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Deciding on cystic fibrosis carrier screening: three citizens' juries and an online survey

Paola Mosconi¹, Cinzia Colombo¹, Anna Roberto¹, Giulia Candiani², Maria Teresa Greco³, Roberto Satolli², Carlo Castellani^{4,5}

1 Laboratory for Medical Research and Consumers Involvement, Department of Public Health, IRCCS Istituto di Ricerche Farmacologiche Mario Negri, Milan, Italy

2 Zadig Agenzia di Editoria Scientifica, Milan, Italy

3 Laboratory of Methodology for Clinical Research, Department of Oncology, IRCCS Istituto di Ricerche Farmacologiche Mario Negri, Milan, Italy

4 Centro Fibrosi Cistica dell'Azienda Ospedaliera Interuniversitaria Integrata, Verona, Italy

5 Centro Fibrosi Cistica, Ospedale Gaslini, Genoa, Italy

Correspondence: Paola Mosconi, IRCCS Istituto di Ricerche Farmacologiche Mario Negri, Via G. La Masa 19, 20156 Milan, Italy, Tel: +39 (0) 2 39014503, Fax: +39 (0) 2 33200231, e-mail: paola.mosconi@marionegri.it

Background: Health technology assessment and ethical issues have to be dealt with in deciding on national carrier screening for cystic fibrosis (CF)—the most frequent severe autosomal recessive disease in Caucasian populations and several stakeholders need to be involved. A citizens' jury is one way to ask citizens to deliberate on controversial topics in the interests of a society. The aims of this project were to gather opinions about CF carrier screening through citizens' jury deliberations and to match them with the findings of a large online consultation survey open to the general population, people with CF and families and health professionals. **Methods:** Three citizens' juries and an online survey were asked: 'Should the Health Service organize screening of the population with the aim of identifying healthy people who may have children with CF?' The jurors had no medical background and no personal or family CF history. The survey was open to people with CF, families, and healthcare professionals. **Results:** Jurors and survey respondents were in favour of CF carrier screening, mainly considering the severity of CF, the value of informed reproductive choices and the equality of the screening. All the citizens' juries felt positively about the health service actively offer CF carrier screening to provide women and couples of reproductive age equal access and standardized information on the pros and cons. **Conclusion:** Considering the favourable attitude towards CF screening, the feasibility of CF screening, in terms of best setting, target age and healthcare professionals providing it, should be tested in a clinical trial.

Introduction

Cystic fibrosis (CF) is the most frequent severe autosomal recessive disease in Caucasian populations.¹ By far the commonest cause of mortality is chronic lung infection, progressive pulmonary function deterioration and parenchymal damage, leading to respiratory failure. CF may also affect the pancreas with exocrine and endocrine insufficiency, the liver with focal biliary cirrhosis, the vas deferens with obstructive azoospermia and the sweat glands with increased salt concentrations in sweat. Specialized care in dedicated centers and an aggressive therapeutic approach have changed the natural history of the disease and considerably improved survival expectancy. However, CF is still a life-limiting disease and patients' median age in Europe ranges from 4.8 to 22.6 years.² In Italy, the median age is 20.5 years² and estimated carrier frequency between 1 in 25 and 1 in 30.³ Quality of life is often seriously affected by chronic symptoms and by the burden of care, which includes daily chest physiotherapy sessions, use of pancreatic enzymes, multivitamins, inhaled antibiotics and, in advanced stages of disease, oxygen supplementation and non-invasive ventilation. CF is caused by defects in the synthesis or function of a protein called CF transmembrane regulator, and the gene coding for it has been known since 1989.⁴ Since its discovery more than 2000 sequence variations have been found,⁵ and solid evidence has been collected to tell apart those that actually cause

CF.⁶ Progress in molecular biology techniques has also produced genetic tests to detect promptly and relatively inexpensively common mutations in most parts of the world and ethnic groups affected by CF. The combination of these factors has made it possible to offer carrier testing not only to relatives of CF patients whose mutation has been detected as running in the family but also to individuals with no family history of CF and a generic probability of being heterozygotes.

