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Stakeholders in psychiatry and their attitudes toward receiving pertinent and incident findings in genomic research

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Increasingly more psychiatric research studies use whole genome sequencing or whole exome sequencing. Consequently, researchers face difficult questions, such as which genomic findings to return to research participants and how. This study aims to gain more knowledge on the attitudes among potential research participants and health professionals toward receiving pertinent and incidental findings. A cross-sectional online survey was developed to investigate the attitudes among research participants toward receiving genomic findings. A total of 2,637 stakeholders responded: 241 persons with mental disorders, 671 relatives, 1,623 blood donors, 74 psychiatrists, and 28 clinical geneticists. Stakeholders wanted both pertinent findings (95%) and incidental findings (91%) to be made available for research participants. The majority (77%) stated that researchers should not actively search for incidental findings. Persons with mental disorders and relatives were generally more positive about receiving any kind of findings than clinical geneticists and psychiatrists. Compared with blood donors, persons with mental disorders reported to be more positive about receiving raw genomic data and information that is not of serious health importance. Psychiatrists and clinical geneticists were less positive about receiving genomic findings compared with blood donors. The attitudes toward receiving findings were very positive. Stakeholders were willing to refrain from receiving incidental information if it could compromise the research. Our results suggest that research participants consider themselves as altruistic participants. This study offers valuable insight, which may inform future programs aiming to develop new strategies to target issues relating to the return of findings in genomic research.

KEYWORDS
attitude, genomics, incidental findings, mental disorders, surveys

1 | INTRODUCTION

Increasingly more research studies are based on genome sequencing (whole genome sequencing or whole exome sequencing). Researchers consequently face difficult questions, such as which genomic results to return to research participants and how. In recent years, a debate has evolved as to whether pertinent and incidental findings should be returned and whether the genetic researcher has a duty to inform about

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individual genetic research results (Christenhusz, Devriendt, & Dierickx, 2013; McGuire & Lupski, 2010; Parens, Appelbaum, & Chung, 2013; Ryan, De Vries, Uhlmann, Roberts, & Gornick, 2017; Solberg & Steinsbekk, 2012; Steinsbekk & Solberg, 2012; Viberg, Segerdahl, Langenskiöld, & Hansson, 2015; Wolf, 2012).

Genomic research potentially provides clinical information about the current and future health of the research participant, and a result with no interest for the research study could still be important for the research participant. The standard practice in genetic research has been not to give participants access to their personal sequencing data. The research participants are seen as altruistic individuals, who do not expect to derive any personal health benefits from participating, and researchers do not analyze the entire genome for all possible risk variants for any disorder (Kaye et al., 2014; McCann, Campbell, & Entwistle, 2010). Rather, researchers restrict their analyze to the variants associated with the disorder that is the focus of their study.

The American College of Medical Genetics and Genomics (ACMG) recommends that 59 specified pathogenic mutations (incidental findings) should be searched for each time a clinical genome sequencing is done, regardless of the indication for ordering the clinical sequencing (ACMG Board of Directors, 2015; Green et al., 2013; Kalia et al., 2017). In the research setting, the same variants may also be identified and the ACMG's recommendations are thus relevant to discuss among researchers because this specific genomic information is medically actionable. However, when sequencing is considered in the research setting, researchers have expressed concern that the delivery of results at the individual level may risk to divert resources, slow down the science, place a considerable burden on researchers, and yet may have only limited benefit to the research participants (Gliwa & Berkman, 2013; Wolf, 2012).

In the last few years, a strong interest has been taken in the identification of the causes of mental disorders by studying genetic factors using sequencing approaches (Merico et al., 2015; Yu et al., 2013). Great hopes are attached to these technologies that may help identify rare variants related to development of mental disorders and introduce new, more effective methods of medical treatment (Cirulli & Goldstein, 2010; Rabbani, Tekin, & Mahdieh, 2014).

Although the attitudes toward the return of pertinent and incidental findings vary among experts, it is relevant to discuss the attitudes among potential research participants and health professionals by exploring their views on the use of genomic research and their expectations to forming part of genomic research. It is especially important to explore the attitudes among relevant stakeholders in psychiatric genomic research as individuals with mental disorders are a vulnerable group, who may be subjected to stigmatization from genomic research. Yet, at the same time, they also have the potential benefit that discovering genetic causes of mental disorder may alleviate self-blame and shame (Phelan, 2002).

