosis (CF), we interviewed parents from 19 families, who already had one child with CF. Nine women had chorion villus sampling in a subsequent pregnancy and 6/19 said they would consider termination of pregnancy if the result confirmed CF. These results differ from the results of antenatal screening studies of previously unaffected families, where most couples opt for termination.

Cystic fibrosis (CF) is the most common autosomal recessive disorder among whites (carrier prevalence 1/25). Carriers can be detected using DNA analysis on mouthwash samples, thus enabling high risk pregnancies to be tested using chorion villus sampling (CVS). CVS can also be offered to families who already have one child with CF. However, the interest shown in antenatal diagnosis by parents seems to vary between countries,^{1–3} and in some instances has been shown to be very low.^{1–4} There are also differences in the attitudes of parents who already have a child with CF, compared to those who do not have first hand experience of CF.^{2–5} This issue is important in view of the burden of care, morbidity of the condition, and the understandable distress of parents when given the diagnosis of CF in their child. No recent study of this issue has been carried out in the UK.

Our study aimed to:

- (1) Evaluate the usefulness of antenatal screening by documenting reproductive decisions.
- (2) Evaluate parents' understanding of CF, to determine whether their reproductive decisions were truly informed.
- (3) Assess whether all parents, who wished it, had received genetic counselling.

SUBJECTS AND METHODS

We examined the attitudes of 19 families (they already had one child with CF and a further pregnancy since 1996) at the Children and Young Person's CF centre in Nottingham (UK). A brief, investigator administered questionnaire comprising open and closed questions (see table 1) was used. The study period was 1999–2000.

Enrolment was by letter, followed up by a telephone call, to the 27 families in the initial sample. Interviews were arranged with those who agreed to participate (n = 19). This included two families, both of whom had in vitro fertilisation (IVF) (using screened donor sperm) for a second pregnancy (having had one child with CF), and one family in which one parent had CF and was embarking on a first pregnancy. Although not strictly fitting the criteria, it was felt that these families' experience of CF would be useful.

Thematic analysis of qualitative questionnaire responses was undertaken by the investigators independently, noting

recurring words, phrases, and themes. SPSS for Windows version 8 was used to analyse the data.

Consent was confirmed in writing and approval was obtained from the local research ethics committee.

RESULTS

Table 1 presents full responses. "Strongly agree" and "agree" responses were combined, as were "strongly disagree" and "disagree".

Nine families had chorion villus sampling (CVS), six indicating they would have a termination of pregnancy (TOP) if the test was positive. Five wished to know whether the baby was affected to give them time to prepare, and two wanted to know if the baby was a carrier. Of the 10 families who opted not to have CVS, two had IVF with screened donor sperm, and seven did not agree with termination, feeling that it would devalue the life of their child with CF. Six said the risk of miscarriage discouraged them from having CVS. Five of these six also felt that termination would devalue the life of their child with CF and would not have a TOP.

Of the eight who refused CVS (excluding the two IVF participants), seven correctly estimated the probability of having another child with CF.

Fourteen couples remembered being offered genetic counselling after the birth of their first child, one could not remember, and four said they had not been offered counselling. Of the 14 who remembered the offer of counselling, five refused this facility, feeling it was not relevant (n = 4) or too much to take in at the time (n = 1). Fifteen families did not discuss the second pregnancy with the CF team, giving a variety of reasons (see table 2) such as: already decided to have CVS (n = 3), felt they knew the options (n = 3), felt they might be dissuaded from continuing the pregnancy (n = 1), and concerned how the team would view a TOP (n = 1). Eight families felt the genetics team approved of TOP. The thematic analysis showed the wide range of feelings held by these families (table 2).

DISCUSSION

Although this is a small sample, we obtained detailed qualitative information. The views of the eight families that refused consent could not be explored, a problem inherent in qualitative research.

Of particular interest is the contrast of attitudes when diagnosis is detected for the first time in an antenatal screening situation, where in one large study⁵ only 24% of parents did not wish to be screened, and all fetuses diagnosed with CF were terminated.

The results do seem to vary in this study from those found elsewhere.^{1–4} However, this may be a result of the fact that some of these studies were done some time ago and in different countries.

Abbreviations: CF, cystic fibrosis; CVS, chorion villus sampling; IVF, in vitro fertilisation; TOP, termination of pregnancy

