

*Title page***Title**

‘We did everything we could’ – A qualitative study exploring the acceptability of maternal-fetal surgery for spina bifida to parents

**Short title**

Parental acceptability of maternal-fetal surgery

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There are no conflicts to report.

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### **What is already known about this topic?**

- Maternal-fetal surgery for open spina bifida has the potential to improve long-term outcomes but at significant procedure-related fetal and maternal risk;
- Prospective parents receiving an antenatal diagnosis of open spina bifida, face a range of uncertainties regarding the future of their unborn child, and the options provided pose major ethical challenges.

### **What does this study add?**

- Despite significant impact, maternal-fetal surgery is highly acceptable for those parents who opt for it; Strong feelings of parental responsibility, direct them to do anything in their power to improve their future child's situation;
- In the small group of parents who opted for termination of pregnancy, maternal-fetal surgery was felt to offer insufficient certainty of substantial improvement in quality of life and the perceived severe impact of SB drove their decision to end the pregnancy.

**Data availability statement**

The data that support the findings of this study are available on request with reasonable motivation from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

## **Abstract**

### **Objective:**

To explore the concepts and strategies parents employ when considering maternal-fetal surgery (MFS) as an option for the management of spina bifida (SB) in their fetus, and how this determines the acceptability of the intervention.

### **Methods:**

A two-centre interview study enrolling parents whose fetuses with SB were eligible for MFS. To assess differences in acceptability, parents opting for MFS (n=24) were interviewed at three different moments in time: prior to the intervention, directly after the intervention and 3-6 months after birth. Parents opting for termination of pregnancy (n=5) were interviewed only once. Themes were identified and organised in line with the framework of acceptability.

### **Results:**

To parents opting for MFS, the intervention was perceived as an opportunity that needed to be taken. Feelings of parental responsibility drove them to do anything in their power to improve their future child's situation. Expectations seemed to be realistic yet were driven by hope for the best outcome. None expressed decisional regret at any stage, despite substantial impact and, at times, disappointing outcomes. For the small group of participants, who decided to opt for termination of pregnancy (TOP), MFS was not perceived as an intervention that substantially could improve the quality of their future child's life.

### **Conclusion:**

Prospective parents opting for MFS were driven by their feelings of parental responsibility. They recognise the fetus as their future child and value information and

care focusing on optimising the child's future health. In the small group of parents opting for TOP, MFS was felt to offer insufficient certainty of substantial improvement in quality of life and the perceived severe impact of SB drove their decision to end the pregnancy.

### **Keywords**

Maternal-fetal surgery, parental perspectives, acceptability, qualitative research

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## *Main text*

### **Introduction**

Maternal-fetal surgery (MFS) for open spina bifida (SB) has the potential to improve long-term outcomes but at significant procedure-related fetal and maternal risk<sup>1-5</sup>. Following antenatal diagnosis of open SB, prospective parents face a range of uncertainties regarding the future of their unborn child, and the options provided pose major ethical challenges<sup>6</sup>. In this situation parents frequently choose termination of pregnancy (TOP), as opposed to expectant prenatal management (EM) followed by postnatal assessment and surgery<sup>7, 8</sup>. MFS provides now an additional third option in which antenatal coverage of the fetal neural tissue is undertaken to prevent further damage. This intervention improves medium-term neurological outcomes, but risks premature membrane rupture, preterm delivery and rarely fetal death. For the mother, the procedure can cause perioperative morbidity and complications and risks uterine dehiscence in the current or future pregnancies, for which an elective caesarean section is required<sup>1-3, 9, 10</sup>. For parents, balancing these competing risks is challenging.

Successful implementation and evaluation of an intervention not only requires it to be effective, but also for it to be acceptable to those undergoing the intervention<sup>11</sup>. Acceptability can be defined as a construct that reflects the extent to which people receiving a healthcare intervention consider it appropriate, based on anticipated or experienced responses to the intervention<sup>12</sup>. Several aspects of the management pathways of fetal SB may be perceived to affect the acceptability of the intervention to parents. Little is known about the processes that parents experience in choosing a specific course, particularly as MFS is an option that could improve the long-term outcome but which also carries a significant risk to the pregnant woman and her fetus. This study investigates

the concepts and strategies parents employ when considering MFS as an option for the management of SB in their fetus, and how this determines the acceptability of the intervention.

## **Methods**

### *Theoretical Framework*

To assess the acceptability of MFS the Framework of Acceptability is used<sup>12</sup>. It is the result of a systematic approach to how the acceptability of healthcare interventions should be defined, assessed and theorised. The framework has been applied to define and assess acceptability of healthcare interventions in a variety of healthcare settings<sup>13-16</sup>. It comprised seven component constructs (Figure 1), i.e affective attitude (AA), burden (B), ethicality (E), intervention coherence (IC), perceived effectiveness (PE), opportunity costs (OC), and self-efficacy (SE). The component constructs reflect the extent to which parents consider the intervention to be appropriate, based on cognitive as well as emotional responses to the intervention<sup>12</sup>. As anticipated and actual lived experiences may differ, the framework measures acceptability over time: a) *prospective acceptability*: prior to participation in the intervention, b) *concurrent acceptability*: whilst in the intervention c) *retrospective acceptability*: after participating in the intervention.

### *Participant recruitment and selection*

Participants were recruited at two MFS partner centres to ensure standardized specialist assessment (University Hospitals Leuven, Belgium; University College London Hospital, United Kingdom) between July 2018 and February 2021. Prospective parents



that met the inclusion criteria for MFS, identical to those of the MOMS trial<sup>1</sup>, were eligible to participate, irrespective of their subsequent decision. All eligible parents were informed about the study at the fetal medicine unit, and if interested their details were shared with the researcher (NC). Interested parents were contacted via email by the researcher (NC) with study information (Patient Information Sheet, appendix 1). Participants were excluded if they were under 18 years old or unable to speak English or Dutch. Women were given the option of whether their partner joined them in the interview or not. All participants gave written informed consent. For participants undergoing TOP one interview was planned; this was conducted as least 6 weeks post termination to avoid distress and emotional burden. For participants opting for MFS three interviews were planned: the first two interviews were undertaken face-to-face during hospital admission, one prior to MFS and one after the intervention during hospitalization. The third interview took place 3-6 months after the birth of the child, by video-phone or video-conference, unless parents requested an alternative. For participants opting for EM two interviews were planned: one during pregnancy after making their decision and one after the birth of their child. Recruitment ceased when saturation was reached.