Several previous studies have focused on the attitudes toward the return of pertinent and incidental findings (Bui, Anderson, Kassem, & McMahon, 2014; Klitzman et al., 2013; Middleton et al., 2016; Yu, Harrell, Jamal, Tabor, & Bamshad, 2014). Yet, to our knowledge, no larger studies have investigated the attitudes of patients, relatives, and health professionals in psychiatry and genetics toward the return of pertinent and incidental findings in genome sequencing in the research setting.

Therefore, we designed a study to explore the attitudes among five different groups of such stakeholders: persons with mental disorders, relatives, healthy individuals (blood donors), psychiatrists, and clinical geneticists. In this article, we analyze attitudes toward the sharing of raw genomic data, pertinent and incidental findings, and the duty of the researchers to search for incidental findings.

2 | MATERIALS AND METHODS

2.1 | Qualitative interviews

Semi-structured interviews were conducted to explore the attitudes toward ethical issues regarding genome sequencing among health professionals engaged in genomic sequencing and among potential research participants. Six interviews were conducted with genetic researchers from Iceland and Denmark, including the Faroe Islands, three interviews were conducted with patients with schizophrenia, and four focus group interviews were conducted with clinical geneticists, relatives to individuals with ADHD, individuals with ADHD, blood donors from The Danish Blood Donor Study (Pedersen et al., 2012; The Danish Blood Donor Study, 2016).

All interviews were conducted from December 5, 2012 to December 11, 2013. Each interview lasted between 30 and 90 min. All interviews were audio-recorded, transcribed verbatim, and coded in NVivo (QSR International. Daresbury, Cheshire, United Kingdom, 2017).

2.2 | The survey

This study was carried out as a cross-sectional study based on survey data. The survey was developed to explore attitudes toward the use of genome sequencing in research. The survey was web-based (https://svaros.dk/holdning) and a modified version of the web-based survey (www.genomethics.org) developed at the Wellcome Trust Sanger Institute in Cambridge, United Kingdom (Middleton, Bragin, Morley, & Parker, 2013; Middleton, Bragin, & Parker, 2014; Middleton et al., 2015, 2016; Middleton, Parker, Wright, Bragin, & Hurles, 2013). The development process and validation techniques of the English survey are described elsewhere (Middleton, Bragin et al., 2013). Ten video films with subtitles and voice-over were used to explain the survey background and to illustrate the ethical issues relating to nine focus areas of the survey. The survey, the voice-over, and the subtitles for the video films were translated into Danish using the cross-cultural adaption process described by (Beaton, Bombardier, Guillemin, & Ferraz, 2000).

On the basis of the information in the qualitative interviews, we modified the Danish version of the survey to also include items about: 1) informed consent; 2) inclusion of children in genomic research; and 3) personal experience with mental disorders.

The final survey included a total of nine themes and focused on attitudes toward: 1) sharing pertinent findings; 2) sharing incidental

findings; 3) receiving incidental findings in different categories of severity and treatability; 4) sharing of raw genomic data; 5) the duty of genomic researchers to search for incidental findings; 6) risk perception; 7) filtering genomic data; 8) potential consenting procedures for genomic research studies; and 9) children in genomic research. The majority of items were responded to by dichotomous answers and a "Don't know" option. The survey also collected socio-demographic information. In addition, the survey requested information about personal and familial experience with mental disorders and about prior personal experience with participation in genetic research, genetic testing, or genomic sequencing.

A number of pilot tests of the survey were performed. After each pilot test, the survey was revised in accordance with the findings in the pilot testing. The first draft of the survey was pilot tested in a small sample of genetic researchers ($n = 11$) in April 2014. The second draft was tested in a group of blood donors from the Danish Blood Donor Study (Pedersen et al., 2012; The Danish Blood Donor Study, 2016) and participants in the qualitative interviews ($n = 54$) in May 2014. It took approximately 23 min to complete the final survey. Responses to the survey were anonymous.

2.3 | Recruitment of stakeholders

Stakeholders were identified and recruited: 1) persons with mental disorders (who are potential cases in psychiatric genomic research); 2) relatives to individuals with mental disorders (who are potential controls in psychiatric genomic research and also potential stakeholders who can benefit from both pertinent and incidental findings); 3) blood donors from the Danish Blood Donor Study (who are potential healthy controls in genomic research) (Pedersen et al., 2012; The Danish Blood Donor Study, 2016); 4) clinical geneticists (who in their clinical work conduct, analyze, return, and explain genomic data to patients and their relatives, and who may have to validate sequencing findings obtained in the research context); and 5) psychiatrists (who diagnose, treat, and care for people with mental disorders). As genomic analyze is incorporated in both research and clinical practise, it is imperative to explore the attitudes of clinical geneticists and psychiatrist as their attitudes may affect their advice to patients, relatives, and healthy controls.

