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Original Article

Newborn screening for cystic fibrosis — The parent perspective☆

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Abstract

Background: Newborn screening for CF started 01/2011 in Switzerland. We investigated the parents' opinions about the information received, their feelings, and overall approval of the screening.
Methods: This is a prospective questionnaire survey of all parents of positively screened children. Parents were phoned by CF-centres and invited for diagnostic investigations. They completed a questionnaire after the visit to the CF-centre.
Results: From 2011–2013, 246 families received the questionnaire and 138 (56%) replied. Of these 77 (60%) found the information received at birth satisfactory; 124 (91%) found the information provided in the CF-centre satisfactory. Most parents (n = 98, 78%) felt troubled or anxious when the CF-centre called, 51 (38%) remained anxious after the visit. Most parents (n = 122; 88%) were satisfied with the screening, 4 (3%) were not, and 12 (9%) were unsure.
Conclusions: The smooth organisation of the screening process, with personal information by a CF specialist and short delays between this information and the final diagnostic testing, might have contributed to reduce anxiety among parents. Most families were grateful that their child had been screened, and are happy with the process.

Keywords: Cystic fibrosis; Newborn screening; Parents; False-positives

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A partial version of this work that included fewer parent participants, 1 year after CF newborn screening has been implemented, was published in the following journals:

- *Rueegg CS*, et al. *One-year evaluation of a neonatal screening program for cystic fibrosis in Switzerland. Dtsch Aerztebl Int 2013*; *110*: *356–63.*
- *Rueegg CS*, et al. *Neugeborenen-Screening auf Cystische Fibrose — Evaluation nach einem Jahr. Paediatrica 2013*; *24*: *26–31.*

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1. Introduction

Newborn screening (NBS) for cystic fibrosis (CF) has many benefits. When CF is diagnosed and treated early, infant's food intake and digestion, growth, cognitive development and lung function can be improved, and exacerbations and hospitalizations decreased [1–3]. However, one of the drawbacks of NBS is that it can cause anxiety and distress to parents of children with positive screening results. After a positive screening, parents go through a phase of uncertainty and anxiety until the diagnosis of CF is confirmed or rejected [4–6]. Parents may also remain concerned after a false-positive screening result, even if CF is ruled out [7–11]. A second possible drawback is that NBS may identify CF-carriers, or children with rare and/or mild CF-mutations, whose further clinical course cannot be clearly predicted and who might be asymptomatic for years [7,12]. This may lead to unnecessary medicalization and impair mother–child bonding [5,9,13,14].

Several investigations have shown that distress can be reduced if parents are properly informed, and if the period between receiving the positive screening result and confirmation through diagnostic evaluation in a specialised clinic is kept short [10,15–22]. Other studies have investigated the opinion of unaffected parents toward genetic screening in general or screening for CF [23–26]. But we are not aware of a neonatal screening programme for CF, which, at the time of its introduction, has assessed prospectively the feedback of parents on a broad range of factors relevant for the screening process. Feedback from parents who have gone through the whole process could help to identify weaknesses of the current screening procedures and show areas that need further research.

In Switzerland, CF-NBS was implemented nation-wide in January 2011 [27–29]. Along with its introduction, we wanted to evaluate and assess the opinions of affected parents in relation to relevant steps of the screening process. The results of this evaluation will help to further improve the screening process in Switzerland, and might provide useful information for other countries that plan to establish or adapt their screening procedures for CF.

In particular, this study aimed to investigate: 1) the information about NBS provided to parents at birth; 2) parental satisfaction with the information they received during the CF-NBS; 3) their feelings at different stages of the CF-NBS; and, 4) their overall approval of the CF-NBS. Furthermore, we investigated the association of socio-demographic and clinical factors with information provision, parents' satisfaction with the information received, parents' feelings during the screening and parents' approval of the screening.