#### *Data collection*

All interviews were conducted by an experienced researcher (NC) and started with an open question inviting participants to share their thoughts, views, feelings and experiences whether to participate or not in MFS<sup>17, 18</sup>. The seven framework components were used as prompts to further explore the different levels of the decision (Interview-guide, appendix 2). Interviews lasted between 20 and 114 minutes, were

audio-recorded and transcribed verbatim by a professional transcription service (LSD Business Services, Derbyshire, UK; Amberscript, Amsterdam, the Netherlands).

Interview transcripts in Dutch were translated into English by NC for subsequent analysis.

### *Data analysis*

Data analysis was performed using the Qualitative Analysis Guide of Leuven (QUAGOL)(19). This method is characterized by the repetitive process and team approach. It consists of two parts, each with five analysis steps: the first aims to determine a conceptual understanding of the research data as a whole and the second comprises the coding process. Two researchers (AS and NC) read and re-read the interview transcripts, discussed observations and ideas, then formulated initial codes. Themes were identified and organised in line with the acceptability framework. Alternating between various stages of the process was required as new data and themes emerged, resulting in interaction between different parts of the analysis. The process was continued until data saturation was reached; no new information was obtained from subsequent interviews and thus recruitment was stopped<sup>20</sup>. NVivo V12 software (QSR International Pty Ltd. (2018)) facilitated data management, organisation and analysis. To enhance the rigour of the study the 32-item Consolidated Criteria for Reporting Qualitative research checklist was used (COREQ)<sup>21</sup>.

### *Data presentation*

The themes derived from the interview analysis of parents opting for MFS are detailed in Figure 2, arranged in temporal order to indicate differences in acceptability over time,

using the seven constructs identified by the framework of acceptability. Available results of the limited number of participants who opted for TOP and who were willing to participate, are presented descriptively. Themes extracted from the interviews with parents opting for MFS are illustrated by participants quotes, indicated in the text (e.g. Q1) and provided in Tables 1-3. Quotes are identified by father or mother and the patient's number (1-28). The acceptability framework component construct relating to each theme is presented in parenthesis.

### *Ethics*

This study was approved by the Ethics Committee of the University Hospitals Leuven, Belgium (S61586) and the South-Central Berkshire Research Ethics Committee UK National Research Ethics Committee (ref: 18/SC/0475).

## Results

Of 68 pregnant women with antenatally diagnosed fetal SB referred to the fetal surgical team over a 31-month period from July 2018 to February 2021, 47 were eligible for MFS. They were counselled about this option alongside potential alternatives including postnatal management or TOP. Of those, 26 opted for MFS, 19 for TOP and two for EM. Twenty-nine (62%) consented to study participation, with 24 parents opting for MFS (recruitment ceased October 2019 due to saturation) and only five (17%) opting for a TOP. No participants who were opting for postnatal surgery could be recruited to date (Figure 3). This paper reports the findings of the interviews with 24 participants (20 with partner present) who opted for MFS (Interview I: *prospective acceptability*; Interview II: *concurrent acceptability*; Interview III *retrospective acceptability*) and provides perspectives of the five participants (3 with partner present) who opted for TOP (Interview I *retrospective acceptability*). Upon the request of both parents, one interview took place after their decision, but prior to TOP. The socio-demographic characteristics of the parents and neonatal outcomes are displayed in Table 4.

In total we conducted 75 interviews. Five with parents opting for TOP and 70 with parents opting for MFS. For the latter, interviews were conducted at three different time points: prior to the intervention (n=24), during hospitalization (n=24) and after birth (n=22). Of the parents opting for MFS, two final interviews did not take place: one patient was lost to follow up and one mother who opted for neonatal palliative care following premature delivery declined further participation.

### *Attitudes and values*

Parents opting for MFS actively chose to proceed with their pregnancy; for some because of the MFS itself however for most because they did not want to terminate the pregnancy. Parents reported strong feelings of parental responsibility and were determined to do anything to improve their future child's health outcomes (E, Q1).

Following successful MFS, parents expressed enormous relief. Firstly, because all went well, but moreover because they perceived MFS as an opportunity they were given (AA) to fulfil their perceived responsibility towards their unborn child's wellbeing and outcome (E). The option of MFS provided hope for their child's future, in contrast to the initial presentation of the condition as a hopeless perspective (E, Q2). Almost all parents opting for MSF perceived that TOP at diagnosis was the recommended option.

After birth, parents either felt guilty at having considered TOP, or angry at their perception of being misinformed (E, Q16-18). None of the parents opting for MFS, regretted their initial decision (AA, Q9).

For the limited number of parents opting for TOP, the foreseen quality of life of their future child was decisive. SB was considered a condition impacting their future child's quality of life to such an extent they felt it would be unfair to proceed with the pregnancy. For four parents, TOP was the only right decision, while one family weighed MFS against the option of TOP. Additionally, they considered the impact of a child with SB on (future) siblings and the family. MFS was perceived as providing too little certainty on the potential positive impact on their future child's quality of life; maternal risks were not mentioned as reasons to decline. After the decision for TOP was taken, parents recalled ambivalent feelings of detachment from their fetus, often with

the intention to protect their selves from too much grieving, coexisting with feelings of attachment towards their fetus, as well as intense grief.

#### *Knowledge and risk perception*

In general, all parents were knowledgeable and aware of the potential and substantial risks associated with surgery. For example, parents could recall the odds of their child being able to walk without surgery as well as the potential for improvement with the surgery. Parents opting for MFS were determined to proceed with surgery (AA, Q3) because it seemed to them the only option that could alter the natural course of the condition and potentially improve their child's outcome (PE). Parents expressed hope that surgery would preserve motor functioning, but even more important, they hoped to preserve neuro-cognitive function (PE, Q4). Although driven by their hopes, potential complications and the uncertainty of surgical benefit made them fear their decision (IC).

The potential to increase the likelihood of their child being independent was perceived as a benefit to their family. In contrast, parents opting for TOP, did not consider MFS to add significantly to their future child's outcomes and its quality of life. Risk related to the current pregnancy in both groups of parents were not considered a decisive reason to proceed or not with MFS. For parents opting for TOP, impact on potential future pregnancies was among the reasons to decline MFS.