Stakeholders were recruited via direct invitations by email, paper flyers at psychiatric hospitals, invitations posted in an ADHD Facebook group, and links at the homepages of the Danish Psychiatric Association, the Danish Society of Medical Genetics, and for user groups of psychiatric patients and their relatives. The data collection began on 12 August 2014 and ended on 17 April 2015.

2.4 | Ethical considerations

The project was approved by the Danish Data Protection Agency (file no. 2007-58-015). The Committee on Health Research Ethics in the Capital Region of Denmark reviewed the project description and concluded that the study did not require ethical approval (file no. H-4_2013_FSP-051). As all data are based on anonymous interviews and survey information, no other ethical clearance is required for this type of study according to Danish law.

2.5 | Statistical analyze

The analyze of the data was carried out using SAS® 9.3 (SAS Institute Inc., Cary, USA, 2017). Descriptive statistics were used to characterize the study sample. Unadjusted associations between items and stakeholder groups were estimated using χ^2 tests. Binary logistic regression models were used to estimate these associations adjusted for gender, age, level of education, marital status, and parenthood, with 99% confidence intervals (CI). A p-value of 0.01 was chosen to denote statistical significance and minimize Type 1 errors from multiple testing. Blood donors were chosen as reference group because they are healthy individuals and therefore potential healthy controls in genomic research. "Don't know" answers from respondents were omitted from the corresponding binary logistic regression models.

3 | RESULTS

3.1 | Sample characteristics

The distribution of socio-demographic characteristics is presented by stakeholder group in Table 1. The survey was completed by 2,637 individuals: 241 persons with mental disorders, 671 relatives, 1,623 blood donors, 74 psychiatrists, and 28 clinical geneticists. There were more females (53%) than males (47%), except among blood donors. As shown in Table 1, the majority of the stakeholders were aged 41–60 years and had a medium higher education (31%) or a long higher education (34%). The majority of the sample was married/cohabiting (67%) and had no children (53%).

3.2 | Attitudes toward receiving pertinent and incidental findings

Table 2 shows attitudes toward receiving pertinent and incidental findings. A majority (95%) of the stakeholders had a positive attitude toward the sharing of pertinent findings. A total of 91% wanted incidental findings from genomic studies to be made available to research participants, although psychiatrists (74%) and clinical geneticists (86%) were less likely to hold this view (Table 2).

In total, 77% of the stakeholders thought that genomic researchers should not actively search for incidental findings. Clinical geneticists (96%) and psychiatrists (91%) were significantly more negative than the three other groups. In total, 15% of the stakeholders supported that genomic researchers should actively search for incidental findings. Of the 15%, 38% agreed that genomic researchers should actively search for incidental findings, even if it would be expensive and time-consuming (Table 2).

TABLE 1 Distribution of socio-demographic characteristics for the five stakeholders groups