2. Patients and methods

2.1. Newborn screening in Switzerland

Screening for CF was implemented in the regular NBS programme. In Switzerland, most women give birth in a hospital; few give birth at home or in special birthing centres. Before or after delivery, all families must be informed about the NBS and receive a brochure about the screening, including a section on cystic fibrosis (INFO 1&2 in Fig. 1; brochure available under http://www.neoscreening.ch/en/download4.htm). By default, all children are screened, unless parents actively refuse to participate (possibility to opt out; done by about 5–10 families per year). The NBS requires a midwife or a nurse to take a heel-prick blood sample on filter paper (Guthrie Card) on the 4th day of life. The Guthrie Cards are sent to the national Swiss Newborn Screening Laboratory (SNSL) in Zurich for analysis.

2.2. Procedure of the Swiss CF-NBS

The CF-NBS has two parts (Fig. 1) [28,29]. The first is the *screening part*, done in the SNSL. The second, the *diagnostic part*, is done in dedicated CF-centres.

2.2.1. Screening part

In the SNSL, immunoreactive trypsinogen (IRT) is measured in the dried blood-spot of the Guthrie Card. If IRT is ≥50 ng/ml (≥99.2 percentile), the sample is genetically screened for a limited number of mutations [28,29]. If one or two mutations are found, the child of the sample is considered screening positive. If no mutations are found, and the initial IRT is ≥60 ng/ml, the midwife or family physician is asked to recall the family to perform another heel-prick test within 2–3 weeks. If the IRT of the second test is ≥50 ng/ml, the child of the sample is also considered as screening positive. All positively screened children are notified by phone from the SNSL to the CF-specialist of the respective family's nearest CF-centre.

2.2.2. Diagnostic part

The responsible CF-specialist of the centre calls the parents of children with positive screening result, informs them (INFO 3 in Fig. 1), and invites them for diagnostic evaluation (sweat test) on the next day. He/She tells the parents, that the screening result for CF was positive and needs further diagnostic investigation to find out if it reflects true CF disease, or the result is false positive. If the parents have more questions regarding CF, the CF specialist takes time to answer them on the phone but emphasises, that they will discuss these questions in more detail during the visit.

If the *sweat test is positive* ($Cl^- \geq 60$ *mmol/l*) *or borderline* ($Cl^- = 30–59$ *mmol/l*), a blood sample is taken, after obtaining written informed consent, for detailed genetic analysis. Parents are informed by the CF specialist about the test results and the disease (INFO 4 in Fig. 1), and appropriate treatment is initiated.

If the *sweat test is negative* ($Cl^- \leq 30$ *mmol/l*), parents are also informed by the CF-specialist in person on the same day. They are being explained that their child is healthy, but could be a CF carrier. In addition, they receive an information leaflet that explains in detail the meaning of a positive screening result with a normal sweat test result (INFO 4 in Fig. 1). The same leaflet is sent to their family doctor or paediatrician.

All parents are given an information sheet about voluntary genetic counselling.

