Clinical obligations and public health programmes: healthcare provider reasoning about managing the incidental results of newborn screening

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► Additional supplemental tables 1–4 are published online only at http://jme.bmj.com/content/vol35/issue10

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ABSTRACT

Background: Expanded newborn screening generates incidental results, notably carrier results. Yet newborn screening programmes typically restrict parental choice regarding receipt of this non-health serving genetic information. Healthcare providers play a key role in educating families or caring for screened infants and have strong beliefs about the management of incidental results.

Methods: To inform policy on disclosure of infant sickle cell disorder (SCD) carrier results, a mixed-methods study of healthcare providers was conducted in Ontario, Canada, to understand attitudes regarding result management using a cross-sectional survey (N = 1615) and semistructured interviews (N = 42).

Results: Agreement to reasons favouring disclosure of SCD carrier results was high (65.1%–92.7%) and to reasons opposing disclosure was low (4.1%–18.1%). Genetics professionals expressed less support for arguments favouring disclosure (35.3%–78.8%), and more agreement with arguments opposing disclosure (15.7%–51.9%). A slim majority of genetics professionals (51.9%) agreed that a reason to avoid disclosure was the importance of allowing the child to decide to receive results. Qualitatively, there was a perceived "duty" to disclose, that if the clinician possessed the information, the clinician could not withhold it.

Discussion: While a majority of respondents perceived a duty to disclose the incidental results of newborn screening, the policy implications of these attitudes are not obvious. In particular, policy must balance descriptive ethics (ie, what providers believe) and normative ethics (ie, what duty-based principles oblige), address dissenting opinion and consider the relevance of moral principles grounded in clinical obligations for public health initiatives.

Newborn screening programmes assess infants who appear to be well to identify those few at increased risk of having a treatable disorder.[1] Although designed to reduce mortality and morbidity,[2] newborn screening programmes may also generate incidental information, such as carrier status (ie, unaffected heterozygotes). Newborn screening for the sickle cell disorders (SCD) provides a paradigmatic case. Screening for SCD is justified by high quality evidence that treatment with prophylactic penicillin leads to reduced mortality in affected children.[3 4] However, screening technologies that identify affected infants also identify virtually all SCD carriers.

In most jurisdictions where newborn screening for SCD occurs, carrier results are routinely disclosed. Commentators state that to do otherwise would result in withholding relevant information from parents.[5–9] Yet whether these incidental results should be routinely disclosed remains controversial. The Committee on Bioethics of the American Academy of Pediatrics,[10] and the US Institute of Medicine Committee on Assessing Genetic Risks concluded that information about an infant's carrier status generated through newborn screening should be disclosed only if informed consent is obtained.[11] As these recommendations imply, the routine disclosure of infant carrier results in the absence of consent imposes a moral burden by requiring parents to receive information about their infant that has no clear health implications.[12] This is especially troubling in light of an extensive literature arguing that genetic information should not be disclosed in childhood unless medically necessary,[10 13 14] even when results are incidental.[14]

In addition to ethical disquiet, concerns remain about other harms that may ensue from disclosure. Whereas little research on the effects of SCD carrier result disclosure has been conducted,[3 15] investigators have long been concerned about parents experiencing undue stress or anxiety, perceiving their child as excessively "vulnerable",[16] leading to over-medicalisation or stigmatisation.[17–21] It is also feared that parents and providers might misunderstand the meaning of carrier status and confuse it with the disease itself, potentially adding to any related harms.[22] Finally, concern about SCD carrier identification is exacerbated by its high prevalence in African-origin populations and the charged history of population screening for SCD in the USA.[23–25] Even if careful communication strategies might allay certain harms, evidence suggests that disclosure practices vary widely,[26 27] and that many of those who communicate with families are ill prepared.[28–30]

In Ontario, Canada, expansion of the newborn screening panel in 2006 to include SCD generated policy interest in the issue of carrier identification. We conducted a mixed-methods study of consumer and healthcare provider attitudes to inform policy on the disclosure of SCD carrier results; in the interim, the province adopted a provisional non-disclosure policy. In this paper, we report the results of research with providers regarding the management of carrier status information generated through newborn screening.

METHODS

With approval from the Hamilton Health Sciences Research Ethics Board, we conducted a mixed-methods study in Ontario, Canada, in 2007

involving a postal survey of providers, and qualitative research with providers, consumers and advocates. We report here on results from the provider survey and from open-ended, semistructured provider interviews.