An Italian study provides evidence on the impact of different policies regarding CF carrier screening. Since the early 1990s, CF carrier screening has been offered and conducted extensively only in the north-eastern regions, while in the west it has not been offered. The increase in the number of carriers screened over time was significantly related with the decrease in the incidence of CF births. These results show how reproductive attitudes can be influenced by the availability of a screening test.⁷ When a healthcare professional or the public health service offers screening, citizens are put in a position where they cannot avoid making a choice.⁸ They have to assess the screening offered and decide, and it is very hard to refuse it. Individual informed consent is therefore partial, as it is driven by an offer already available, decided by someone else (healthcare professional or public health service). Engaging the public to express their opinions on the pros and cons of screening could therefore be the way to obtain informed choices from the public's point of view, before offering or implementing screenings. This is also important as

the benefits and harms of screenings can be assessed from a public perspective, as it is impossible to know which person might benefit or be harmed by the screening.

In the case of CF carrier screening in Italy, fertile couples living in the north-eastern and western regions had only to respond yes or no to a healthcare offer, with no role in the decision about which offer they considered the best. In Italy health policy programs for CF carrier screening are rare, and the test is sporadically offered to couples planning a pregnancy or to pregnant women, on the basis of the healthcare professional's personal attitudes.

Scientific societies tend to take a positive attitude to CF carrier screening, with some clearly arguing that it should be offered to all individuals of reproductive age,^{9–11} and others delegating the decision to local health authorities.^{12,13} The latter have shown little interest in implementing wide-range programs, possibly partly because little information has been available about the level of acceptance by people with CF and society as a whole. Patient associations have rarely expressed a strong opinion, and the general public has been involved in the debate only occasionally and without targeted information.¹⁴

Complex logistic, ethical and health technology assessment issues have to be faced when deciding about CF carrier screening.^{13,15} Several stakeholders need to be involved and their views may be at odds or unvoiced. This is a matter of concern because screening is a medical intervention aimed at supposedly healthy people with consequences at the community and individual levels and entails ethics and value-based decisions at community level.

A citizens' jury is a method of deliberative democracy to consult citizens on controversial topics, where a group of lay people with different backgrounds, values and attitudes are given adequate and independent information and asked to deliberate in the interests of society. This method has already been applied to decisions about screening.¹⁶

The aims of this project were to gather opinions about CF carrier screening through citizens' jury deliberations and to match them with the findings of an open online survey.

Methods

The project was promoted by researchers, science communication experts and clinicians in partnership with lay people and patients; these promoters were in charge and shared all the phases of the project. The process was superintended by a multidisciplinary committee: genetics, general practice, reproductive medicine, counselling, population screening, representatives of advocate groups of patients and families, communication, laboratory medicine and healthcare organizations.

The citizens' juries

Verona, Pistoia and Palermo were the headquarters of the three juries, respectively, in the north, center and south of Italy, to cover different socio-economic conditions. The first, as a pilot study, was in Verona in 2012,¹⁷ and the next two were in 2014.

The main question 'Should the Health Service organize screening of the population with the aim of identifying healthy people who may have children with CF?' was accompanied by five sub-questions regarding information to be highlighted on CF and the genetic test, responsibility for the information and suggestions for research. The questions were defined by the promoters.

People with no personal or family history of CF were selected. A call for interest was launched to local cultural, assistance, sport and religious voluntary associations registered in each area. Each association was asked to suggest at least two members willing to participate. The jurors were balanced for sex, age and education.

According to the citizen juries' method,¹⁸ the jurors have to be informed in a complete and balanced way and facilitated in a discussion meeting in order to reach a deliberation. The information

provided was covered by a booklet and a group of experts spoke during the jury meeting. The booklet was drafted from the literature and documents from consumer and CF family-patients' websites. It was reviewed by the experts and by patients and consumers' representatives.¹⁷

Two weeks before, the meeting members were sent the 26-page booklet covering: what a citizen jury is, information on CF (incidence, prevalence, symptoms, variability of symptoms, severity and follow-up), daily life impact, what screening and carrier screening are (logistics, organization and economic evaluation), what the carrier screening test is (including specificity and sensitivity), information on the carrier screening test in relatives, in the general population, and a glossary. The contents were written in plain language and figures and tables summarized the concepts. Questions for knowledge self-assessment were provided.

Clinicians, general practitioners, experts in laboratory tests, gynecologists, epidemiologists dealing with screening and policy makers were invited as speakers. They were invited to prepare presentations in plain language and to deal with their topic in a balanced way.

The first pilot jury meeting in Verona lasted one day, and the other two lasted 2 days each. Experts presented evidence on the topics covered in the booklet. One session specifically explored issues related to CF carrier screening, showing videos of three people who had been tested. The meetings ended with a pros and cons debate. There was time for questions and discussion with experts after each session.