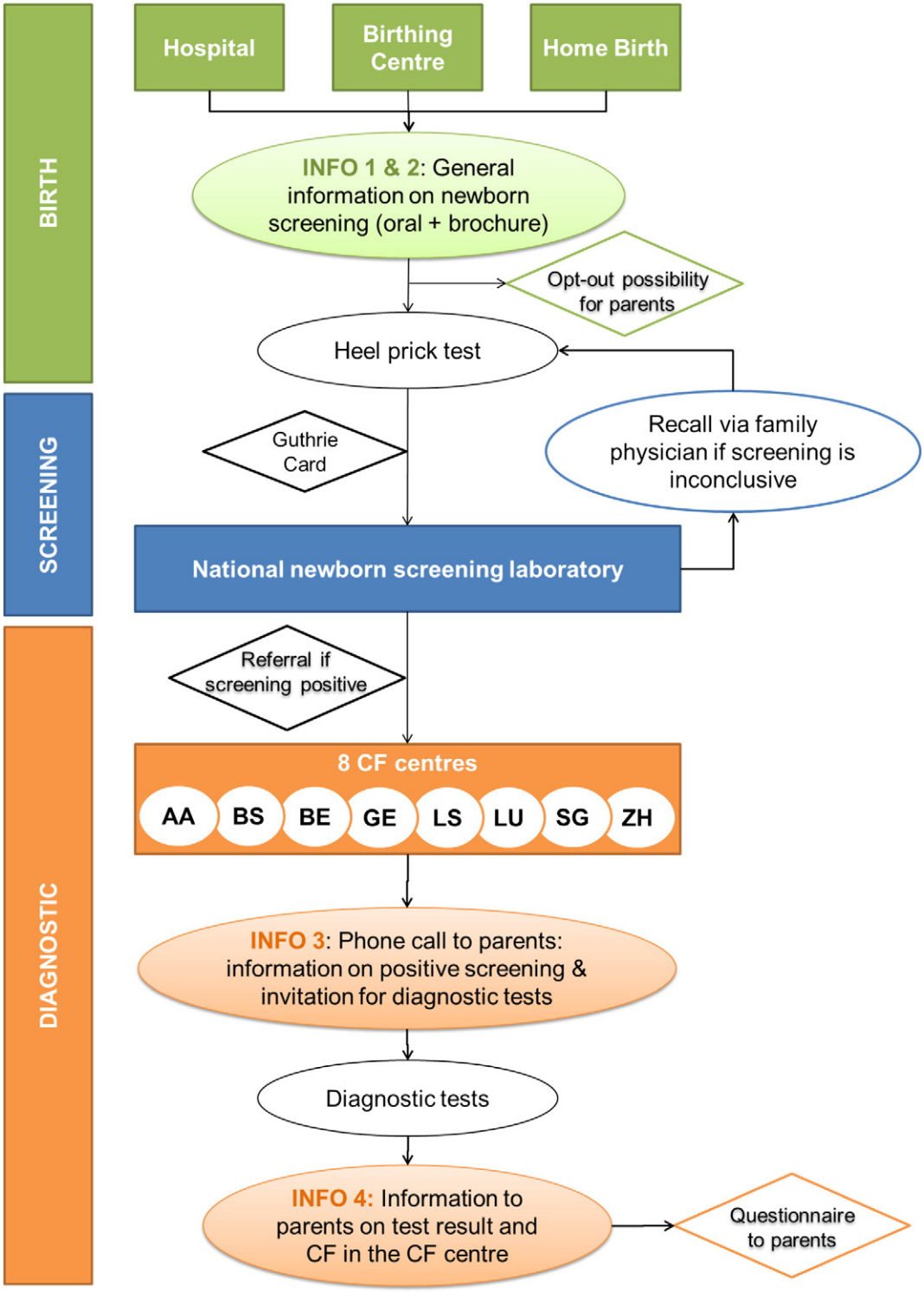

Fig. 1. **Procedure of the Swiss newborn screening for CF.** Fig. 1 shows the general procedure of the Swiss newborn screening for CF with a focus on the interaction with parents. It starts from the birth of the child to the final diagnosis. The detailed algorithm of the Swiss newborn screening for CF was published elsewhere (Ref. [28]). Abbreviations: AA, Aarau; BE, Bern; BS, Basel; CF, Cystic Fibrosis; GE, Geneva; LS, Lausanne; LU, Lucerne; SG, St. Gallen; ZH, Zurich.

2.2.3. Central database

The screening results of all positively screened children are entered into a central project database by SNSL staff. After the diagnostic evaluation, clinical data and diagnostic test results are entered on a form by the CF-physician, sent to the central database at the University of Bern and then entered by a central data manager. For this analysis, we used the following information from the database: child's birthdate; sex; responsible CF-centre

(categorised as large if they diagnosed >30 patients in 3 years); date of phone call to parents; date of visit to the CF-centre; final diagnosis; and, date of genetically-confirmed diagnosis.

2.2.4. Parental questionnaire

At the visit in the CF-centre, all parents received a questionnaire (4 pages, 25 items) in German (**Appendix I**), French or Italian, with a prepaid return envelope. The questionnaire was developed by clinicians (CF specialists) and epidemiologists to evaluate the new CF-NBS in Switzerland with specific questions related to the following topics: 1) the information received during the screening and the satisfaction with the information received; 2) parent's knowledge on CF prior to the screening; 3) parent's feelings during the screening procedure; 4) parent's overall opinion on the screening; 5) type of birth institution. In addition, we assessed socio-demographic characteristics of parents with standardised questions from the Swiss Census (language, education, migration background, number of children in the family) [30]. The questionnaire was pilot tested by members and relatives of the study team and translated by a professional translator into French and Italian. Parents completed the questionnaire at home, after the visit. At that point, their certainty about the final diagnosis depended on the sweat test result (conclusive or inconclusive).