We generated a random sample of healthcare providers, stratified into seven groups. Obstetrician/gynaecologists (n = 498), midwives (n = 339), obstetric/postpartum nurses employed in hospitals (n = 725), family physicians (n = 729) and paediatricians (n = 569) have a current or potential role in preparing parents for newborn screening. In addition, genetics professionals (clinical and biochemical geneticists, genetic counsellors; n = 105) were included because they may be involved in the follow-up of positive newborn screening test results. Finally, haematologists (n = 148) were included because of their particular interest in newborn screening for SCD. Potential respondents were identified using MDSelect, the Canadian Medical Directory, and directories from the College of Midwives of Ontario, the College of Nurses of Ontario and the Canadian Association of Genetic Counsellors.

A self-administered questionnaire was mailed to providers accompanied by a covering letter, a Can$2 coffee-shop coupon and a postage-paid reply envelope. In accordance with the Dillman tailored design method,[31] five mailings were completed over an 8-week period. The questionnaire was developed by a multidisciplinary team based on a review of the literature and pilot tested among two to three members of each group surveyed to ensure face validity. The initial sections of the questionnaire (up to 15 items) were provider group specific to assess involvement in prenatal care or care of infants in the first days of life and to gauge perceptions of barriers related to newborn screening care. The bulk of the questionnaire (69 items) was the same for each provider group. Using categorical responses and five-point Likert scales measuring strength of agreement or frequency of practice, as appropriate, we assessed: (1) knowledge and confidence about newborn screening and SCD; (2) involvement in newborn screening or care of persons with SCD; (3) practices and attitudes regarding informing parents about newborn screening before sample collection or caring for families with positive screening results; (4) core beliefs about newborn screening, generally and (5) demographics. We focus here on analysis of the 12 questions that assessed attitudes towards the management of incidental carrier results (see tables 1 and 2).

In addition, we conducted open-ended, semistructured interviews with a purposive sample of 42 providers across the same seven groups. Provider respondents included key informants designated by professional associations or known to be interested in newborn screening, postal survey respondents willing to participate in an interview and individuals referred through snowball sampling. The interviews engaged respondents about their experience with newborn screening or SCD and posed three core questions: (1) what should be done with SCD carrier results generated through newborn screening; (2) did the generation of SCD carrier results alter the way in which newborn screening should be provided (eg, consent for newborn screening or carrier result disclosure); and (3) if disclosed, how should this be done. The interviews were conducted in a conversational style and involved both open-ended questions to elicit opinion, and probes to allow respondents to consider the potential benefits or harms of routine disclosure.

Table 1 Reasons to disclose SCD carrier status results—parents should be provided with the information that their infant is a sickle cell carrier because…

	All	OB	RN	MW	FP	PED	GEN	HEM	p Value
…of the importance for identifying future reproductive risks for carrier infant									
A/SA	1393 (88.1)	182 (88.3)	444 (92.9)	217 (87.5)	253 (88.2)	231 (85.6)	28 (53.8)	38 (95)	<0.01
N/D/SD	188 (11.9)	24 (11.7)	34 (7.1)	31 (12.5)	34 (11.8)	39 (14.4)	24 (46.2)	2 (5)	
…of the importance for informing parents their own reproductive risks									
A/SA	1354 (85.6)	168 (81.6)	427 (89.3)	205 (82.7)	254 (88.5)	227 (84.1)	41 (78.8)	32 (80)	0.02
N/D/SD	227 (14.4)	38 (18.4)	51 (10.7)	43 (17.3)	33 (11.5)	43 (15.9)	11 (21.2)	8 (20.0)	
…of the newborn screening programmes' responsibility to disclose information it generates									
A/SA	1345 (85.4)	172 (84.3)	425 (88.7)	219 (88.7)	241 (84.9)	220 (81.5)	33 (63.5)	35 (89.7)	<0.01
N/D/SD	230 (14.6)	32 (15.7)	54 (11.3)	28 (11.3)	43 (15.1)	50 (18.5)	19 (36.5)	4 (10.3)	
…of parents' right to information that exists about their infant									
A/SA	1464 (92.7)	184 (89.3)	466 (97.3)	236 (94.4)	261 (91.6)	247 (91.8)	35 (67.3)	35 (89.7)	<0.01
N/D/SD	116 (7.3)	22 (10.7)	13 (2.7)	14 (5.6)	24 (8.4)	22 (8.2)	17 (32.7)	4 (10.3)	
…it is important to avoid misleading parents who might believe nothing was found									
A/SA	1461 (92.7)	181 (88.3)	458 (95.6)	233 (94.3)	265 (93.3)	250 (92.6)	37 (71.2)	37 (94.9)	<0.01
N/D/SD	115 (7.3)	24 (11.7)	21 (4.4)	14 (5.7)	19 (6.7)	20 (7.4)	15 (28.8)	2 (5.1)	
…of the importance of using infrastructure for reporting results that is already in place									
A/SA	1009 (65.1)	125 (63.1)	360 (75.9)	161 (66.3)	174 (62.4)	152 (56.9)	18 (35.3)	19 (48.7)	<0.01
N/D/SD	542 (34.9)	73 (36.9)	114 (24.1)	82 (33.7)	105 (37.6)	115 (43.1)	33 (64.7)	20 (51.3)	