#### *Emotional impact*

Shortly before having MFS women felt emotional and feared losing their unborn child. Some described a fear of not waking up after the operation, in particular those who

already had children. Mothers termed this a 'mother's dilemma': by trying to do the best for their unborn child they feared their own mortality, and even more that their other children would grow up without a mother (AA, Q6). For most parents, the main emotion after waking from their anaesthetic was relief (AA). Fathers reported their fears that they could lose both their partner and their unborn child, and described the intra-operative waiting time as stressful. This was even more pronounced when that period was longer than expected (AA, Q7). Facing hospital discharge, some parents feared a break in continuity of care up until the planned delivery date, as they had been monitored intensely throughout their stay (SE). They described the intensive monitoring as reassuring, and as a consequence the thought of being without it whilst at home prior to delivery was worrying (AA). Fear of premature birth was reported by some parents, whilst others felt confident all would be fine (AA). Parents opting for TOP expressed ambiguity: on one hand they indicated they would have wanted to care for this baby, but at the same time would feel selfish, if they would decide to put the burden of a disability on this child, just because they wanted to become parents. They grieved the loss of their (dreamed) child and some expressed feelings of guilt; all described it as an impossible choice they had to make.

Postnatally in the MFS group three mothers suffered from mental health issues following the birth of their child (B). One mother mentioned she was diagnosed with post-traumatic stress disorder and two mothers with depression; all had received treatment. Parents reported feeling the effects of all that had happened only after they got their baby home. One mother expressed extreme fears of losing her child that were triggered both at the moment of antenatal diagnosis as well as when the neonatal team

indicated medical problems following her baby's birth. She later experienced the same fears of death for her older child. Two further mothers reported a diagnosis of depression in the postnatal period, which they related to the distress they experienced from living through a "rollercoaster" that started at the moment of diagnosis (B, Q19-20). In the TOP group, three mothers and one father received bereavement support, which helped them to cope with their loss.

### *Burden and impact*

Women opting for MFS discussed the logistical effort required to have MFS and reported significant and stressful difficulties organising last minute travel and accommodation (B). The stress of travelling and time away from work had a substantial financial impact on the families, and for some prompted the need to find additional funding for the MFS (B, Q5). Parents with children at home reported having to arrange care for their other children both during the hospital stay and recovery (OC).

The recovery in hospital, for some, was easier than anticipated but many mothers described the in-hospital recovery period as both physically and emotionally overwhelming. For some women the postoperative pain was described as extremely intense, however most women considered their hospital recovery as manageable due to the day-to-day improvements (B, Q8). Women experienced their home-based recovery very differently. Some were capable of doing some easy home and caring tasks, but others remained bed- or couch-ridden the whole pregnancy. One mother described so much pain following discharge she thought she and her baby were going to die (B, Q12). Hope for positive outcomes was the participants' main drive to undergo the burden of the recovery process (PE).



### *Outcomes*

At the final interview, all parents were happy with their child's outcome which they reported as being often better than they hoped for (PE). For some parents, however, certain outcomes such as hip-dislocation, despite being diagnosed, was disappointing or unexpected. Along with any additional unexpected findings such as reasons for longer hospitalization or not meeting certain milestones, challenged parents' hopes (PE, Q14). All parents discussed that there remained many uncertainties and acknowledged that the child's outcome would be revealed in time (IC, Q15).

### *Continuity of care*

Following MFS, a substantial proportion of women reported a break in the continuity of their pregnancy care, caused by what they perceived as lack of knowledge or understanding about their situation amongst their routine antenatal healthcare providers (SE, Q10). Some parents reported their concerns were not taken seriously by local clinical staff, which exacerbated their worries (AA, Q11) and made them feel responsible for their unborn child's wellbeing and outcome (SE). These concerns continued into the neonatal period, where parents reported a perceived lack of understanding or knowledge of their situation amongst the neonatal staff, giving them a feeling of not being taken seriously. Parents worried these challenges could offset all the investments they had made so far for their child, reporting a perception that they needed to provide extra protection for their newborn (E, Q13).

## **Discussion**

In this cohort of parents, acceptability for MFS was assessed by using the acceptability framework. For parents opting for MFS, acceptability remained high at each of the three interviews undertaken throughout their journey. Prior to the intervention it was perceived as an opportunity that needed to be taken. Parents seemed well aware of the risks involved and the uncertainty about individual outcomes. Expectations seemed to be realistic yet were driven by hope and expectation of the best outcome. None expressed regrets about their decision at any stage, despite the substantial impact. Few parents who opted for TOP were willing to participate. Those who did, considered the severe impact of SB on the quality of life of their future child as decisive. For most, TOP was the preferred option, one family weighed MFS against the option of TOP, but concluded that it would not add substantially to improve the quality of their future child's life.

Earlier studies providing an insight into parental experiences about MFS for SB confirm the high acceptability of the intervention. Most parents do not express regrets about their decision<sup>22</sup> and hold positive attitudes towards MFS<sup>23</sup>. Among parents opting for MFS, parental-fetal attachment, in this study described as the perceived parental responsibility towards their unborn child, is a construct that indicates a unidirectional and 'abstract' relation of the parents towards their fetus<sup>24-26</sup>. This seems an important motivating factor in the parents' decision to undertake MFS. Studies have indicated an increase in this phenomenon among parents receiving a prenatal diagnosis of a serious congenital malformation, including surgical conditions and twin-to-twin transfusion syndrome<sup>27, 28</sup>. In contrast, parents opting for TOP express this parental responsibility in

the context of protecting their child from a life with significant challenges and MFS was not considered to improve this significantly.

Many parents in this study recalled a strong contrast between the initial prenatal diagnosis and the subsequent option of MFS. MFS was perceived as an opportunity for potential improvement of their baby's outcome, giving hope for a more positive future outlook. Some parents even experienced feelings of guilt for having considered TOP. This memory remained powerful, and parents often reflected on it, both after surgery and the birth of their child, a finding also seen in mothers who have experienced severe congenital heart defect in their unborn child<sup>29</sup>. The parents in this study valued information and care focusing on the future from the healthcare providers, and the recognition of the fetus as a future child, similar to prenatal diagnostic trajectories of parents of children with Down syndrome<sup>30-32</sup>. A perceived break in the continuity of care between treatment centre and referral centre caused additional stress to an already intense trajectory. Families often are referred long-distance, or even international, which stresses the importance of a smooth transition and demands clear communication between centres.

The observation of attachment to their unborn child is interesting in the context of the debate on whether a fetus should be considered as a patient<sup>33, 34</sup>. Our interviews make it clear that the study participants already perceived their fetus as their future child from the moment they entered the MFS trajectory, and is similar to parents confronted with antenatal diagnosis of twin-to-twin transfusion syndrome, early onset fetal growth restriction and fetal congenital cardiac defects<sup>27, 28, 35</sup>. In contrast, in the TOP group,

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parents grieve the loss of their (dreamed) child, which they have tried to protect from a life with additional challenges, leading to coexistent feelings of detachment and attachment, which are known in comparable groups of parents<sup>36</sup>.