At the end of the meeting, a 3–4 h section behind closed doors was dedicated to discussion among the jurors. The process was assisted by a skilled psychologist as facilitator, the same for all three juries; she made particularly sure that every juror could freely express his/her opinion and she guaranteed everyone's equal share of time.

Pro and cons were collected from each juror. They were asked not to declare their preference until all the elements were listed, and then they discussed the points listed. As far as possible, the final decision was reached by consensus. Minority positions at the end of the jury meeting were reported in the final deliberative document. A preliminary draft of the conclusions and recommendations from jurors was prepared. Later, a final report was drafted by the facilitator and approved by all the jurors.

Each citizens' jury experience was assessed using two questionnaires: one before and one after the jury meeting. The first comprised 12 questions on: the CF citizens' jury method, information in the booklet, any additional information sought before the meeting. The second explored the level of appreciation of the speakers, the time dedicated to the jury discussion, the facilitator and the method itself.

Online survey

A survey aimed at the general population, people with CF, families, and health professionals was organized and published on the PartecipaSalute ('Participate in healthcare') website¹⁹ from February to June 2015. Contact details were retrieved from databases of promoters, the Italian CF Foundation, CF patient associations and professional societies. Target groups were also informed through e-mails and articles published on websites, lay journals, bulletins and newsletters. People entering the survey were shown a brief presentation and then directed to 'General population' or 'Health professionals'. The information material aimed at jurors was the basis for the slides provided online: eight slides for the general population and six for health professionals, covering a general presentation and the main question, information on CF, the carrier test, screening and a list of pros and cons.

At the end, six questions were shown on socio-demographic characteristics and direct experience with CF, followed by the main question. Health professionals responding affirmatively had three

more questions on the target of the screening and organizational aspects. Space was available for comments.

Due to the nature of this research, no ethic committee approval was required, by law.

Results

The call for interest reached 891 (Verona), 281 (Pistoia), 411 (Palermo) voluntary associations. Respectively, 17, 13 and 23 expressed interest and suggested volunteers as potential jurors. Table 1 describes the selection and characteristics of the jurors. The mean age ranged from 49 to 60 years; except in Pistoia the male/female ratio was close to 1:1; in general, there were more people with higher education.

In Verona, all jurors expect one, in Pistoia unanimously, and in Palermo a majority of 9–7 were in favour of national CF carrier screening. Table 2 summarizes the human, scientific, economic and social reasons. The literally translated documents of Pistoia and Palermo citizens' juries are reported in Supplementary table S1, and the pilot jury (Verona) document is published elsewhere.¹⁷ The answers to the five sub-questions (Supplementary table S2) indicated that the health service should provide broad, clear information about screening and CF, involving primary care physicians and, gynecologists, and organizing *ad hoc* information campaigns.

In the pre-jury assessment, most of the jurors considered the meeting long enough to reach a deliberation (33/47, 70%). The majority rated the information booklet good (29/47, 61%); most gave the right answer to three questions regarding the meaning of a positive (41/47, 87%) or negative (39/47, 83%) test, and the organ most affected by CF (44/47, 94%). Half the jurors collected additional information, mostly from Internet. At the end of the day, the jurors rated the experience as good (36/43, 83%) and the presentations of the speakers as good (34/43, 79%); five jurors considered the closed doors discussion time too short.

In all, 904 responders completed the online survey; most were women among the general population (753/904; 73%) and among healthcare professionals (233/338; 69%), with mean age, respectively, 44.2 years and 47.9 years. Most of the general population had a high level of education.

The majority of respondents were in favour of a national CF carrier screening program. The general population was more often in favour than physicians; non-physicians health professionals were closer to the general population (table 3).

People with cases of CF or CF carriers in their families or among friends were more often in favour of the screening (table 4).

Discussion

The citizens' juries opted in favour of CF carrier screening in all three areas of Italy, supporting the generalizability of this deliberation. The deliberations were confirmed by the online survey. The main motivations, based on the jurors' deliberations and some comments in the online survey, were the good acceptance of the test (considered easy to do and with high sensitivity), the severity of CF and its impact on the quality of life. Jurors did not express great concern about the possibility of a 'eugenics' effect.