	Persons with mental disorders[a] (*n* = 241)	Relatives[a] (*n* = 671)	Blood donors[a] (*n* = 1,623)	Psychiatrists[a] (*n* = 74)	Clinical geneticists (*n* = 28)	Total[a] (*n* = 2,637)
	% (*n*)	% (*n*)	% (*n*)	% (*n*)	% (*n*)	% (*n*)
Gender						
Female	75 (180)	55 (372)	47 (769)	58 (42)	86 (24)	53 (1,387)
Male	25 (61)	45 (299)	53 (851)	42 (31)	14 (4)	47 (1,246)
Age groups						
20–30 years	13 (31)	15 (97)	11 (176)	3 (2)	0 (0)	12 (306)
31–40 years	20 (48)	21 (139)	20 (327)	33 (24)	25 (7)	21 (545)
41–50 years	27.5 (66)	25 (171)	27 (437)	16 (12)	28 (8)	26 (694)
51–60 years	27.5 (66)	26 (177)	26 (417)	20 (15)	36 (10)	26 (685)
61–70 years	11 (26)	12 (79)	15 (249)	27 (20)	11 (3)	14 (377)
71–76 years	1 (3)	1 (6)	1 (14)	1 (1)	0 (0)	1 (24)
Educational level						
None	5 (12)	2 (13)	2 (26)	0 (0)	0 (0)	2 (51)
One or more short courses	4 (10)	2 (16)	2 (25)	0 (0)	0 (0)	2 (51)
Skilled worker in craft, office, etc.	16 (39)	15 (102)	17 (276)	0 (0)	0 (0)	16 (417)
Short higher education <3 years	15 (35)	13 (86)	10 (170)	0 (0)	0 (0)	11 (291)
Medium higher education, 3–4 years	30 (72)	35 (232)	31 (502)	0 (0)	0 (0)	31 (806)
Long higher education, >4 years	24 (58)	29 (196)	34 (554)	100 (74)	100 (28)	34 (910)
Other education	6 (14)	4 (25)	4 (70)	0 (0)	0 (0)	4 (109)
Marital status						
Married/living together	57 (136)	62 (417)	70 (1,128)	79 (58)	82 (23)	67 (1,762)
Partnership	13 (30)	17 (116)	10 (161)	4 (3)	4 (1)	12 (311)
Divorced/separated	8 (20)	5 (34)	5 (79)	6 (4)	7 (2)	5 (139)
Widowed	2 (5)	1 (7)	1 (17)	3 (2)	0 (0)	1 (31)
Single	20 (49)	15 (97)	14 (236)	8 (6)	7 (2)	15 (390)
Children						
0	57 (138)	52 (348)	54 (871)	53 (39)	36 (10)	53 (1,406)
1	16 (38)	18 (123)	18 (293)	14 (10)	18 (5)	18 (469)
2	21 (51)	22 (146)	22 (362)	20 (15)	21 (6)	22 (580)
3	5 (13)	6 (43)	5 (82)	8 (6)	25 (7)	6 (151)
4+	1 (1)	2 (10)	1 (12)	5 (4)	0 (0)	1 (27)

[a]*n* varies because of missing data.

3.3 | Attitudes toward receiving different categories of findings

To gain a deeper understanding of attitudes toward receiving findings, we asked stakeholders which kind of findings they would be interested in receiving (Tables 3 and 4). Table 3 provides the unadjusted associations between statements and stakeholder groups, and Table 4 provides the adjusted and significant associations.

Most stakeholders (68%) did not want to receive all their raw genomic data. More clinical geneticists (85%) had this view (Table 3). Significant differences existed between the groups: persons with mental disorders were more likely to agree than blood donors (OR = 1.962) (Table 4).

In relation to the questions about receiving findings at different levels of severity and treatability of a disorder, a total of 54% of the stakeholders would prefer to receive information about

TABLE 2 Attitudes toward return of pertinent[a] and incidental[b] findings

	Persons with mental disorders[c] (*n* = 241)	Relatives[c] (*n* = 671)	Blood donors[c] (*n* = 1,623)	Psychiatrists (*n* = 74)	Clinical geneticists (*n* = 28)	Total[c] (*n* = 2,637)
	% (*n*)	% (*n*)	% (*n*)	% (*n*)	% (*n*)	% (*n*)
Do you think that pertinent findings from genomic studies should be made available to research participants?						
Yes	96 (230)	95 (638)	95 (1,545)	89 (66)	93 (26)	95 (2,505)
No	4 (9)	4 (23)	4 (61)	7 (5)	3 (1)	4 (99)
Do not know	1 (1)	1 (9)	1 (17)	4 (3)	3 (1)	1 (31)
$\chi^2 = 10.40$, df = 8, p-value = 0.24						
Do you think that incidental findings from genomic studies should be made available to research participants?						
Yes	95 (227)	91 (610)	91 (1,481)	74 (55)	86 (24)	91 (2,397)
No	3 (8)	6 (40)	7 (112)	20 (15)	7 (2)	7 (117)
Do not know	2 (5)	3 (20)	2 (28)	6 (4)	7 (2)	2 (59)
$\chi^2 = 37.57$, df = 8, p-value = < 0.0001						
Assuming research participants' consent, do you think that genomic researchers should actively search for incidental findings that are not relevant to the research study?						
Yes	18 (44)	16 (110)	14 (223)	5 (4)	4 (1)	15 (382)
No	71 (170)	75 (500)	78 (1,264)	91 (67)	96 (27)	77 (2,028)
Do not know	11 (25)	9 (61)	8 (135)	4 (3)	0 (0)	8 (224)
$\chi^2 = 21.77$, df = 8, p-value = 0.005						
Actively searching for incidental findings that are not relevant to the research study is likely to be very expensive and time-consuming. This may mean that the research is compromised. Given this caveat, do you still feel it is important for genomic researchers to actively search for incidental findings that are not relevant to their research study?						
Yes	55 (23)	41 (45)	31 (69)	100 (4)	100 (1)	38 (142)
No	36 (15)	39 (43)	45 (99)	0 (0)	0 (0)	41 (157)
Do not know	9 (4)	20 (22)	24 (53)	0 (0)	0 (0)	21 (79)
$\chi^2 = 19.03$, df = 8, p-value = 0.014						