Three out of 24 mothers who opted for MFS were diagnosed with mental health issues after birth of their baby, which they relate to events they experienced during their pregnancy trajectory. Earlier studies have reported depression and anxiety in patients undergoing fetal diagnosis or receiving fetal therapy, suggesting that women underdoing MFS require appropriate follow-up and support to detect increased risks of mood and anxiety disorders<sup>37-39</sup>. This was already the case in women with fetal SB in the pre-fetal surgery era<sup>40, 41</sup>, to which the stress of the MFS and potential complications of prematurity is now added.

In this cohort, most parents, including the three mothers that were diagnosed with mental health issues, reported adjusting adequately to their new situation, regardless of the individual outcome. Hope, parenting satisfaction, income, marital adjustment, economical resources and (social) support are factors improving parental psychological adjustment in parents of children with SB, in contrast to disability related stress, negative family impact and negative coping strategies<sup>41</sup>. In this cohort, parental hopes, parenting satisfaction, and their social support network may have positively contributed to this early adjustment. Participants also reported that they felt adequately supported by the team's counselling and interaction, and even by study participation itself, the latter has been confirmed by others as well<sup>42-44</sup>.

#### *Strengths and limitations*

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One strength of this study is the high participation rate of parents who opted for MFS (24/ 26) in the period under study. A diverse case mix was seen, with heterogeneity for parity, age and educational level, enhancing the generalisability. We assessed their experience against a framework of acceptability using previously validated methodology. Another strength is that this study provides a first in-depth exploration of parental experience of fetal surgical SB repair focussing on the acceptability of the intervention, and thereby supports the provision of patient-centred care<sup>45-47</sup>. Two recent studies describing the information recommended to counsel parents whose fetus has SB did not address the patient perspective, or did so to a limited extent<sup>48, 49</sup>. However, this study has several limitations. We only had access to women referred for potential surgery at two centres managed by identical surgical teams. This ensured that parents received standardized specialist assessment and counselling regarding all three options. Patients who were referred were more likely to opt for MFS. This may explain the lower number than expected of parents opting for TOP and even no inclusions in the group of parents choosing postnatal surgery. Therefore, we are actually only able to provide some insights into these populations. We acknowledge the importance of the contrasting parental perspectives on providing a full scope and balanced presentation of what defines the acceptability of MFS to all parents. Given the relevance of different parental perspectives, further recruitment for TOP and EM should go on. Lastly, one mother experienced a neonatal loss. Due to persisting grief, this mother preferred not to participate in the final (postnatal) interview, therefore her perception of acceptability of what she was offered has not been explored.

## **Conclusion**

MFS for SB remains highly acceptable from diagnosis until 3-6 months postnatally. In contrast, parents who opted for TOP, MFS was not perceived as an intervention that substantially could improve the quality of their future child's life. For those opting for MFS, expectations seemed to be realistic yet were driven by hope and expectation of the best outcome. For parents opting for TOP, the potential benefit of MFS seems to play a minimal role in their final decision.

## References

1. Adzick NS, Thom EA, Spong CY, et al. A randomized trial of prenatal versus postnatal repair of myelomeningocele. *N Engl J Med*. 2011;364(11):993-1004.
2. Sacco A, Van der Veecken L, Bagshaw E, et al. Maternal complications following open and fetoscopic fetal surgery: A systematic review and meta-analysis. *Prenat Diagn*. 2019;39(4):251-68.
3. Inversetti A, Van der Veecken L, Thompson D, et al. Neurodevelopmental outcome of children with spina bifida aperta repaired prenatally vs postnatally: systematic review and meta-analysis. *Ultrasound Obstet Gynecol*. 2019;53(3):293-301.
4. Joyeux L, De Bie F, Danzer E, et al. Learning curves of open and endoscopic fetal spina bifida closure: systematic review and meta-analysis. *Ultrasound Obstet Gynecol*. 2020;55(6):730-9.
5. Sacco A, Simpson L, Deprest J, David AL. A study to assess global availability of fetal surgery for myelomeningocele. *Prenat Diagn*. 2018;38(13):1020-7.
6. Cao KX, Booth A, Ourselin S, et al. The legal frameworks that govern fetal surgery in the United Kingdom, European Union, and the United States. *Prenat Diagn*. 2018;38(7):475-81.
7. Ovaere C, Eggink A, Richter J, et al. Prenatal diagnosis and patient preferences in patients with neural tube defects around the advent of fetal surgery in Belgium and Holland. *Fetal Diagn Ther*. 2015;37(3):226-34.
8. Morris JK, Rankin J, Draper ES, et al. Prevention of neural tube defects in the UK: a missed opportunity. *Arch Dis Child*. 2016;101(7):604-7.
9. Johnson MP, Bennett KA, Rand L, et al. The Management of Myelomeningocele Study: obstetrical outcomes and risk factors for obstetrical complications following prenatal surgery. *Am J Obstet Gynecol*. 2016;215(6):778 e1- e9.
10. Farmer DL, Thom EA, Brock JW, 3rd, et al. The Management of Myelomeningocele Study: full cohort 30-month pediatric outcomes. *Am J Obstet Gynecol*. 2018;218(2):256 e1- e13.
11. Diepeveen S, Ling T, Suhrcke M, et al. Public acceptability of government intervention to change health-related behaviours: a systematic review and narrative synthesis. *BMC Public Health*. 2013;13:756.
12. Sekhon M, Cartwright M, Francis JJ. Acceptability of healthcare interventions: an overview of reviews and development of a theoretical framework. *BMC Health Serv Res*. 2017;17(1):88.
13. McInnerney D, Candy B, Stone P, Kupeli N. Let It Out (LIO) study: protocol for a mixed-methods study to optimise the design and assess the feasibility of an online emotional disclosure-based intervention in UK hospices. *BMJ Open*. 2021;11(5):e047135.
14. Galea S, Salvaris CA, Yap MBH, et al. Feasibility and acceptability of an enhanced cognitive behavioural therapy programme for parent-child dyads with anxiety disorders: a mixed-methods pilot trial protocol. *Pilot Feasibility Stud*. 2021;7(1):109.