Values are number (%).
FP, family physicians; GEN, genetics professionals; HEM, haematologists; MW, midwives; OB, obstetricians; PED, paediatricians; RN, registered nurses; SCD, sickle cell disorder. A, agree; SA, strongly agree; N, neutral; D, disagree; SD, strongly disagree.

Data analysis

Data from completed questionnaires were entered using Snap Survey Software (version 8), and analysed using SPSS (version 16). Likert scales were collapsed into binary categories. Descriptive statistics were computed for all variables measured, including frequency counts and percentages. We used the χ^2 test to determine differences in categorical variables. Unadjusted odds ratios (OR), 95% CI and p values are reported as appropriate. A probability level of <0.05 was used to determine statistical significance.

All interviews were transcribed, entered into our database and coded by two to three members of the research team using qualitative data analysis software (NVivo, version 7). For the current paper, we coded transcript sections that explored rationales regarding disclosure of carrier results. Next, we categorised coherent rationales related to disclosure, most of which were prestructured, arising from the 12 reasons for and against disclosure included in the questionnaire. We next identified dissonant cases to capture which pro or antidisclosure rationales changed respondents' views on disclosure. This analysis suggested that respondents' rationales could be organised into a hierarchy, as some reasons were motivating and others supportive of a given orientation towards disclosure. Throughout, we adopted a modified grounded theory approach[32–34] integrating the iterative and constant comparative method,[34] and writing as an analytical device,[35] with a reflexive approach to interpretation that drew on pre-existing concepts to guide us in understanding what was in the data.[36]

RESULTS

About respondents

Of the 3113 surveys that were mailed, 1615 were completed and returned, generating an uncorrected response rate of 51.9%. After adjusting the response rate for all ineligible respondents (ie, those we could not contact despite repeated efforts, and those not in practice or not eligible to respond because their practice involves neither providing care for newborns nor providing medical care for individuals with SCD; n = 544), our corrected response rate was 62.9% (adjusted N = 2569; 54.1% of obstetrician/gynaecologists, 76.7% of midwives, 72.6% of nurses, 50.6% of family physicians, 63.1% of paediatricians, 68.0% of genetics professionals, 51.9% of haematologists).

Data related to respondents' practice characteristics, confidence in and familiarity with Ontario's newborn screening programme as well as SCD, and attitudes regarding when and how SCD carrier results should be reported to parents are presented in supplemental tables 1–4, respectively (available online only).

About majority/dominant opinion

Majority: survey results

Agreement (defined as agree or strongly agree) with reasons favouring disclosure of SCD carrier status was high (65.1%–92.7%) (table 1) and with reasons opposing disclosure was low (4.1%–18.1%; table 2). Genetics professionals expressed less agreement than the majority for arguments favouring disclosure (35.3%–78.8%) and more agreement than the majority for arguments opposing disclosure (15.7%–51.9%). The sole reason for avoiding disclosure that attracted majority support from any provider group was the importance of allowing the child to decide about the receipt of carrier results; a slim majority of genetics professionals agreed with this item (51.9%). Differences in agreement across provider groups are statistically significant for all items.

Table 2 Reasons to not disclose SCD carrier status results—parents should not be provided with the information that their infant is a sickle cell carrier because it is important…