Information provision

We asked parents whether or not, and from whom they had received the information brochure or any orally conveyed information on the NBS, before or after birth. Parents could choose from a list of possible specialists or add another person in free text.

Satisfaction with the information

Parents could indicate, for each type of information (INFO 1–4 in Fig. 1), if it had satisfied them. If not, they could explain why in an open format.

Parents' feelings

Parents could describe how they felt after the phone call from the CF-centre, and after the diagnostic evaluation in the CF-centre. They could choose between the answer categories "optimistic", "calm", "troubled", "very troubled", or add their own description of their feelings. We categorised answers into a binary variable ("calm & optimistic" vs. "troubled & anxious") for the analysis.

Overall approval

We asked parents if they were glad that their child had been screened (Yes/No). They could state why they approved or disapproved of the screening in an open format. They were also asked if they would change the screening procedure (Yes/No) and, if so, what they would change in an open format.

2.3. Statistical analysis

We used descriptive statistics (proportions and 95% confidence intervals (CI)) to describe the information provision to parents, parents' satisfaction with the information received, parents' feelings and their approval of the screening. To summarise individual open format statements, we manually combined statements of the same meaning into groups of similar content. The classification was done by one author (CSR); ambiguities were solved in discussion with CEK and JB. Because the final diagnosis could be a strong predictor of parents' feelings and approval of the screening, we stratified the analysis by final diagnosis (CF vs. no CF). We used univariable logistic regression models to assess the association of socio-demographic and clinical factors with parents' satisfaction with the information received, parents' feelings during the screening and parents' approval of the screening. We used univariable logistic regression models to investigate whether information provision differed by type of birth institution or screening year.

3. Results

3.1. Study population

Within the first 3 years of the Swiss CF-NBS, 253,346 IRT analyses were performed and 260 children with a positive screening result were referred to a CF-centre. Of these, 246 received a questionnaire and 138 returned it (response rate = 56%; **Supplemental** Fig. 1). Responders were more often of Swiss origin ($p < 0.001$) and had a child with CF ($p = 0.023$; Table 1), but they were otherwise similar.

In 54% (n = 74) of responders, the screened infant was their first-born; 52% (n = 71) had heard of CF before; and, 17% (n = 23) knew someone with CF. Most children were born in a hospital (94%; n = 129). Finally, 47 (34%) were diagnosed with CF, 2 (2%) with a CFSPID (CF Screen Positive, Inconclusive Diagnosis), and CF was ruled out in 89 (65%). The mean age of infants seen at the CF-centre was 22 days. On average, one day passed between the telephone call to parents and their visit in the CF-centre. On average, questionnaires arrived 11 days after the clinic visit.

3.2. Aim 1: information provision at birth

Of 138 parents, 13 (11%) remembered that they received the NBS information brochure before birth, 76 (58%) remembered receiving it after birth (**Supplemental Table 1**); 26 (20%) remembered receiving information orally before birth, and 106 (84%) remember receiving oral information after birth. Both brochures and oral information were most commonly received from nurses or midwives. Information was more often provided to parents of children born in birthing centres or at home (**Supplemental Table 2**). The rate at which information was provided did not change over the three years of the screening.

3.3. Aim 2: satisfaction with information provided

Of the 81 parents who remembered the brochure, 69 (85%) thought it was good (Fig. 2**;** INFO 1&2 in Fig. 1). The oral information received at birth was satisfactory to 77 parents (60%; INFO 1&2 in Fig. 1). The information given by the CF-physician over the telephone was satisfactory to 100 (74%; INFO 3 in Fig. 1) parents, and the information given personally

Table 1
Characteristics of parents responding and not responding to the CF newborn screening questionnaire.