15. Roponen J, Ruusunen A, Absetz P, et al. Nutrition-focused group intervention with a strength-based counseling approach for people with clinical depression: a study protocol for the Food for Mind randomized controlled trial. *Trials*. 2021;22(1):344.
16. Morton K, Dennison L, Band R, et al. Implementing a digital intervention for managing uncontrolled hypertension in Primary Care: a mixed methods process evaluation. *Implement Sci*. 2021;16(1):57.
17. Yeo A, Legard R, Keegan J, et al. In-depth interviews. In: Ritchie J, Lewis J, McNaughton-Nicholls C, Ormston R, editors. *Qualitative Research Practice*. 2nd ed. London: Sage; 2014.
18. Rubin H, Rubin I. *Qualitative interviewing*. Thousand Oaks: Sage; 2005.
19. Dierckx de Casterle B, Gastmans C, Bryon E, Denier Y. QUAGOL: a guide for qualitative data analysis. *Int J Nurs Stud*. 2012;49(3):360-71.
20. Ritchie J, Lewis J, Elam G, et al. Designing and selecting samples. In: Ritchie J, Lewis J, McNaughton-Nicholls C, Ormston R, editors. *Qualitative Research Practice*. London: Sage; 2014.
21. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care*. 2007;19(6):349-57.
22. Antiel RM, Janvier A, Feudtner C, et al. The experience of parents with children with myelomeningocele who underwent prenatal surgery. *J Pediatr Rehabil Med*. 2018;11(4):217-25.
23. Fry JT, Frader JE. "We want to do everything": how parents represent their experiences with maternal-fetal surgery online. *J Perinatol*. 2018;38(3):226-32.
24. Van den Bergh B, Simons A. A review of scales to measure the mother-foetus relationship. *Journal of Reproductive and Infant Psychology*. 2009;27(2):114-26.
25. McNamara J, Townsend ML, Herbert JS. A systemic review of maternal wellbeing and its relationship with maternal fetal attachment and early postpartum bonding. *PLoS One*. 2019;14(7):e0220032.
26. Doan HM, Zimerman A. Conceptualizing prenatal attachment: Toward a multidimensional view. *Journal of Prenatal & Perinatal Psychology & Health*. 2003;18(2):109-29.
27. Ruschel P, Zielinsky P, Grings C, et al. Maternal-fetal attachment and prenatal diagnosis of heart disease. *Eur J Obstet Gynecol Reprod Biol*. 2014;174:70-5.
28. Mackie FL, Pattison H, Jankovic J, et al. Parental attachment and depressive symptoms in pregnancies complicated by twin-twin transfusion syndrome: a cohort study. *BMC Pregnancy Childbirth*. 2019;20(1):4.
29. Bertaud S, Lloyd DFA, Sharland G, et al. The impact of prenatal counselling on mothers of surviving children with hypoplastic left heart syndrome: A qualitative interview study. *Health Expect*. 2020;23(5):1224-30.
30. Skotko B. Mothers of children with Down syndrome reflect on their postnatal support. *Pediatrics*. 2005;115(1):64-77.
31. Skotko BG. Prenatally diagnosed Down syndrome: mothers who continued their pregnancies evaluate their health care providers. *Am J Obstet Gynecol*. 2005;192(3):670-7.



32. Crombag NM, Page-Christiaens GC, Skotko BG, de Graaf G. Receiving the news of Down syndrome in the era of prenatal testing. *Am J Med Genet A*. 2020;182(2):374-85.
33. Lyerly AD, Little MO, Faden RR. A critique of the 'fetus as patient'. *Am J Bioeth*. 2008;8(7):42-4; discussion W4-6.
34. Chervenak FA, McCullough LB. Ethical dimensions of the fetus as a patient. *Best Pract Res Clin Obstet Gynaecol*. 2017;43:2-9.
35. Sheppard M, Spencer RN, Ashcroft R, David AL. Ethics and social acceptability of a proposed clinical trial using maternal gene therapy to treat severe early-onset fetal growth restriction. *Ultrasound Obstet Gynecol*. 2016;47(4):484-91.
36. Lou S, Hvidtjørn D, Jørgensen ML, Vogel I. "I had to think: This is not a child." A qualitative exploration of how women/couples articulate their relation to the fetus/child following termination of a wanted pregnancy due to Down syndrome. *Sex Reprod Healthc*. 2021;28:100606.
37. Vergote S, Lewi L, Gheysen W, et al. Subsequent fertility, pregnancy, and gynecologic outcomes after fetoscopic laser therapy for twin-twin transfusion syndrome compared with normal monochorionic twin gestations. *Am J Obstet Gynecol*. 2018;218(4):447 e1- e7.
38. Gregoir C, Engels AC, Gomez O, et al. Fertility, pregnancy and gynecological outcomes after fetoscopic surgery for congenital diaphragmatic hernia. *Hum Reprod*. 2016;31(9):2024-30.
39. Beck V, Opdekamp S, Enzlin P, et al. Psychosocial aspects of invasive fetal therapy as compared to prenatal diagnosis and risk assessment. *Prenat Diagn*. 2013;33(4):334-40.
40. Pinquart M. Parenting stress in caregivers of children with chronic physical condition-A meta-analysis. *Stress Health*. 2018;34(2):197-207.
41. Driscoll CFB, Stern A, Ohanian D, et al. Parental perceptions of child vulnerability in families of youth with spina bifida: The role of parental distress and parenting stress. *Journal of Pediatric Psychology*. 2018;43(5):513-24.
42. Lakeman R, McAndrew S, MacGabhann L, Warne T. 'That was helpful ... no one has talked to me about that before': Research participation as a therapeutic activity. *Int J Ment Health Nurs*. 2013;22(1):76-84.
43. Peddie VL, Porter M, Van Teijlingen E, Bhattacharya S. Research as a therapeutic experience? An investigation of women's participation in research on ending IVF treatment. *Hum Fertil (Camb)*. 2006;9(4):231-8.
44. Colbourne L, Sque M. The culture of cancer and the therapeutic impact of qualitative research interviews. *Journal of Research in Nursing*. 2005;10(5):551-67.
45. Coulter A, Collins A. Making Shared Decision-Making a Reality. No decision about me, without me. London; 2011. Report No.: 1126980.
46. Stacey D, Legare F, Lewis K, et al. Decision aids for people facing health treatment or screening decisions. *Cochrane Database Syst Rev*. 2017;4:CD001431.
47. Black N. Patient reported outcome measures could help transform healthcare. *BMJ*. 2013;346:f167.
48. Gotha L, Pruthi V, Abbasi N, et al. Fetal spina bifida: What we tell the parents. *Prenat Diagn*. 2020.