[a]A result from a genetic test or genomic study which is directly relevant to the condition being investigated.
[b]A result from a genetic test or genomic study which is not directly related to the condition being explored.
[c]*n* varies because of missing data.

life-threatening conditions even if it cannot be prevented (Table 3). Clinical geneticists were significant less likely to hold this view than blood donors (OR = 0.312) (Table 4). A large majority of the stakeholders (97%) were interested in receiving findings relating to preventable life-threatening conditions (Table 3).

Fewer though, and still the majority (61%), would like to receive information about a serious but not life-threatening condition that cannot be prevented (Table 3). Clinical geneticists (37%) and psychiatrists (39%) were less interested in receiving information about this type of condition (Table 3). When we explored these results by stakeholder group, psychiatrists were significant less likely than blood donors to be interested in receiving findings concerning a serious unpreventable condition (OR = 0.43) (Table 4). A total of 96% would prefer to receive information about serious preventable conditions (Table 3).

As Table 3 shows, 68% of our stakeholders agreed with the survey statement "I would like to receive information that is not immediately relevant but could be useful later in life." Psychiatrists were less likely than blood donors to be interested in receiving not immediately relevant information (OR = 0.46) (Table 4). In total, 64% of the stakeholders would prefer not to receive "information that is uncertain and cannot be interpreted at the moment" (Table 3).

When asked about receiving information that is not likely to be of serious health importance, half of the stakeholders (50%) indicated that they would not like to receive this information (Table 3). Persons with mental disorders tended more likely to agree with this statement than blood donors (OR = 1.51) (Table 4).

A total of 83% of the stakeholders would like to receive information that could predict how they might respond to different medications or drugs, and 87% preferred to know if they are carrier of a condition that could be relevant to their children (Table 3).

Finally, we asked about their attitude toward receiving information about ancestry: 74% of the persons with mental disorders, 72% of

TABLE 3 Attitudes toward receiving genomic information

	Persons with mental disorders[a] (*n* = 241)	Relatives[a] (*n* = 671)	Blood donors[a] (*n* = 1,623)	Psychiatrists[a] (*n* = 74)	Clinical geneticists[a] (*n* = 28)	Total[a] (*n* = 2,637)
	% (*n*)	% (*n*)	% (*n*)	% (*n*)	% (*n*)	% (*n*)
If you were a research participant in a whole genome study, would you want to be able to receive all of your raw genomic data?						
Yes	31 (75)	20 (136)	21 (341)	20 (15)	14 (4)	22 (571)
No	59 (141)	67 (450)	70 (1,134)	77 (57)	85 (24)	68 (1,806)
Do not know	10 (23)	13 (84)	9 (145)	3 (2)	0 (0)	10 (254)
χ^2 = 30.86, df = 8, *p*-value = **0.0001**						
I would like to know about life-threatening conditions that cannot be prevented						
Yes	60 (141)	53 (344)	55 (874)	41 (28)	32 (9)	54 (1,396)
No	24 (55)	28 (186)	27 (435)	39 (27)	61 (17)	28 (720)
Do not know	16 (37)	19 (121)	18 (277)	20 (14)	7 (2)	18 (451)
χ^2 = 25.40, df = 8, *p*-value = **0.001**						
I would like to know about life-threatening conditions that can be prevented						
Yes	99 (237)	97 (647)	97 (1,572)	93 (68)	96 (27)	97 (2,551)
No	0.4 (1)	2 (12)	2 (35)	6 (4)	4 (1)	2 (53)
Do not know	0.4 (1)	1 (10)	1 (13)	1 (1)	0 (0)	1 (25)
χ^2 = 11.84, df = 8, *p*-value = 0.16						
I would like to know about serious (but not life-threatening) conditions that cannot be prevented						
Yes	68 (158)	60 (398)	62 (997)	39 (28)	37 (10)	61 (1,591)
No	23 (53)	28 (182)	26 (417)	45 (32)	56 (15)	27 (699)
Do not know	9 (22)	12 (78)	12 (195)	16 (11)	7 (2)	12 (308)
χ^2 = 31.58, df = 8, *p*-value = **0.0001**						
I would like to know about serious (but not life-threatening) conditions that can be prevented						
Yes	98 (234)	96 (642)	96 (1,548)	89 (65)	96 (27)	96 (2,516)
No	2 (4)	3 (16)	3 (50)	8 (6)	4 (1)	3 (77)
Do not know	0.4 (1)	1 (8)	1 (18)	3 (2)	0 (0)	1 (29)
χ^2 = 12.68, df = 8, *p*-value = 0.12						
I would like to receive information that could predict how I might respond to different medications or drugs (e.g., cholesterol-lowering drugs, anti-depressants)						
Yes	87 (205)	82 (540)	83 (1,316)	82 (58)	85 (23)	83 (2,142)
No	8 (20)	11 (70)	11 (117)	14 (10)	11 (3)	11 (280)
Do not know	5 (12)	7 (44)	6 (95)	4 (3)	4 (1)	6 (155)
χ^2 = 4.02, df = 8, *p*-value = 0.86						
I would like to receive information that could tell me if I am a carrier of a condition that could be relevant to my children						
Yes	92 (219)	87 (581)	87 (1,414)	77 (57)	93 (26)	87 (2,297)
No	5 (12)	8 (51)	8 (127)	12 (9)	7 (2)	8 (201)
Do not know	3 (8)	6 (38)	5 (77)	11 (8)	0 (0)	5 (131)
χ^2 = 14.28, df = 8, *p*-value = 0.07						
I would like to receive information that is not immediately relevant but could be useful later in life (e.g., very late onset cancer or predisposition to strokes)						