	All	OB	RN	MW	FP	PED	GEN	HEM	p Value
…to avoid risk that parents may misunderstand the meaning of carrier status									
A/SA	65 (4.1)	11 (5.3)	12 (2.5)	10 (4.1)	10 (3.5)	12 (4.5)	8 (15.7)	2 (4.9)	<0.01
N/D/SD	1506 (95.9)	195 (94.7)	461 (97.5)	236 (95.9)	276 (96.5)	256 (95.5)	43 (84.3)	39 (95.1)	
…to minimise provision of information that does not influence the medical management of the child									
A/SA	72 (4.6)	14 (6.8)	9 (1.9)	9 (3.7)	14 (4.9)	12 (4.5)	11 (21.2)	3 (7.3)	<0.01
N/D/SD	1493 (95.4)	192 (93.2)	461 (98.1)	235 (96.3)	271 (95.1)	255 (95.5)	41 (78.8)	38 (92.7)	
…to avoid creating additional costs for healthcare system									
A/SA	98 (6.3)	16 (7.8)	15 (3.2)	20 (8.1)	19 (6.7)	12 (4.5)	14 (27.5)	2 (4.9)	<0.01
N/D/SD	1469 (93.7)	189 (92.2)	456 (96.8)	227 (91.9)	266 (93.3)	255 (95.5)	37 (72.5)	39 (95.1)	
…that the child decide if and when they want this information									
A/SA	123 (7.9)	15 (7.3)	13 (2.8)	24 (9.8)	17 (6.0)	25 (9.4)	27 (51.9)	2 (4.9)	<0.01
N/D/SD	1438 (92.1)	191 (92.7)	455 (97.2)	221 (90.2)	267 (94.0)	240 (90.6)	25 (48.1)	39 (95.1)	
…not to assume that people want to learn about their or their infant's carrier status									
A/SA	278 (17.9)	29 (14.3)	61 (13)	64 (25.9)	53 (18.8)	40 (15.2)	25 (48.1)	6 (14.6)	<0.01
N/D/SD	1279 (82.1)	174 (85.7)	408 (87)	183 (74.1)	229 (81.2)	223 (84.8)	27 (51.9)	35 (85.4)	
…not to assume that people will want to learn about their or their infant's reproductive risks									
A/SA	281 (18.1)	33 (16.2)	67 (14.3)	59 (24.3)	51 (18.0)	42 (15.9)	22 (42.3)	7 (17.1)	<0.01
N/D/SD	1274 (81.9)	171 (83.8)	401 (85.7)	184 (75.7)	232 (82.0)	222 (84.1)	30 (57.7)	34 (82.9)	

Values are number (%).
FP, family physicians; GEN, genetics professionals; HEM, haematologists; MW, midwives; OB, obstetricians; PED, paediatricians; RN, registered nurses; SCD, sickle cell disorder. A, agree; SA, strongly agree; N, neutral; D, disagree; SD, strongly disagree.

Table 3 Pattern of attitudes for dissenting individuals: reasons to disclose—parents should be provided with the information that their infant is a sickle cell carrier because…

	Dissenters	Non-dissenters	Unadjusted OR (95% CI)
…of the importance for identifying future RR for carrier infant			
A/SA	305 (78.4)	1071 (91.2)	0.35 (0.25 to 0.48)*
N/D/SD	84 (21.6)	103 (8.8)	1.0
…of the importance for informing parents their own RR			
A/SA	309 (79.4)	1030 (87.7)	0.54 (0.40 to 0.73)*
N/D/SD	80 (20.6)	144 (12.3)	1.0
…of the newborn screening programmes' responsibility to disclose information it generates			
A/SA	302 (77.6)	1029 (88.1)	0.47 (0.35 to 0.63)*
N/D/SD	87 (22.4)	139 (11.9)	1.0
…of parents' right to information that exists about their infant			
A/SA	334 (86.1)	1112 (94.8)	0.34 (0.23 to 0.50)*
N/D/SD	54 (13.9)	61 (5.2)	1.0
…it is important to avoid misleading parents who might believe nothing was found			
A/SA	337 (87.1)	1106 (94.9)	0.39 (0.26 to 0.56)*
N/D/SD	50 (12.9)	60 (5.1)	1.0
…of the importance of using infrastructure for reporting results that is already in place			
A/SA	205 (53.1)	791 (68.8)	0.51 (0.41 to 0.65)*
N/D/SD	181 (46.9)	358 (31.2)	1.0

Values are number (%).
*$p<0.01$. OR, odds ratio; RR, reproductive risk. A, agree; SA, strongly agree; N, neutral; D, disagree; SD, strongly disagree.

Dominant: qualitative results

A dominant emergent theme was the perception of a clinical duty to disclose SCD carrier status results. This imperative was seen to motivate disclosure independently of the perceived benefits (notably, reproductive risk identification) or harms (eg, parental misunderstanding, revelation of misattributed paternity, etc) of doing so.

A duty to disclose was perceived to arise from the consumer's ownership of their information:

> "You don't do a test and not reveal the information. […] It's their results. It's their knowledge." (obstetrician, I 11)

Some providers feared the harm that secrets could create:

> "I have a problem with secrets. I, I think that secrets of any kind are destructive, are potentially destructive or harmful and that if there is information it should be given." (clinical geneticist, I 3)

Reflecting concerns about legal liability from non-disclosure, respondents noted that

> "…wrongful birth suits could be avoided down the road for some physicians." (haematologist, I 4)

Respondents argued that test results should be provided even when they are not anticipated and suggested several analogies to the context of clinical care.