49. Ravindra VM, Aldave G, Weiner HL, et al. Prenatal counseling for myelomeningocele in the era of fetal surgery: a shared decision-making approach. *J Neurosurg Pediatr.* 2020:1-8.

Title of the study: [The parental perspectives in decision-making for fetal surgery](#)

Sponsor: [University Hospitals Leuven](#)

Department: [Development and Regeneration, Biomedical Sciences, Herestraat 49, 3000 Leuven](#)

Ethical committee: [Ethical Committee Research UZ/ KU Leuven](#).

Study supervisor and researchers: [Prof. Jan Deprest, \[jan.deprest@uzleuven.be\]\(mailto:jan.deprest@uzleuven.be\), Fetal Medicine, UZ Gasthuisberg, Herestraat 49, bus 7003 06, 3000 Leuven](#); researcher: [Neeltje Crombag \[neeltje.crombag@kuleuven.be\]\(mailto:neeltje.crombag@kuleuven.be\)](#)

## **I Important information for deciding whether to take part**

### **Introduction**

You are being invited to take part in an interview study to investigate the parental perspectives in pregnancies where fetal surgery is an option. Before you decide, we would like you to understand why the interviews are being done and what they involve. Please take time to read the following information carefully and discuss it with others if you wish. Your decision will be respected and will not affect the standard of care you receive. One of our team will go through the information sheet with you and will answer any questions.

Before you agree to take part in this study, we would like you to be aware of the time commitment, possible disadvantages and benefits, to allow you to make a decision with full awareness of the implications. This is known as giving “informed consent”.

Please read this information carefully and ask the investigator (Prof Jan Deprest) or his representative any questions you have. There are three parts to this document: the information essential to your decision, your written consent and supplementary information (appendices) detailing certain aspects of the study.

### **If you take part in this clinical study, you should be aware that:**

- This clinical study is being conducted after review by one or more ethics committees.
- Your participation is voluntary and must remain free from any coercion. It requires the signature of a document expressing your consent. Even after signing this document, you can stop taking part by informing the investigator. Your decision not to take part or to stop taking part in the study will have no impact on the quality of your care or on your relationship with the investigator.
- The data collected is confidential and your anonymity is guaranteed during publication of the results.
- Insurance has been taken out in case you should suffer any damage in connection with your participation in this clinical study.
- You will not incur any charges for the visits/consultations, examinations or treatments specific to this study.
- You may contact the investigator or a member of his team at any time should you need any additional information.

Further information about your “Rights as a participant in a clinical study” can be found on page 5.

### **Objectives and description of the study protocol**

During this pregnancy, your baby has been diagnosed with a structural (physical) anomaly (abnormality) and fetal surgery is among the options. As you might know, structural anomalies are often detected in pregnancy and have a large variety of possible effects on the future child’s health.

You will have hopefully spoken with an expert and had the chance to obtain all of the information that you need about your baby's diagnosis and about all of the choices available to you. Among other options, such as postnatal surgery or termination of pregnancy, there is the option for fetal surgery. Fetal surgery is a procedure carried out on a baby before it is born. We are inviting you to take part in an interview study to investigate the parental perspectives in pregnancies in which fetal surgery is among one of the options. This study is part of a larger research project, called GIFT-Surg, which aims to improve the results of fetal surgery, primarily for spina bifida and congenital diaphragmatic hernia. As surgery on the baby before it is born is a new treatment, we are interested in the factors that influence your decision about what to do in this pregnancy, *whether or not you decide to proceed with the pregnancy or to take up the option of fetal surgery*. So that we can improve the support we give to parents in these situations, we would like to find out how *you* view these difficult decisions, and what *your* thoughts, feelings and needs are during this stressful period. We call this **the parental perspective**. Learning more about your perspective will help us to improve support and care for future parents facing the same difficult decisions.

### **Course of the study**

We would like to have a face-to-face interview, alongside a hospital visit, either with or without your partner. The interview will be carried out by a midwife and research fellow, Mrs Neeltje Crombag. She is experienced both in carrying out these types of interviews, and is experienced in caring for patients in similar situations. During the interview, we will ask you to share your thoughts, views, feelings and experiences related to the decision about what to do in this pregnancy. As we are interested in the individual experiences of expecting parents, there are no right or wrong answers, and we would like to learn about *your thoughts and experiences*.

If you decide to take part in the study, you will be invited for at least two interviews: the first interview will take place shortly after one of your hospital consultations. The second interview will take place several months after the birth of the baby, or termination of the pregnancy. This will be scheduled according to your preference and availability, either by Skype or phone. If you decide to have fetal surgery, a third interview will take place after the surgery, while you are still in the hospital and feel well enough to speak with the researcher.

The interview will last approximately 30-60 minutes, in which the interviewer will mainly listen to your personal experiences. The interviews will be recorded as an audio-file, and transcribed verbatim (word-for-word) to facilitate the subsequent analysis.

Any information that can be used to identify you (name, address, telephone number) will be stored securely at UZ Leuven, the hospital that is sponsoring the study, either in a locked filing cabinet or on a securely protected computer. The recording of the interview will be deleted from the devices and computer, after the transcription and accuracy check. The written version of the interview and the information we get from these will be stored securely at UZ Leuven servers, on a password protected computer. Information about you may be looked at by authorised staff only: Mrs Crombag and Professor Jan Deprest. Both have a duty of confidentiality to you. Your personal details will only be kept whilst we need to contact you, following which they will be securely destroyed. The written versions will be kept for 20 years after the study ends. There will be nothing that identifies you in this material or in the results.

### **Benefits and disadvantages**

#### *Possible benefits:*

There is no immediate and direct benefit to you. However, by taking part in the study you are supporting research into fetal therapy, and in particular, care for future parents who also have a pregnancy affected by similar conditions. For some parents sharing their feelings, experiences and thoughts helps them to cope with the stressful period they go through.

*Possible disadvantages:*

You will need to speak to us prior to the interview, for about 20 minutes to discuss the study and arrange for the interviews. The first interview will be face-to face but combined with a scheduled hospital visit and lasts 30-60 minutes. Subsequent interviews can be done via Skype or phone upon your preference.

**Withdrawal from the study**

Your participation is voluntary and you are entitled to withdraw from the study for any reason, without having to justify your decision. Nevertheless, it may be useful for the investigator and for the sponsor of the study to know if you are withdrawing because the demands of participation are too great.