Each juror had the chance to address ethical stands while discussing the pros and cons of the screening. The deliberations stressed the value of informed reproductive choices and equity of access to the test and to the information on its availability, which would be guaranteed by a national screening program, an issue well addressed in the literature.²⁰ The balance of benefits and costs, including direct and indirect costs of the disease compared with the cost of nation-wide CF carrier screening, was judged positive, with differences due to the perception of local organizational difficulties. Palermo's jurors had a negative opinion about the

Table 1 Selection and characteristics of jurors

	Verona N (%)	Pistoia N (%)	Palermo N (%)
Call for interest in the project			
Voluntary associations contacted	891	281	411
Voluntary associations suggesting members as jurors	17	13	23
Selection of jurors			
People interested in participating as jurors	24	20	34
People selected	16 (67)	16 (80)	17 (50)
Characteristics of jurors			
14 ^a	14 ^a	16 ^a	
Sex			
Male	8 (57)	3 (21)	8 (50)
Female	6 (43)	11 (79)	8 (50)
Age (years) mean (range)			
30–44	2 (14)	1 (7)	6 (37)
45–54	5 (36)	3 (21)	5 (31)
55–65	7 (50)	6 (43)	4 (25)
66–81	0	4 (29)	1 (6)
Education			
Primary school	0	0	0
Middle school	2 (14)	3 (21)	1 (6)
High school	6 (43)	5 (36)	7 (43)
University	6 (43)	5 (36)	8 (50)
Missing	0	1 (7)	0

a: Two, two and one people did not participate in the final deliberation for personal reasons (many could not attend in the afternoon dedicated to the deliberation).

Regional Health Service's ability to organize screening programs, and those who voted against the screening were only sceptical about its feasibility, but otherwise they were in favour. This is of interest to policymakers and for health technology evaluations, suggesting that trust in the ability of healthcare services to implement the screening plays a key role.

Concerns about the balance of benefits and costs of CF carrier screening were mentioned in the online survey mainly by physicians not in favour. This could help explain the lower percentage of positive attitudes toward CF carrier screening in this professional group. It was noted that a major investment would be necessary to organize a CF carrier screening program. The initial annual cost is around \$2.5 million²⁰ so, despite the savings to the health system, the funding could be an important barrier.

The online survey suggested that being a relative of a person with CF produced a positive attitude toward the screening. Studies focused on couples with a family history of CF reported more than 80% of participants in favour of the population screening,²¹ while a recent review noted that an important obstacle to screening was the wrong perception of the risk of disease for people without a family history of CF.²² These results support a family history of CF as an exclusion criterion from the citizens' juries and the addition of an extra day to the jury meeting. CF is a rare disease and the general population is not aware of its characteristics, as shown in a recent Italian survey.¹⁴

The present study has some limitations. Due to a lack of resources, in Verona, the meeting lasted only 1 day, limiting discussion with the experts, while the other jury meetings lasted 2 days. This might make the experience not fully comparable. However, the topics covered during the jury meeting were the same, though in Pistoia and Palermo more time was dedicated to the population screening and the cost issues. In designing the project, the promoters did not consider the possible influence of local health systems' efficiency and reputation on the jurors' attitudes towards screening. Neither did they assess the jurors' opinion on the use of screening in general.

Regarding the citizens' jury method, there are some issues related to the representativeness of jurors, tied to a potential selection bias

Table 2 Synopsis of the three final documents

Verona	Pistoia	Palermo
Human reasons		
The severity of CF and the impact on quality of life and family life	The severity of CF and the impact on quality of life and family life	The need for the health service to do more to inform the public about CF and enable individuals/couples to make informed reproductive choices
Low life expectancy	Awareness of being at risk	More awareness of genetics as a consequence of CF carrier screening
CF carrier screening as a tool to avoid considerable suffering for many children and their families	Screening as a tool to convey more information for more informed reproduction. The stress of being found positive in the test should be balanced by information and psychological support	Screening is an effective tool to help reduce the incidence of the disease, with considerable advantages in human terms—avoidance of suffering to families and potential patients
Scientific reasons		
The availability of a test, with 85% sensitivity, and the results from previous experience	The availability of a simple blood test, with good sensitivity (85%). The small risk of false negatives CF carrier screening as a model for other inheritable diseases	The limits of the test were discussed, and its characteristics (85% sensitivity) were considered good
Economic reasons		
The balance between future expected decreases in the cost of the test and future increases in the cost of care	The balance between expected decreases in the cost of the test and expected increases in the cost of care	Positive cost-benefit balance considering both direct and indirect costs of the disease and the increasing cost as new treatments become available. Nation-wide screening would also lower the cost of the test
Social fairness		
Screening would enable participants to make better-informed reproductive choices, without affecting their freedom of choice	Screening would guarantee uniformity among the Italian regions and therefore greater fairness	Screening the population ensures greater social justice throughout the country, whereas the active offer campaign might deprive the citizens of some regions and the less-well-off.