(Continues)

TABLE 3 (Continued)

	Persons with mental disorders[a] (*n* = 241)	Relatives[a] (*n* = 671)	Blood donors[a] (*n* = 1,623)	Psychiatrists[a] (*n* = 74)	Clinical geneticists[a] (*n* = 28)	Total[a] (*n* = 2,637)
	% (*n*)	% (*n*)	% (*n*)	% (*n*)	% (*n*)	% (*n*)
Yes	69 (166)	66 (439)	69 (1,118)	53 (39)	59 (16)	68 (1,178)
No	18 (44)	24 (162)	23 (368)	36 (27)	26 (7)	23 (608)
Do not know	13 (30)	10 (65)	8 (134)	11 (8)	15 (134)	9 (241)
χ^2 = 17.98, df = 8, *p*-value = 0.021						
I would like to receive information that is uncertain and cannot be interpreted at the moment						
Yes	26 (63)	22 (147)	24 (382)	16 (12)	10 (3)	23 (607)
No	58 (138)	65 (427)	63 (1,028)	73 (54)	78 (22)	64 (1,669)
Do not know	16 (37)	13 (87)	13 (207)	11 (8)	12 (207)	13 (342)
χ^2 = 9.82, df = 8, *p*-value = 0.28						
I would like to receive information that is not likely to be of serious health importance (e.g., mild eyesight problems)						
Yes	52 (125)	40 (266)	43 (701)	28 (21)	21 (6)	43 (1,119)
No	42 (100)	51 (334)	50 (802)	65 (48)	79 (22)	50 (1,306)
Do not know	6 (14)	9 (62)	7 (112)	7 (5)	0 (0)	7 (193)
χ^2 = 30.77, df = 8, *p*-value = **0.0002**						
I would like to receive information that tell me about my ancestry						
Yes	74 (178)	72 (478)	69 (1,122)	51 (38)	39 (11)	70 (1,827)
No	17 (41)	19 (130)	23 (373)	37 (27)	54 (15)	22 (586)
Do not know	9 (21)	9 (59)	8 (123)	12 (9)	7 (2)	8 (214)
χ^2 = 35.63, df = 8, *p*-value = < **0.0001**						

Numbers in bold are significant at 1%.
[a]*n* varies because of missing data.

the relatives, 69% of the blood donors, 51% of the psychiatrists, and 39% of the clinical geneticists were interested in receiving this type of information (Table 3). Significant differences were found between stakeholder groups; psychiatrists (OR = 0.48) and clinical geneticists (OR = 0.28) were less likely than blood donors to believe that information of ancestry should be shared (Table 4).