**If you take part in this clinical study, we ask you to:**

- Help us in the smooth running of this study.
- Be present at the scheduled timing of the interview and to participate in the interview
- Inform the researcher if you consider participating in another study, to discuss whether you can participate in both studies or not

When the study is finished, you will receive a summary of the results.

**Contact**

If you need additional information or if you have any concerns you can contact the investigator (Professor Jan Depreest) by phone +32 16 34 51 23 (working hours) or e-mail ([Jan.Depreest@uzleuven.be](mailto:Jan.Depreest@uzleuven.be)) or a member of his research team (Neeltje Crombag, [neeltje.crombag@kuleuven.be](mailto:neeltje.crombag@kuleuven.be), by phone: +32 16345222 who will answer your questions or contact the study supervisor.

If you have any questions relating to your rights as a participant in a clinical study, you can contact the patient rights ombudsman of UZ Leuven on this telephone number +3216 34 48 18. If necessary, he/she can put you in contact with the ethics committee.

Title of the study: [The parental perspectives in decision-making for fetal surgery](#)

## II Informed consent

### Participant

I declare that I have been informed of the nature of the study, its purpose, its duration, any risks and benefits and what is expected of me. I have taken note of the information document and the appendices to this document.

I have had sufficient time to think about it and discuss it with a person of my choice, such as my GP or a member of my family.

I have had the opportunity to ask any questions that came to mind and have obtained a satisfactory response to my questions.

I understand that my participation in this study is voluntary and that I am free to end my participation in this study without this affecting my relationship with the therapeutic team in charge of my health.

I understand that data about me will be collected throughout my participation in this study and that the investigator and the sponsor of the study will guarantee the confidentiality of these data.

I agree to my personal data being processed as described in the section dealing with confidentiality guarantees (page 5). I also consent to these data being transferred to and processed in countries other than Belgium.

**I agree/ I do not agree** (delete as appropriate) to the study data being processed at a later date, provided this processing is within the context of the present study, for a better understanding of the disease and its treatment or improvements in patient communication and care.

I have received a copy of the information to the participant and the informed consent form.

*Surname:* ..... *First name:* .....

*Date:* .....

*Signature of the volunteer:* .....

**Investigator**

I, the undersigned, [surname, first name] investigator/clinical study assistant, confirm that I have verbally provided the necessary information about the study and have given the participant a copy of the information document.

I confirm that no pressure was applied to persuade the patient to agree to take part in the study and that I am willing to answer any additional questions if required.

I confirm that I operate in accordance with the ethical principles set out in the latest version of the "Helsinki Declaration", the "Good Clinical Practices" and the Belgian Law of 7 May 2004 related to experiments on humans.

*Surname of the investigator:* .....

*First name of the investigator:* .....

*Date:* .....

*Signature of the investigator:* .....

*Surname of the investigator's representative:* .....

*First name of the investigator's representative:* .....

*Date:* .....

*Signature of the investigator's representative:* .....

Accepted Article

Titel van de studie: **Het ouderperspectief bij besluitvorming rondom foetale chirurgie**

### **III Supplementary information**

#### **The protection and the rights of the participant in a clinical study :**

##### ***Ethics Committee***

This study has been reviewed by an independent Ethics Committee, namely the Ethics Committee of UZ/KU Leuven, which has issued a favourable opinion. It is the task of the Ethics Committees to protect people who take part in a clinical trial. They make sure that your rights as a patient and as a participant in a clinical study are respected; that based on current knowledge, the balance between risks and benefits remains favourable to the participants and that the study is scientifically relevant and ethical.

You should not under any circumstances take the favourable opinion of the Ethics Committee as an incentive to take part in this study.

##### ***Voluntary participation***

Before signing, do not hesitate to ask any questions you feel are appropriate. Take the time to discuss matters with a trusted person if you so wish.

Your participation in the study is voluntary and must remain free of any coercion: this means that you have the right not to take part in the study or to withdraw without giving a reason, even if you previously agreed to take part. Your decision will not affect your relationship with the investigator or the quality of your future therapeutic care.

However, it is advisable for your safety to inform the investigator if you have decided to stop taking part in the study.

If you agree to take part, you will be asked to sign the informed consent form. The investigator will also sign this form to confirm that he/she has provided you with the necessary information about the study. You will receive a copy of this form.

##### ***Costs associated with your participation***

If you decide to take part in this study, this will not involve any extra costs for you or your insurer, and you will not be reimbursed for your participation.

##### ***Guarantee of confidentiality***

Your participation in the study means that you agree to the investigator collecting data about you and to the study sponsor using these data for research purposes and in connection with scientific and medical publications.

Your data will be processed in accordance with the European General Data Protection Regulation (GDPR) and with the Belgian legislation on the protection of natural persons with regard to the processing of personal data.

You are entitled to ask the investigator what data are being collected about you and what its use is in connection with the study. This data concerns your current clinical situation but also some of your background, the results of examinations carried out and the results of examinations required by the protocol. You have the right to inspect this data and correct it if it is incorrect<sup>1</sup>.

The investigator has a duty of confidentiality regarding the data collected.

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<sup>1</sup> These rights are guaranteed by the Law of 8 December 1992 on the protection of privacy in relation to the processing of personal data and by the Law of 22 August 2002 on patient rights.



This means that he/she undertakes not only to never to reveal your name in the context of a publication or conference but also that he/she will encode your data (i.e. your identity will be replaced by an ID code in the study) before sending it to the manager of the database of collected data (KU/UZ Leuven, Leuven).

The investigator and his/her team will therefore be the only ones to be able to establish a link between the data transmitted throughout the study and your medical records<sup>2</sup>.

The personal data transmitted will not contain any combination of elements that might allow you to be identified<sup>3</sup>.

For the study data manager designated by the sponsor, the data transmitted will not allow you to be identified. The latter is responsible for collecting the data gathered by all investigators taking part in the study, processing them and protecting them in accordance with the requirements of the Belgian law on the protection of privacy.

To verify the quality of the study, it is possible that your medical records will be examined by persons subject to professional secrecy and designated by the ethics committee, the sponsor of the study or an independent audit body. In any event, this examination of your medical records may only take place under the responsibility of the investigator and under the supervision of one of the collaborators designated by him/her.

The encoded study data will be able to be sent to Belgian or other regulatory authorities, to the relevant ethics committees, to other doctors and/or to organisations working in collaboration with the sponsor.

Your consent to take part in this study therefore also implies your consent to the use of your encoded medical data for the purposes described in this information form and to their transmission to the aforementioned people and authorities.

The sponsor undertakes only to use the data collected within the context of the study in which you are taking part.