	Responders (n = 138)		Non-responders (n = 108)		
	n	%[a]	n	%[a]	p-value[b]
Child's demographic characteristics					
Child's year of birth					0.495
2011	48	35	36	33	
2012	47	34	31	29	
2013	43	31	41	38	
Child's sex					0.706
Male	65	47	53	50	
Female	73	53	54	50	
Family's socio-demographic characteristics					
Spoken language					n.a.[c]
Swiss German	92	68	n.a.[c]	n.a.[c]	
French	36	27			
Italian	7	5			
Highest parental education					n.a.[c]
Primary	47	35	n.a.[c]	n.a.[c]	
Secondary	24	18			
Tertiary	64	47			
Country of origin of the mother					**<0.001**
Switzerland	95	69	50	53	
Other	42	31	45	47	
Number of children					n.a.[c]
Screened child is the first child	74	54	n.a.[c]	n.a.[c]	
Screened child has older siblings	64	46			
Family has heard of CF before					n.a.[c]
No	71	52	n.a.[c]	n.a.[c]	
Yes	67	48			
Family knows someone with CF					n.a.[c]
No	115	83	n.a.[c]	n.a.[c]	
Yes	23	17			
Clinical characteristics					
Birth institution					0.195
Hospital	129	94	94	88	
Birthing centre	2	2	1	1	
Home	7	5	12	11	
Type of CF-centre					0.990
Small	64	46	50	46	
Large (>30 patients diagnosed in 3 years)	74	54	58	54	
Child's final diagnosis					**0.023**
No CF	89	65	77	71	
CF	47	34	24	22	
CFSPID	2	2	7	7	

	Median	SD	Median	SD	p-value[d]
Time variables					
Time between telephone and clinic visit (d)	1.0	1.8	1.0	2.3	0.168
Age at visit in the CF-centre (d)	22.0	12.3	23.0	33.2	0.136
Time between clinic visit and questionnaire reply (d)	11.0	57.0	n.a.[c]	n.a.[c]	n.a.[c]
Age at genetically confirmed diagnosis (d)	34.5	21.1	35.0	23.1	0.952

Note: Percentages are based upon available data for each variable.
Abbreviations: CF, cystic fibrosis; CFSPID, cystic fibrosis screen positive, inconclusive diagnosis; d, days; n.a., not available.
p-values smaller than 0.05 are highlighted in bold.
[a] Column percentages are given.
[b] p-value calculated from chi-square statistics comparing responders and non-responders.
[c] Information not available for non-responders.
[d] p-value calculated from two-sample mean-comparison test (t-test) comparing responders and non-responders.

at the CF-centre was satisfactory to 124 parents (91%; INFO 4 in Fig. 1).

Some parents (n = 4) were dissatisfied with the brochure because they felt it did not offer a good explanation of the