Table 3 Online survey responses to the main question

	General population (566)	Health professionals non-physicians (169)	Health professionals physicians (169)
<i>'Should the Health Service organize screening of the population with the aim of identifying healthy people who may have children with CF?'</i>			
Yes—N (%)	488 (86)	126 (75)	87 (51)
No—N (%)	78 (14)	43 (25)	82 (49)

Table 4 Respondents in favour of CF carrier screening depending on their closeness to CF, i.e. CF cases or carriers in family

	In favour of CF carrier screening
General population	86% (488/566)
No CF in family and I don't know	83% (250/301) ^a
Yes CF in family and Yes carrier	91% (204/225) ^a
Health professionals non-physicians	75% (126/169)
No CF in family and I don't know	72% (111/154) ^a
Yes CF in family and Yes carrier	100% (10/10) ^a
Health professionals physicians	51% (87/169)
No CF in family and I don't know	51% (83/164)
Yes CF in family and Yes carrier	80% (4/5)

a: Differences in total numbers in each group are due to people in the group 'No CF in family and Yes carrier' or 'Yes CF in family and No carrier'.

due to the sources used to invite people, the characteristics of people deciding to participate and the small number of participants. These are common features of this method and have to be assessed considering that people must be willing to deliberate for the community,

and represent no individual or stakeholders interest.²³ Within the citizens' juries we organized, the jurors were asked to express a view point bearing in mind the community.

In the online survey there may have been some selection bias due to the internet tool used, depending on web access, digital health literacy, and the way people were invited. In Italy, 57% of people used Internet in 2014,²⁴ and the sample of the general population responding had a high level of education, as is common to online surveys. Furthermore, polls or consultations inevitably risk collecting only superficial opinions since they do not require a thorough understanding of the issue. Nevertheless, we believe this survey provides a broad and interesting insight into public perceptions and opinions on CF screening.

Before opting for the organization of national CF carrier screening, public health authorities can promote participatory action such as citizens' juries, which appear to produce statements in the societal perspective. This kind of deliberative consultation depicts the citizens' outlook in an affordable way and can make a valuable contribution to decision-making on public health matters, particularly on complex and/or controversial issues,^{23,25} such as CF carrier screening.

This project combined a deliberative method where CF carrier screening was thoroughly discussed in an *ad hoc* informed group with an online survey, with widely disseminated summary information. The broad range of views supports CF carrier screening. Considering the uncertainties and the controversial positions among scientific societies,^{10–13} a clinical trial could now be organized to test targets, organizational aspects, and methods of informing the public in order to find the best way to provide CF carrier screening, drawing on the preliminary information about different models: prenatal carrier screening, pre-conception carrier screening, carrier screening outside the clinic (workplaces, or schools).²⁰ Factors and barriers determining the intention to participate and implementation of the screening should also be carefully assessed.²⁶ In the meantime, public health authorities could plan to offer the test, promoting evidence-based information through

medical education programs, to ensure that balanced communication reaches women and couples of reproductive age, and monitoring the process and costs throughout the country, to guarantee equal access.

Supplementary data

Supplementary data are available at *EURPUB* online.

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Conflicts of interest: None declared.

Key points

- Three Citizens' Juries and an online survey opted in favour of t CF carrier screening. This provides a contribution and an example for decision-making on public health matters.
- CF carrier screening is a matter of concern because screening is a medical intervention aimed at supposedly healthy people with consequences at the community and individual levels and entail ethics and value-based decisions at community level.
- Involving citizens in health debates is very important, particularly in decisions about health such as CF carrier screening, which cannot be taken without consulting the general population for its preferences and values. A citizens' jury is a method of deliberative democracy to consult citizens on controversial topics.
- CF carrier screening is an important public health issue. Various scientific societies have produced statements about this screening, but the offer of carrier screening at a population level should not be decided by local health authorities only, without consulting citizens' preferences.