4 | DISCUSSION

Through an online survey, we asked the stakeholders to imagine that they were participating in a whole genome sequencing research study and presented them to several hypothetical scenarios. Our analyze show that a majority in the stakeholder groups would like to receive health-related individual level findings, depending on the severity and treatability of the condition, information about drug response and carrier conditions that could be relevant to their children. The access to this kind of information may provide the stakeholders with a better understanding of their own health and make it easier to take preventive steps. The research participants thus see themselves as potential patients, who might benefit from the research. We also found that genomic information about ancestry was more important to persons with mental disorders, relatives, and blood donors than to clinical geneticists and psychiatrists. A previous study found that the primary motivation for accessing DNA ancestry test information was educational advancement, interest in genealogical research, and entertainment. Very few of the respondents who had experience with DNA ancestry testing indicated that medical research was a motivational factor for the test (Wagner & Weiss, 2012). For the health professionals in our study, the low interest in ancestry may reflect that they do not believe that this kind of information is sufficiently important for the health to be returned to the participants by the researchers.

Despite interest in findings, changes in health behavior are not likely to be realized. A Cochrane review by (Marteau et al., 2010) explored if risk estimates derived from a genetic test could motivate people to change their behavior. They found no evidence that receiving DNA-based test results motivated people to change their behavior (Marteau et al., 2010). Even if the stakeholders would like to receive individual results and believe that they will find the information useful, they may not actually act on it.

However, only 22% of the stakeholders were interested in receiving their own raw data, although persons with mental disorders

TABLE 4 Attitudes toward receiving genomic information with blood donors as reference group: adjusted[a] results

	OR	99% CI	*p*-value
If you were a research participant in a whole genome study, would you want to be able to receive all of your raw genomic data?			
Persons with mental disorders	1.96	1.28–3.00	**< 0.0001**
Relatives	1.04	0.76–1.41	0.75
Psychiatrists	0.85	0.38–1.90	0.61
Clinical geneticists	0.66	0.16–2.77	0.46
p-value = **0.001**			
I would like to know about life-threatening conditions that cannot be prevented.			
Persons with mental disorders	1.34	0.85–2.10	0.10
Relatives	0.94	0.70–1.25	0.56
Psychiatrists	0.54	0.25–1.13	0.032
Clinical geneticists	0.31	0.10–0.92	**0.006**
p-value = **0.004**			
I would like to know about serious (but not life-threatening) conditions that cannot be prevented.			
Persons with mental disorders	1.32	0.85–2.08	0.11
Relatives	0.94	0.71–1.25	0.58
Psychiatrists	0.43	0.21–0.88	**0.003**
Clinical geneticists	0.38	0.13–1.13	0.02
p-value = **0.002**			
I would like to receive information that is not immediately relevant but could be useful later in life. (e.g., very late onset cancer or predisposition to strokes)			
Persons with mental disorders	1.29	0.80–2.08	0.17
Relatives	0.90	0.68–1.21	0.37
Psychiatrists	0.46	0.23–0.93	**0.004**
Clinical geneticists	0.82	0.24–2.76	0.67
p-value = 0.02			
I would like to receive information that is not likely to be of serious health importance (e.g., mild eyesight problems)			
Persons with mental disorders	1.51	1.03–2.22	**0.01**
Relatives	0.94	0.73–1.21	0.52
Psychiatrists	0.53	0.26–1.10	0.03
Clinical geneticists	0.37	0.11–1.27	0.04
p-value = **0.001**			
I would like to receive information that tell me about my ancestry			
Persons with mental disorders	1.47	0.90–2.39	0.04
Relatives	1.26	0.93–1.71	0.05
Psychiatrists	0.48	0.24–0.97	**0.007**
Clinical geneticists	0.28	0.10–0.83	**0.002**
p-value = < **0.0001**			

Numbers in bold are significant at 1%.

[a]Adjusted for: gender, age, educational level, marital status, and parenthood.

took a more positive approach than blood donors. Middleton et al. (2015) found that most genomic researchers (68%), non-genetic health professionals (57%), and the general public (61%) were interested in receiving their own raw sequence data (Middleton et al., 2015). The raw sequencing data do not seem to have the same personal utility for the Danish stakeholder groups in our study compared with the non-Danish stakeholder groups in the study by Middleton et al (participants in the Middleton study were from 75 countries across the world) (Middleton et al., 2015). Our data suggest that the stakeholders perceive the raw genomic data as less interpretable and more complex than individual level results.