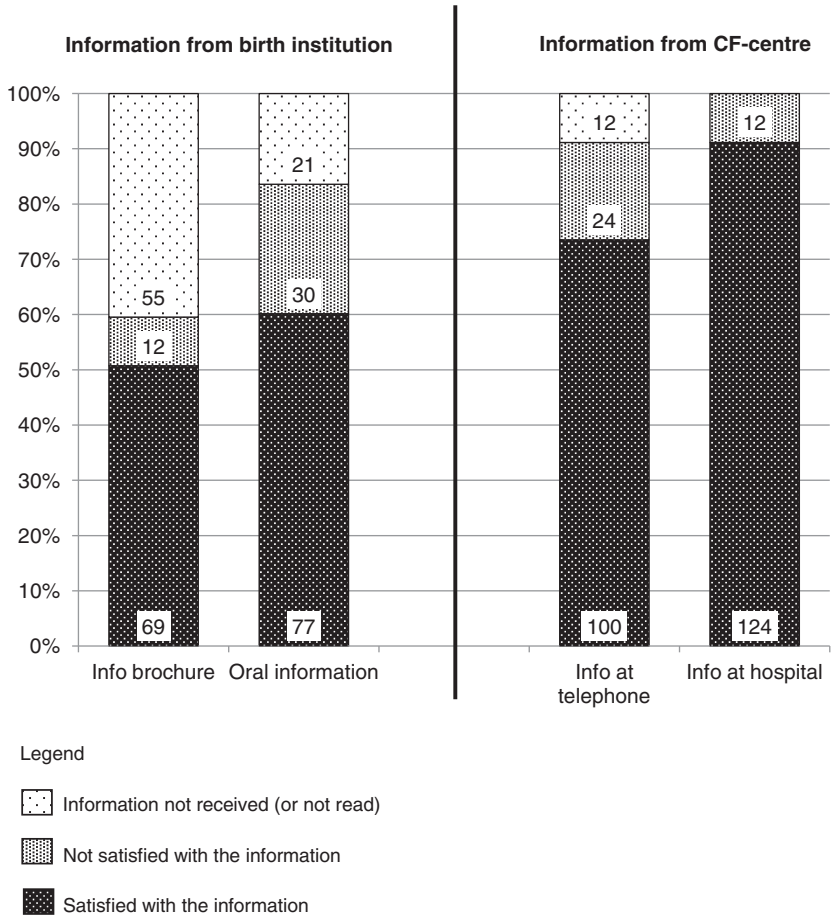

Fig. 2. Parents' satisfaction with the information received throughout the screening process. Fig. 2 shows the satisfaction of parents with the information received at different stages of the screening procedure. The numbers in the columns reflect the number of parents (N) within each category. Note: Percentages are based upon available data for each variable. This figure was published in part before including fewer participants after 1 year of the implementation of the newborn screening for CF in Switzerland (in *Rueegg CS*, et al. *Deutsches Aerzteblatt International 2013*; *110*: *356–63* (Ref. [29])).

diseases that were screened for, or that they did not understand the information (n = 2; **Supplemental Table 3**). Common reasons for parental dissatisfaction with oral information given at birth were that the screened diseases were not well explained (n = 14), or that they would have liked more details (n = 9). The 24 parents who were unsatisfied with the information they received by phone said that the caller had not explained the test result and the disease (n = 9), or had provided superficial information and instead focused on arranging the appointment (n = 5). Parents who were unhappy with the information they received in the CF-centre wanted to be told more clearly that a negative sweat test meant a healthy child (n = 3), or wished they had been given more information at an earlier stage of screening (n = 3).

In univariable logistic regression, we found that satisfaction with the *information received at birth* (brochure and oral information) was determined mainly by socio-demographic factors. Parents of foreign origin and of a first child were more dissatisfied (**Supplemental Table 4**).

The *information received by telephone* was less satisfactory to parents of children diagnosed with a CF (OR 2.23, p = 0.044), or parents of younger infants (OR 0.93 per day older, p = 0.001).

Parents of a first child were more dissatisfied with *the information in the CF-centre* (OR 0.21, p = 0.024).

3.4. Aim 3: determining the feelings of parents during the screening process

Most parents (n = 98; 78%) were troubled or anxious after the CF-centre called, but only 51 (38%) were still anxious after the visit (Table 2; 19/88 families with a healthy child (21.6%) and 32/48 families with a child with CF [66.7%; p < 0.001]).

Negative feelings after the phone call from the CF-centre were more frequent in more highly educated parents (p = 0.003; **Supplemental Table 5**), and after a call from large CF-centres (p = 0.005). *Negative feelings after the visit in the CF-centre* were more often found in families of foreign origin (p = 0.002) and if their infant had CF (p < 0.001).

3.5. Aim 4: parents' overall approval

Most parents (122 of 138; 88%) were glad that their child had been screened: 84% (n = 75) of parents without CF compared to 96% (n = 47) of families with CF (p = 0.103; Table 2). Few parents gave specific reasons for their

Table 2
Parents' feelings during the screening and overall approval of the screening.