References

- Spoonhower KA, Davis PB. Epidemiology of cystic fibrosis. *Clin Chest Med* 2016;37:1–8.
- ECFSRP Annual Report 2015. 2017, https://www.ecfs.eu/sites/default/files/general-content-images/working-groups/ecfs-patient-registry/ECFSRP_Report2015_Nov2017.pdf (15 December 2017, date last accessed).
- Castellani C, Picci L, Tridello G, et al. Cystic fibrosis carrier screening effects on birth prevalence and newborn screening. *Genet Med* 2016;18:145–51.
- Rommens JM, Iannuzzi MC, Kerem B, et al. Identification of the cystic fibrosis gene: chromosome walking and jumping. *Science* 1989;245:1059–65.
- Cystic Fibrosis Mutation Database. <http://www.genet.sickkids.on.ca/app> (15 December 2017, date last accessed).
- Sosnay PR, Siklosi KR, Van Goor F, et al. Defining the disease liability of variants in the cystic fibrosis transmembrane conductance regulator gene. *Nat Genet* 2013;45:1160–7.
- Castellani C, Picci L, Tamanini A, et al. Association between carrier screening and incidence of cystic fibrosis. *JAMA* 2009;302:2573–9.
- Johansson M, Jorgensen KJ, Getz L, Moynihan R. 'Informed choice' in a time of too much medicine—no panacea for ethical difficulties. *BMJ* 2016;353:i2230.
- Genetic testing for cystic fibrosis. National Institutes of Health Consensus Development Conference Statement on genetic testing for cystic fibrosis. *Arch Intern Med* 1999;159:1529–39.
- ACOG Committee Opinion No. 486: update on carrier screening for cystic fibrosis. *Obstet Gynecol* 2011;117:1028–31.
- Grody WW, Thompson BH, Gregg AR, et al. ACMG position statement on prenatal/preconception expanded carrier screening. *Genet Med* 2013;15:482–3.
- Società Italiana Fibrosi Cistica: Raccomandazioni sul test del portatore di mutazioni del gene CFTR. https://www.sifc.it/sites/default/files/documento_cftr.pdf (15 December 2017, date last accessed).
- Castellani C, Macek M, Cassiman J-J, et al. Benchmarks for cystic fibrosis carrier screening: a European consensus document. *J Cyst Fibros* 2010;9:165–78.
- Braido F, Baiardini I, Sumberesi M, et al. Public awareness on cystic fibrosis: results from a national pragmatic survey. *Eur Respir J* 2015;46:264–7.
- Metcalfe SA. Carrier screening in preconception consultation in primary care. *J Commun Genet* 2012;3:193–203.
- Rychetnik L, Carter SM, Abelson J, et al. Enhancing citizen engagement in cancer screening through deliberative democracy. *J Natl Cancer Inst* 2013;105:380–6.
- Mosconi P, Castellani C, Villani W, Satolli R. Cystic fibrosis: to screen or not to screen? Involving a Citizens' jury in decisions on screening carrier. *Health Expect* 2015;18:1956–67.
- The Jefferson Center. The Jefferson Center. Citizens jury Handbook. (2004). http://www.rachel.org/files/document/Citizens_Jury_Handbook.pdf (15 December 2017, date last accessed).
- Mosconi P, Colombo C, Satolli R, Liberati A. PartecipaSalute, an Italian project to involve lay people, patients' associations and scientific-medical representatives in the health debate. *Health Expect* 2007;10:194–204.
- Maxwell S, Brameld K, Youngs L, et al. Informing policy for the Australian context—costs, outcomes and cost savings of prenatal carrier screening for cystic fibrosis. *Aust. N Z J Obstet Gynaecol* 2010;50:51–9.
- Janssens S, Chokoshvili D, Binst C, et al. Attitudes of cystic fibrosis patients and parents toward carrier screening and related reproductive issues. *Eur J Hum Genet* 2016;24:506–12.
- Massie J, Ioannou L, Delatycki M. Prenatal and preconception population carrier screening for cystic fibrosis in Australia: where are we up to? *Aust N Z J Obstet Gynaecol* 2014;54:503–9.
- Mosconi P, Colombo C, Satolli R, Carzaniga S. Involving a citizens' jury in decisions on individual screening for prostate cancer. *PLoS One* 2016;11:e0143176.
- Italian National Institute of Statistics-ISTAT. Cittadini e nuove tecnologie. Report. 2014. <https://www.istat.it/it/archivio/143073> (15 December 2017, date last accessed).
- Rychetnik L, Doust J, Thomas R, et al. A Community Jury on PSA screening: what do well-informed men want the government to do about prostate cancer screening—a qualitative analysis. *BMJ Open* 2014;4:e004682.
- Lakeman P, Plass AMC, Henneman L, et al. Preconceptional ancestry-based carrier couple screening for cystic fibrosis and haemoglobinopathies: what determines the intention to participate or not and actual participation? *Eur J Hum Genet* 2009;17:999–1009.