> "[in] an ultrasound of the kidneys … they found a benign tumour […] I do feel that I should explain that to the patient […] and I realise that, you know, this can be anxiety provoking and ah, you know, the easier thing would be just to say, 'Oh your ultrasound is normal,' and that's the end of it. But […] you know, five years later he goes to some other doctor and has another ultrasound […] So then the doctor says, 'Oh maybe this is something new.' And so on. So I think, really, a patient should, should know […] in general I would say, 'It's better to share'." (family physician, I 42)

Not all clinicians were clear about who was obliged by the duty to disclose. For most, the duty was considered to be incumbent on the clinician who received the report; by extension, if the clinician did not possess the information, no such duty to disclose existed:

> "If it's not reported to me in a report […] there's nothing for me to report or not report to my client. But certainly if, as a clinician, if I'm provided with the information in a report I would … [pause] … I would disclose that to the client." (midwife, I 16)

However, some respondents extended a duty to disclose to the system as a whole:

> "If the health care system knows … that individual has a right to that knowledge."(family physician, I 26)

About dissent

Dissenting individuals: survey results

To understand better minority attitudes regarding the disclosure of SCD carrier status information we defined 'dissenting individuals' (N = 391) to include anyone who agreed or strongly agreed with any of the six reasons opposing disclosure listed in the questionnaire. In addition to supporting one or more reasons opposing disclosure, these individuals were less likely to support any of the six reasons favouring disclosure (OR 0.3 to 0.5, $p<0.01$; table 3).

Individuals with five or fewer years in practice were twice as likely to be dissenters (OR 2.0, $p<0.01$), and those who were more confident to explain various aspects of SCD were 20% more likely to be dissenters (OR 1.2, $p<0.01$). Furthermore, those who agreed that newborn screening should be mandatory were half as likely to be dissenters (OR 0.6, $p<0.01$). Finally, compared with obstetric/postpartum nurses, family physicians and midwives, genetics professionals were 1.5 to 7 times more likely to be dissenters (OR 1.5, 2.0 to 7.8, respectively, $p<0.01$; table 4).

Table 4 What explains dissenting individuals? Bivariate analysis

	Dissenters	Non-dissenters	Unadjusted OR (95% CI)
Practice setting			
Academic	104 (26.9)	292 (24.9)	1.1 (0.85 to 1.44)
Non-academic	283 (73.1)	881 (75.1)	1.0
Method of reimbursement			
Fee for service	116 (30.5)	401 (35.3)	0.80 (0.63 to .03)
Non-fee for service	264 (69.5)	734 (64.7)	1.0
Location			
Metropolitan city/suburb	256 (66.1)	807 (68.8)	0.88 (0.69 to 1.12)
Small town/rural	131 (33.9)	366 (31.2)	1.0
Gender			
Female	292 (75.5)	849 (72.3)	1.18 (0.91 to 1.54)
Male	95 (24.5)	326 (27.7)	1.0
Years in practice			
0–5	126 (33.5)	229 (19.8)	2.04 (1.58 to 2.66)*
6+	250 (66.5)	928 (80.2)	1.0
Familiarity/confidence in newborn screening			
Up to date on conditions included in Ontario's newborn screening programme			
A/SA	154 (41.0)	431 (37.8)	1.14 (0.90 to 1.45)
N/D/SD	222 (59.0)	708 (62.2)	1.0
Confident in ability to explain newborn screening to parents			
A/SA	177 (47.1)	481 (42.3)	1.21 (0.96 to 1.53)
N/D/SD	199 (52.9)	656 (57.7)	1.0
Involvement in related care			
Involved in prenatal care or care of newborns within first days of life			
Involved	324 (82.9)	936 (79.2)	1.27 (0.94 to 1.71)
Not involved	67 (17.1)	246 (20.8)	1.0
Involved in care of families in newborn period			
Involved	315 (80.6)	912 (77.2)	1.23 (0.92 to 1.63)
Not involved	76 (19.4)	270 (22.8)	1.0
HCP group			
RN (reference group)	92 (19.5)	381 (80.5)	1.0
OB	41 (30.8)	165 (30.2)	1.03 (0.68 to 1.55)
MW	81 (46.8)	166 (30.3)	2.02 (1.42 to 2.87)*
FP	75 (44.9)	211 (35.6)	1.47 (1.04 to 2.09)†
PED	60 (39.5)	208 (35.3)	1.20 (0.83 to 1.72)
HEM	8 (8.0)	33 (8.0)	1.00 (0.45 to 2.25)
GEN	34 (27.0)	18 (4.5)	7.82 (4.23 to 14.46)*
Involvement in SCD care			
Ever see patients with SCD			
VF/F/S/R	279 (73.6)	860 (74.8)	0.94 (0.72 to 1.22)
Never	100 (26.4)	289 (25.2)	1.0
Ever provide SCD carrier status results to individuals and families			
VF/F S/R	214 (56.5)	621 (54.1)	1.10 (0.87 to 1.39)
Never	165 (43.5)	526 (45.9)	1.0
Ever inform carriers of SCD about reproductive risks of carrier status			
VF/F S/R	216 (57.1)	591 (51.6)	1.25 (0.99 to 1.58)
Never	162 (42.9)	554 (48.4)	1.0
Confident to explain ….			
Clinical significance of SCD			
A/SA	232 (61.4)	624 (54.5)	1.32 (1.04 to 1.68)†
N/D/SD	146 (38.6)	520 (45.5)	1.0
Meaning of SCD carrier status			
A/SA	246 (65.3)	681 (59.5)	1.28 (1.00 to 1.63)†
N/D/SD	131 (34.7)	464 (40.5)	1.0
Reproductive risks of SCD carrier status			
A/SA	214 (56.6)	570 (49.8)	1.31 (1.04 to 1.66)†
N/D/SD	164 (43.4)	575 (50.2)	