	Overall		Families without CF		Families with CF		p-value[b]
	n	%[a]	n	%[a]	n	%[a]	
Feelings during the screening procedure							
Feeling after phonecall from CF-centre							0.445
Troubled & anxious	98	78	65	77	7	18	
Calm & optimistic	27	22	20	24	33	82	
Feeling after visit in the CF-centre							**<0.001**
Troubled & anxious	51	38	19	22	32	67	
Calm & optimistic	85	62	69	78	16	33	
Approval of the screening							
Families approve the screening							0.103
Yes	122	88	75	84	47	96	
No	4	3	4	5	0	0	
Not sure	12	9	10	11	2	4	
Families would change something							0.625
Yes	53	45	36	46	17	42	
No	66	55	42	54	24	58	

Note: Percentages are based upon available data for each variable.
Abbreviations: CF, cystic fibrosis.
[a] Column percentages are given.
[b] p-value calculated from chi-square statistics comparing families with and without CF.

disapproval or approval of the screening; three families of 16 who disapproved the screening and 40 families of 122 who were in favour of the screening (**Supplemental Table 6**). Three families stated that screening caused anxiety, and that they would have preferred not to know their child had CF so they could enjoy the symptom-free time. Among parents who were happy with the screening, 22 said that timely treatment benefitted the child, nine were glad to know that their child was healthy, four said that screening helps to avoid complications, two said that they always want to know if their child has a disease, and two were glad to know that their child was a carrier, in case the child showed symptoms later or will have children.

Table 3
What parents would change about the national newborn screening for cystic fibrosis, stratified by final CF diagnosis.

	What[a] parents of children with diagnosed CF would change (N=17)
Information provision	• Better information about the screening and the diseases in the birth institution (n=3)
	• More information about cystic fibrosis, also positive aspects and hope (n=3)
	• Inform parents directly about the screening result (n=2)
	• Inform about Guthrie test before birth by gynaecologist or midwife (n=1)
	• Written information about the disease and the meaning for relatives (n=1)
	• Make sure the information brochure is really provided at birth (n=1)
Screening procedure	• Better communication among involved parties(birth hospital, CF-centre, paediatrician, parents) (n=2)
	• Shorter waiting time throughout the screening procedure (n=3)
	• Nothing stated (n=1)
	What[a] parents of children without CF would change (N=36)
Information provision	• More detailed information from the birth institution about the Guthrie test and what a positive screening result means (n=13)
	• Inform already prior to birth, i.e. by gynaecologist (n=2)
	• Better information at the telephone (that also healthy children (carriers) can have a positive screening result; type ofdisease) (n=2)
	• Better information why a 2nd Guthrie test is needed at the paediatrician (n=1)
	• Give written information already after normal sweat test and not only after detailed genetic analysis (n=1)
	• Information at sweat test → state normal (healthy) results immediately (n=1)
	• Tailor information to lay persons without using jargon (n=1)
Screening procedure	• Whole coordination should be done by 1 central station, information gets faster to the family and comes from known sources (i.e. paediatrician, gynaecologist) (n=4)
	• Shorter waiting time throughout the screening procedure (n=3)
	• Do not make a 2nd (or 3rd) Guthrie test but directly the sweat test (n=2)
	• Improve the screening test to reduce false positives (n=2)
	• Make the Guthrie test directly in the birth institution for immediate results (n=1)
	• Give the possibility to make the sweat test in a place more close (n=1)
	• Nothing stated (n=2)

Abbreviations: CF, cystic fibrosis; N, number; NBS, newborn screening.
[a] Parents could indicate in an open format question what they would change in the CF newborn screening. Statements of the same meaning were manually combined in this table.

Changes to the screening procedure were suggested by 45% (n = 53) of parents (Table 2). Among families with a child with CF (n = 17), most suggestions (n = 11) related to improving the way information was provided (Table 3). Other comments were related to the screening procedure, i.e. better communication among involved parties (n = 2), and shorter waiting times throughout the screening procedure (n = 3). Among families whose child did not have CF (n = 36), most suggestions were again related to provision of information (n = 21). Other suggestions concerned the screening procedure: coordination over a central station (n = 4), shorter waiting time (n = 3), reduction of false positives (n = 2), or conducting the sweat test immediately, instead of repeating the heel-prick test (n = 2).

4. Discussion

Most parents (78%) were troubled or anxious when they were informed that the results of the screening test had been positive, but only 22% of families without CF, and 67% with CF, were still worried after the diagnostic evaluation. Overall, most parents (88%) were satisfied with the screening irrespective of the final diagnosis (84% of parents without CF compared to 96% of families with CF).