Previous studies have discussed whether researchers have specific responsibilities to communicate individual research results to research participants (Gliwa & Berkman, 2013; Knoppers, Joly, Simard, & Durocher, 2006). Our study shows that people would generally like to receive incidental findings, but they do not expect this information to be returned at all costs. This indicates that stakeholders can be characterized as altruistic participants who believe that the primary duty of the researchers is to do research and that their own duty is to help others. The stakeholders do not expect research to be compromised by the researchers' active search for incidental findings, even if such findings could be of interest to the research participant.

We found consistent differences in the attitudes between potential participants in research and the stakeholders who deal with genomic data in their clinical work, for example, explaining findings to patients and relatives or providing care for people with mental disorders. For potential participants in research, there may be a clinical motivation to participate as they may gain an opportunity to receive a diagnosis, help identify more effective methods of medical treatment, and obtain more clinical knowledge about their own health. A study by Jallinoja and Aro (2000) found that the individuals in the Finnish population with the highest level of knowledge of genetics were both more enthusiastic and yet more sceptical toward genetic testing than the individuals with the lowest level of knowledge. A study by (Laegsgaard, Kristensen, & Mors, 2009) showed that persons with an anxiety disorder had a higher knowledge score of mental illness and genetics than persons with depression. However, significantly more persons with depression than persons with anxiety have intentions toward undergoing psychiatric genetic testing. Because of the education and professional training of clinical geneticists and psychiatrists, they are much more likely to understand this type of data, but they are also least interested in receiving such information and appear to take a more conservative approach than persons with mental disorders. The health professionals must explain the findings to patients, but they must also provide help and support if the patients are unable to cope with the findings. The implication of this is that health professionals, particularly psychiatrists, must have knowledge of health-related areas outside their field of expertise. The less positive attitudes among health professionals could be caused by inability to cope with the workload and from lack of skills. Some health professionals are familiar with returning findings and may have more concerns about the clinical validity and utility of the genomic findings than patients do.

5 | LIMITATIONS

Stakeholders were recruited using an online advertisement. This required the individuals to have familiarity with computers, laptops, tablets, or smartphones, and this approach excluded anyone without those skills or such access. Advertisements were placed on websites, and participant ascertainment would thus tend to favor the more proactive and information-seeking stakeholders. Stakeholders were recruited through a diversity of sources, and all stakeholders volunteered. Therefore, it is likely that they are a more homogeneous group and more in favor of genomic research than others who did not respond to the survey.

The design of the online survey did not provide us with any details on the non-response rate. According to the Danish Health Data Authority, there were 36 clinical geneticists and 1,125 psychiatrists in total in Denmark as of December 31, 2013 (Sundhedsdatastyrelsen, 2016). Almost all clinical geneticists responded to the survey, whereas only very few psychiatrists responded. According to Galea and Tracy (2007) low interest in participating could arise if the potential participants are much more likely to take part in a study focusing on an issue salient to the participant's life. One hypothesis explaining the low response rate among psychiatrists could be that there is a relation between outcome of interest and likelihood to participate.

Another limitation of this study is that we included persons with different mental disorders as one category. Different mental disorders involve different levels of severity, and attitudes may differ depending on diagnosis. This topic remains to be investigated in future studies.

This study provides new insight into the attitudes among relevant stakeholder groups in psychiatric genomic research toward receiving genomic information in a hypothetical research scenario. The recruitment strategies meant that it would not be possible for the final sample to be representative. Although the study provides evidence about attitudes toward hypothetical scenarios and shows that the majority of the stakeholders reported to have a positive attitude toward receiving genomic information, it does not mean that they would want to receive the information in a real-life situation.

6 | CONCLUSION

We studied the relationship between five groups of selected stakeholders in psychiatry and their attitudes toward receiving genomic research findings. Overall, the attitudes toward the survey statements about receiving findings were very positive. Persons with mental disorders and relatives were generally more positive about receiving any kind of findings than clinical geneticist and psychiatrists. Compared with blood donors, persons with mental disorders reported to be more positive about receiving raw genomic data and information that is not of serious health importance. Psychiatrists and clinical geneticists were less positive about receiving genomic findings compared with blood donors. An important finding is the general support to the return of incidental findings to research participants. Additionally, the stakeholders did not expect researchers to actively search for incidental findings. Most importantly, stakeholders were generally willing to refrain from receiving personal information if it could compromise the research. Our results confirm the assumption that research participants consider themselves as altruistic participants.

This study offers valuable insights, which may inform future programs aiming to develop new strategies to target issues relating to the return of results in genomic research.

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CONFLICTS OF INTEREST

The authors declare to have no conflicts of interest.

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