Continued

Table 4 Continued

	Dissenters	Non-dissenters	Unadjusted OR (95% CI)
When result should be provided			
To parents as soon as available			
A/SA	290 (75.1)	1067 (90.7)	0.31 (0.23 to 042)*
N/D/SD	96 (24.9)	109 (9.3)	1.0
To parents at a later date			
A/SA	47 (12.2)	95 (8.1)	1.57 (1.08 to 2.27)†
N/D/SD	338 (87.8)	1071 (91.9)	1.0
To parents only when they request it			
A/SA	76 (19.7)	53 (4.5)	5.17 (3.56 to 7.50)*
N/D/SD	309 (80.3)	1113 (95.5)	1.0
To child when s/he is in a position to request the information			
A/SA	194 (64.0)	382 (32.9)	2.08 (1.65 to 2.63)*
N/D/SD	109 (36.0)	779 (67.1)	
How provide newborn screening			
Newborn screening should be mandatory			
A/SA	207 (53.4)	784 (66.5)	0.58 (0.46 to 0.73)*
N/D/SD	181 (46.6)	395 (33.5)	1.0

Values are number (%).
*$p<0.01$; †$p \leqslant 0.05$. FP, family physicians; GEN, genetics professionals; HCP, healthcare professional; HEM, haematologists; MW, midwives; OB, obstetricians; PED, paediatricians; RN, registered nurses; SCD, sickle cell disorder. VF, very frequently; F, frequently; S, sometimes; R, rarely; A, agree; SA, strongly agree; N, neutral; D, disagree; SD, strongly disagree.

Dissenting group: survey results

In the descriptive data and in our analysis of dissenting individuals, genetics professionals stood out as a dissenting group. Furthermore, respondents who agreed with any of the six reasons opposing disclosure were more likely to be genetics professionals than other provider groups (OR 3.5 to 15.9, $p<0.01$). In addition, with the sole exception of the importance of disclosure for identifying the parents' reproductive risks, those who agreed with the reasons supporting carrier result disclosure were less likely to be genetics professionals than other provider groups (OR 0.1 to 0.6, $p<0.01$; table 5).

Dissent: qualitative findings

Few providers were definitive in rejecting a duty of disclosure. Rather, they questioned the assumption that this duty always applied. Some suggested a difference between screening and testing that might oblige a different policy for incidental findings without clinical benefits.

> "You've got two medical models. One would say that you should disclose what you know about an individual's status. The other would say that newborn screening disclosure is about disclosure of disease status for which an intervention is both available and effective…" (clinical geneticist, I 37)

For a small minority, an accepted duty of disclosure was superseded in the case of infant carrier results because of the harms that might ensue (eg, identification of misattributed paternity) or the child's independent future right to decide whether to learn this information. Furthermore, the relevant results had to be destroyed, ensuring that the "system" assume the responsibility for non-disclosure rather than the clinician.