Table 1 Questionnaire with results

1.	How old is your child?	Age range 7 months – 7 years		
2.	How well do you feel your child is at present?	Not very well		0
		Quite well		6
		Very well		10
	Not applicable, as first child died			3
3.	Were you offered genetic counselling following the birth of your child?	No		4
		Yes		14
	If NO, go to question 5	Could not remember		1
4.	If you were, did you take this offer?	No		5
		Yes		9
	If NO, why (see text)			
5.	Did you seek counselling prior to the next pregnancy?	No		12
		Yes		6
		No response		1
6.	Did you seek counselling immediately after becoming pregnant (after the birth of your child with CF)?	No		7
		Yes		12
7.	(a) On the subsequent pregnancy, were you offered a test to see if your unborn baby would have CF?	No		4
		Yes		15
	(b) Did you discuss your pregnancy with any member of the CF team?	No		15
		Yes		4
	If YES, did this discussion help you in your decision?	No		3
		Yes		1
	If NO, why not (see table 2)			
8.	(a) If you were offered a test did you take up this facility?	No		10
		Yes		9
	If YES, go to question 10			
	If NO, go to question 9			
9.	If you did not choose to see if the subsequent pregnancy (after the birth of your child with CF) was affected, please fill in the following section:			
	(a) You did not want to know if the baby would be affected by CF?	Agree	No opinion	Disagree
		4	1	3
	(b) The risk of miscarriage of the test discouraged you?	6	1	1
	(c) It is unlikely (less than 1 in 4) that you would have a second child with cystic fibrosis because you already have one with cystic fibrosis	1	0	7
	(d) There might be a cure for cystic fibrosis soon	3	0	5
	(e) Cystic fibrosis is not such a serious disease	1	0	7
	(f) You don't agree with terminations	6	0	2
	(g) You don't agree with terminations due to religious reasons	0	1	7
	(h) You don't agree with terminations in this type of situation	7	0	1
	(i) If you had a termination, it would seem as if you were devaluing the life of your child with cystic fibrosis	7	0	1
	(j) Other reason not mentioned above	1	0	7
	(k) Would you make the same decision again?	6	0	2
	Responses to the above were not applicable for the 2 families who had IVF			
10.	(a) Were you offered the option of a termination of pregnancy if the test was "positive" for CF?	No		1
		Yes		7
		No response		1
	(b) Did you think the genetics team approved of terminating the pregnancy?	Yes		8
		No response		1
	Having taken up the option of having a test, was this because			
	(c) You wanted a termination if test was positive?	Agree	No opinion	Disagree
		6	0	3
	(d) Wanted to know if unborn baby would have CF to give you time to prepare?	5	0	4
	(e) Wanted to know if the baby would be a CF carrier?	2	1	6
	(f) Did not want to risk having another child with CF?	6	0	3
	(g) Would you make the same decision again?	9	0	0
11.	Any other comments (see table 2)			

Conclusion

Although only 6/19 couples, who already had a child with CF, said they would have a TOP if the fetus was affected, access to antenatal diagnosis was deemed essential by half the sample. Some parents said they would not have embarked on a further

pregnancy if CVS was not available. However, several parents felt that even if they knew that their subsequent pregnancy was affected, they would not have a TOP, as it would be devaluing the life of the child they already had. The need for professionals to remain non-judgemental is reinforced.

Table 2 Thematic analysis

<i>Question 7(b): Did you discuss your subsequent pregnancy with any member of the CF team?</i>	
Fifteen respondents gave reasons why they did not discuss their pregnancy with any member of the CF team. They were:	
Already decided to have the test	3
Did not feel the need to discuss with the team as they knew the options	3
Did not have the need to discuss as their new partner was not a carrier in one case and IVF was being undertaken in the other	2
Parent wanted to carry on as normal	1
Because she had CF she felt that the team may dissuade the couple to carry on with the pregnancy	1
Perceived that discussion was not offered with the team	1
Discussed the options with a member of the genetics department	1
Felt that she was unable to cope as she and the child were unwell	1
Felt that it was not important	1
Concerned how the team would view the termination of pregnancy	1
<i>Question 11: Any other comments? (Applicable to all participants)</i>	
The team did not apply any pressure	1
Cystic fibrosis is not that serious	3
Cystic fibrosis is serious	4
Would have a termination if the baby was affected	5
Could not terminate the life of a CF child	5
Had a good service	6
Could not cope with the pressure of another CF child	5
One partner did want the termination of pregnancy, the other did not	2
Felt there was a bias to CVS and a termination of pregnancy	1

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ARCHIVIST

Dirt, flies, and trachoma

Trachoma is the world's second most prevalent cause of blindness. It is estimated that it affects some 146 million people worldwide and has blinded 6 million. It is a priority target of the World Health Organisation and the elimination of blindness due to trachoma by the year 2020 is the aim of the Alliance for the Global Elimination of Trachoma (GET 2020). The mainstays of this programme are surgery, antibiotic treatment, face washing, and environmental improvement (SAFE). Researchers in Mali (J-F Schémann and colleagues. *International Journal of Epidemiology* 2002;**31**:194–201) have sought a better understanding of the last two of these factors.

They performed a national survey between March 1996 and June 1997 examining a total of 15187 children under 10 years in 30 cluster samples in each of the country's seven regions excluding the capital Bamako. The prevalence of active trachoma was 35%, and of intense trachoma 4%. The two factors most strongly linked with trachoma were a dirty face (dust on face or discharge from nose or eye) and flies on the face. Each of these was associated with a more than threefold increase in likelihood of trachoma. Other risk factors were a dry environment, living in a small village, overcrowding in the home, and relative poverty as judged by household possessions. Frequent bathing and face washing, and proximity to a water source, a medical centre, or a social organisation such as a women's association all reduced the risk.

Improved hygiene and fly control together with general environmental and socioeconomic improvements will be important factors in the elimination of trachoma.