4.1. Strengths and limitations of the study

The successful implementation of the CF-NBS programme in Switzerland was attributable to the already well-established nationwide NBS programme with its single, centralised screening laboratory [29]. The opt-out procedure explains the high participation rate in the screening. Our study is limited by the fact that we have answers only from parents whose infants had positive results on screening, and were then seen in a CF-centre. We do not have information from parents recalled for a second heel-prick test, for which the results were normal. Furthermore, parent's feelings were assessed retrospectively, after they had been given the final diagnosis. Therefore, their emotional state when they completed the questionnaire might have affected their recollections of past feelings. The study was relatively small, and comments from individual parents should not be given undue weight. Adapting the screening procedure based on suggestions from the few parents who were not pleased might lead to a decrease in satisfaction of the larger group of parents who were happy with the current procedures.

4.2. Interpretation of the information provision and satisfaction

Consistent with the literature [15–17,20], we found that well-informed parents are less stressed by the testing process. All NBS programmes debate the amount of information that should be given to new parents, who are already overloaded with brochures. In Switzerland, the law on genetic screening within the NBS has been in force since 2007, and all parents must be told about the screening and their option to refuse, so a chapter on 'CF' was added to the existing NBS brochure. It is, however, possible that in daily life not all parents receive the brochure; some might also not remember it. But if the children of these parents would have been screened negative, they would probably never have missed the information.

Some parents felt that details about the screening test and the disease were missing when they were invited by phone for diagnostic evaluation. But information provided during this phone call is deliberately minimal without mentioning the respective disease, so as to reduce parental anxiety and to discourage parents from researching CF on the Internet before they receive accurate information from a specialist.

4.3. Interpretation of parents' feelings

False-positive results and negative second heel-prick tests are a challenge to every NBS programme, since they cause parental anxiety and unnecessary medical examinations. When children with positive screening results are tested in a CF-centre, their parents often feel anxious or depressed while awaiting definitive results [6,10]. A French study investigated the short- and long-term psychological effects of false-positive results in CF-NBS and found that 96.5% of parents were anxious at the time of the sweat test [14]. However, 86% were entirely reassured 3, 12, and 24 months after the test. In our study, 78% of parents were troubled or anxious after the phone call, but after the visit in the CF-centre, only 22% of the families with a false-positive result were still troubled. Since we only asked parents about their feelings once, shortly after diagnostic evaluation in the CF-centre (median time to questionnaire response 11 days, interquartile range 7–24 days), we do not know the long-term effects of a false-positive screening result.

Parents who were approached by larger CF-centres were more anxious after the phone call. It is possible that a call from a large university clinic might trigger more fear than a call from a regional and more familiar hospital. Or it may also be possible that calls from a large clinic that call more patients, may be less personally accessible or reassuring. More educated parents were more concerned after the phone call than less educated parents, perhaps because more educated parents were more aware of the potential consequences. We found that uncertainty and stress persisted more in migrants, perhaps because of language problems that may have prevented them from understanding the information provided.

4.4. Interpretation of the overall satisfaction and implication into practise

Overall, 88% of parents were glad that their child had been screened, independent of final diagnosis. Nearly half of the parents suggested that the screening programme could be improved; most of them wished better information. This means that the NBS brochure should be handed out more systematically, and that midwives or nurses responsible for giving the oral information at the birth clinic might profit from special training.

CF specialists should also be aware of the effect of the information given by phone and in the CF-centres, since this information can either reassure or cause anxiety to parents. The period between informing parents about a positive screening result (phone call) and their appointment at the CF-centre should be as short as possible. Parents should only be called when an appointment can be offered within the following days.

Many of the suggestions made by parents are already being used to improve the screening process. For example, the Swiss CF-NBS is trying to reduce false-positive screening results and recalls for a 2nd heel-prick test by adding an additional parameter: pancreatitis associated protein (PAP)-measurement [31].

5. Conclusion

The large majority of families, independent of the child's final CF diagnosis, were glad that their infants were screened, and their suggestions to improve the process have already been used to improve the screening procedure and this refinement will be continued.

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Conflict of interest

No conflict of interest to state for any of the authors.

Appendix A. Supplementary data

Supplementary data to this article can be found online at http://dx.doi.org/10.1016/j.jcf.2015.12.003.

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