> "Do you have a moral obligation to inform parents of carrier status if you have the information available?… Honestly I would say that they should be destroyed and a record of it kept. I think if the child wants to know, then the child can go and ask for testing. But keeping that in a doctor's file without the doctor having the consent or authority to communicate that with the patient, I think is just unacceptable. […] I think having access to information that you're not allowed to impart to your patient, it really goes against everything we stand for." (obstetrician, I 28)

Respondents entertained contradictory views of minority opinion and rights. Some providers interpreted the possibility that some might not wish to know as sufficient reason to disrupt a blanket policy of disclosure. Other providers who supported disclosure were explicit that minority opinion should not be permitted to trump majority needs:

> "I think we tend to focus a lot on the one or two people who may not want, or may have an objection, but we tend not to focus on the 95% of people who actually want information." (clinical geneticist, I 36)

DISCUSSION

Our study was designed to gauge the views of key stakeholders regarding the appropriate way to manage carrier information that is generated incidentally as a result of newborn screening for SCD. Growing interest in involving stakeholders in public policy development—through information, consultation or more deliberative engagement—is motivated by the need to enhance both the quality and acceptability of ensuing decisions.[37–40] In the absence of consensus, how stakeholder perspectives should inform policy is not well explored.[41] This is especially salient in considering moral issues, in which a strictly proceduralist standard of fairness (ie, that fairly solicited majority opinion is fair) can be inadequate, and some substantive standard of adequacy (ie, that majority opinion meet some principle-based or epistemic standard of fairness) may be required.[42]

As we expected, the majority of respondents supported the reasons provided for disclosure and disagreed with reasons opposing disclosure. Respondents were most attracted to reasons that supported parents' rights to their child's information, and to being informed generally, but a minority was concerned about violating the child's right to decide about receipt of this information. Dissenters were more likely to

Table 5 Pattern of attitudes regarding disclosure of SCD carrier results among genetics professionals versus non-genetics professionals

	Genetics professionals	Non-genetics professionals	Unadjusted OR (95% CI)
Reasons to disclose SCD carrier results—parents should be provided with the information that their infant is a sickle cell carrier because…			
…of the importance for identifying future reproductive risks for carrier infant			
A/SA	28 (53.8)	1365 (89.3)	0.14 (0.08 to 0.25)*
N/D/SD	24 (46.2)	164 (10.7)	1.0
…of the importance for informing parents their own reproductive risks			
A/SA	41 (78.8)	1313 (85.9)	0.61 (0.31 to 1.21)
N/D/SD	11 (21.2)	216 (14.1)	1.0
…of the newborn screening programmes' responsibility to disclose information it generates			
A/SA	33 (63.5)	1312 (86.1)	0.28 (0.16 to 0.50)*
N/D/SD	19 (36.5)	211 (13.9)	1.0
…of parents' right to information that exists about their infant			
A/SA	35 (67.3)	1429 (93.5)	0.14 (0.08 to 0.26)*
N/D/SD	17 (32.7)	99 (6.5)	1.0
…it is important to avoid misleading parents who might believe nothing was found			
A/SA	37 (71.2)	1424 (93.4)	0.17 (0.09 to 0.33)*
N/D/SD	15 (28.8)	100 (6.6)	1.0
…of the importance of using infrastructure for reporting results that is already in place			
A/SA	18 (35.3)	991 (66.1)	0.28 (0.16 to 0.50)*
N/D/SD	33 (64.7)	509 (33.9)	1.0
Reasons not to disclose SCD carrier results—parents should not be provided with the information that their infant is a sickle cell carrier because it is important…			
…to avoid risk that parents may misunderstand the meaning of carrier status			
A/SA	8 (15.7)	57 (3.8)	4.78 (2.15 to 10.62)*
N/D/SD	43 (84.3)	1463 (96.3)	1.0
…to minimise provision of information that does not influence the medical management of the child			
A/SA	11 (21.2)	61 (4.0)	6.39 (3.13 to 13.03)*
N/D/SD	41 (78.8)	1452 (96.0)	1.0
…to avoid creating additional costs for healthcare system			
A/SA	14 (27.5)	84 (5.5)	6.45 (3.36 to 12.40)*
N/D/SD	37 (72.5)	1432 (94.5)	1.0
…that the child decide if and when they want this information			
A/SA	27 (51.9)	96 (6.4)	15.90 (8.88 to 28.45)*
N/D/SD	25 (48.1)	1413 (93.6)	1.0
…not to assume that people want to learn about their or their infant's carrier status			
A/SA	25 (48.1)	253 (16.8)	4.58 (2.62 to 8.03)*
N/D/SD	27 (51.9)	1252 (83.2)	1.0
…not to assume that people will want to learn about their or their infant's reproductive risks			
A/SA	22 (42.3)	259 (17.2)	3.52 (2.00 to 6.20)*
N/D/SD	30 (57.7)	1244 (82.8)	1.0

Values are number (%).
*$p<0.01$. A, agree; SA, strongly agree; N, neutral; D, disagree; SD, strongly disagree. OR, odds ratio; SCD, sickle cell disorder.

support providing carrier results to the child when s/he requested it. Qualitatively, the dominant support for disclosure appeared to be motivated by a perceived clinical duty to disclose information that belongs to others, as well as a reluctance to keep secrets or be exposed to legal risk. The duty to disclose was generally perceived to reside with the clinician who received the report, but some respondents identified a broader "system" responsibility to inform. Dissenters articulated a core difference between clinical testing and population screening contexts, suggesting that "clinician-style" obligations might not apply to population-screening contexts; they were also motivated by a desire to avoid the potential harms of disclosure.

The data generated are not equivocal about majority opinion: provider stakeholders clearly favoured routine disclosure. A fair inference is that it would be easier to enlist providers in a policy of routine disclosure of SCD carrier results than any other policy. Despite this, it does not follow necessarily that the majority opinion of key stakeholders should guide public policy,

nor that an easier implementation process should be preferred. Ultimately, what should be done cannot be derived from descriptive data. Nonetheless, a more developed analysis of both majority and minority opinion can clarify the range of moral interests to be considered. To that end, we explore three considerations that are relevant to the current case and may have wider applicability in the adjudication of stakeholder opinion: (1) whether majority interests might be restrained by minority interests; (2) the nature and significance of dissent; and (3) the relevance of ethical standpoint.

A first issue is whether majority opinion infringes any well-accepted minority interests. In the current case, many of the principles upheld by the majority are non-contentious. Nevertheless, the denial of the interest of a child in deciding on the receipt or disposition of non-health serving medical information conflicts with some normative guidance that supports these interests.[10 13 14] Some respondents argued that minority interests should not derail attention to majority preferences. Nonetheless, within the liberal political tradition, minority interests that do not unduly constrain majority interests warrant protection.

A second issue is how dissenting opinion should be weighed and balanced. In the first instance, this concerns how dissent is to be calculated. What is to be made of the opinions of individuals who side with the majority on most issues but support a dissenting position on at least one other issue? Does a simple majoritarian calculus apply, or is any support for a dissenting reason sufficient to constitute reasonable doubt? If we accept any dissent as substantive, a group of ''dissenters'' can be defined that is consistently and measurably different from the majority, and whose dissent is partly structured by previous experiences and attitudes. Furthermore, though smaller in absolute size than the total dissent, a sizeable proportion of genetics professionals dissented. Arguably, this is not surprising, but reflects this group's familiarity with ethical and policy debates related to genetic screening. Kass[43] has argued that dissent that is concentrated in some morally salient way—for her, dissent that is concentrated in a specific ethnic group or geographical area—may require special consideration when seeking to implement a public health intervention. In this case also, the dissent of the ''dissenting group'', or of genetics professionals as a whole, is concentrated in ways that suggest a coherent and informed moral discourse that deserves careful consideration.

A third issue to consider is how clinical ethics should be considered in guiding public health policy initiatives. Providers emphasised a clinical ethic as a dominant imperative guiding their orientation to this issue: an obligation to share medical information with one's patients. A clinical ''duty'' to disclose is clearly both a meritorious and an understandable imperative. It suggests that policy makers would be unwise to ask clinicians to exercise a non-disclosure policy—by possessing such results but exercising programmatic control over their release (ie, only disclosing upon request, or when the child reaches a suitable age). Instead, should a non-disclosure policy be implemented, central programmatic control over all aspects of release would be required. However, it is not clear that clinical ethics are always consonant with public health ethics, or that an obligation grounded in clinical ethics should determine policy for a public health initiative.[44]

Dissent is inevitable in the face of any public health initiative and should not by its existence alone serve as a veto.[43] Yet in specific cases, as illustrated by the data reviewed here, there may be reasons for taking dissent seriously: when it identifies minority interests that have independent moral standing and that can be respected without undue burden, when it involves a rational moral discourse, or when it stems from an ethical standpoint that is not authoritative in the policy domain under consideration. Recognition of morally salient minority interests does not require that majority interests be overturned. Nonetheless, it does oblige decision-makers to look beyond majority opinion, and to discover ways in which both minority and majority interests might be served.

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