If you withdraw your consent to take part in the study, to guarantee the validity of the research, the data encoded up to the point at which you withdraw will be retained. No new data may be sent to the sponsor.

If you have any questions relating to how your data are being processed, you may contact the investigator. The data protection officer in your hospital can be contacted as well: DPO - UZ Leuven, Herestraat 49, 3000 Leuven, e-mail [dpo@uzleuven.be](mailto:dpo@uzleuven.be).

Finally, if you have a complaint concerning the processing of your data, you can contact the Belgian supervisory authority who ensures that privacy is respected when personal data are processed.

The Belgian supervisory authority is called:

Data Protection Authority (DPA)

Drukpersstraat 35,

1000 Brussels

Tel. +32 2 274 48 00

e-mail: [contact@apd-gba.be](mailto:contact@apd-gba.be)

Website: <https://www.dataprotectionauthority.be>

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<sup>2</sup> For clinical trials, the law requires this link with your records to be retained for 20 years. In the case of an advanced therapy medicinal product using human biological material, this period will be a minimum of 30 years and a maximum of 50 years in accordance with the Belgian Law of 19 December 2008 on the use of human biological material and the applicable royal decrees.

<sup>3</sup> The database containing the results of the study will therefore not contain any combination of elements such as your initials, your gender and your full date of birth (dd/mm/yyyy).

**Insurance**

Any participation in a clinical study involves a risk, however small it is. Even if there is no fault, the sponsor accepts responsibility for damage caused to the participant (or in the event of death, his/her dependants) and directly or indirectly linked to his/her participation in the study. The sponsor has taken out insurance for this responsibility<sup>4</sup>.

Van Breda Risks & Benefits NV, Plantin en Moretuslei 297, 2140 Antwerpen, polisnummer 299.053.700.

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<sup>4</sup>In accordance with Article 29 of the Belgian Law related to experiments on humans (7 May 2004)



Guided Instrumentation for  
Fetal Therapy and Surgery

## Participant Information Sheet

Version: 2.0

Date: 9/OCTOBER/2018

REC Reference: 18/SC/0475

Principal Investigator:

Professor Neil Marlow, Consultant in Neonatal Medicine

Researchers:

Neeltje Crombag PhD, Midwife and Research Fellow,  
Professor Anna David, Consultant in Fetal Medicine

## Fetal surgery interview study: Parental perceptions of Fetal Surgery

You are being invited to take part in an interview study. Before you decide, we would like you to understand why the interviews are being done and what it involves. Please take time to read the following information carefully and discuss it with others if you wish. Your decision will be respected and will not affect the standard of care you are receiving. One of our team will go through the information sheet with you and will answer any questions.

### What is the purpose of the study?

During this pregnancy, your baby has been diagnosed with spina bifida. As you might know these are problems that often arise early in pregnancy and with a large variety of possible effects on your child's health. You will have spoken with a Doctor and you have had the chance to obtain all of the information that you need about your baby's diagnosis and about all of the choices available to you. When your baby is diagnosed with spina bifida, there are three options: postnatal therapy, termination of pregnancy or the option for fetal surgery. Fetal surgery, is a surgical procedure carried on a baby before it is born. This study is part of a larger research project, called Guided Instrumentation for Fetal Therapy and Surgery (GIFT-Surg), which aims to improve the results of fetal surgery, primarily for these two conditions.

As surgery on the baby before it is born is a very new treatment, we are interested in the factors that influence your decision about what to do in this pregnancy, whether or not you decide to proceed with the pregnancy or choose termination, or choose fetal surgery or surgery after birth. So that we can help to design the support we give to parents in these situations, we would like to find out how you view these difficult decisions, and what the thoughts, feelings and needs are of parents experiencing this stressful period. We call this the parental perspective. Learning more about your perspective will help us to improve support and care for future parents facing the same difficult decisions.

### Why have I been invited?

You have been invited in this study as your baby has been diagnosed with spina bifida and you are able to choose the option of fetal surgery. We are interested in all parents facing the dilemmas when expecting a child with likely spina bifida or diaphragmatic hernia, regardless of any decision you have made or will make.



### **Do I have to take part?**

No. It is up to you to decide to be interviewed and your decision will be respected. If you agree to be interviewed, we will ask you to sign a consent form. You are free to withdraw at any time, without giving a reason. If you would rather not take part, or if you later want to withdraw, this will not affect the care you receive.

### **What happens if I am interested in taking part?**

You may discuss this with the member of the team who discussed the project with you or contact Neeltje Crombag. Dr Crombag is a midwife and research fellow employed by the GIFT-Surg study specifically to carry out these interviews. She will arrange a time to speak to you on the telephone to go over the details of the interview study and answer any of your questions, which should not take more than 20 minutes.

### **Can my partner take part in the interview?**

Yes. Please let us know when you contact us whether your partner would like to take part in the interview. If possible we would like to speak to both of you before the interview.

### **What will I have to do if I decide to take part?**

We would like to have two or three face-to-face interviews, with you and your partner. One interview after your first consultation, and one, three months after the birth of the baby, or if you choose to terminate the pregnancy, three months after the termination. If you decide to have fetal surgery, one additional interview will take place during the week of hospitalisation after the intervention. These interviews will all take place alongside a hospital visit, while you are hospitalised and/or via Skype or phone. It will last approximately 30-60 minutes and will be recorded as an audio-file. During the interview, we will ask you to share your thoughts, views, feelings and experiences related to the decision about what to do in this pregnancy. As we are interested in the individual experiences of expecting parents, there are no right or wrong answers, and we would like to learn about your thoughts and experiences.

### **What are the possible inconveniences and risks of taking part?**

It may be distressing for you to speak about your experience in this pregnancy, in which you have heard the news your baby is affected by spina bifida. You will also need to speak to us on the telephone for about 20 minutes to discuss the study and arrange for the interviews. The first interview will be face-to-face but combined with a scheduled hospital visit. Subsequent interviews can be done via Skype or phone upon your preference.

### **What are the possible benefits of taking part?**

There is no immediate and direct benefit to you. However, by taking part in the study you are supporting research into fetal therapy, and in particular care for future parents who also have a pregnancy affected by the same and similar conditions. For some parents sharing their feelings, experiences and thoughts helps them to cope with the stressful period they go through.

### Is there any payment for taking part?

You will receive expenses when you need to travel for the study, but no other payments for taking part.

### Will my taking part in the study be kept confidential?

The interview will be audio-recorded and transcribed for the purpose of analysis. Transcription will be carried out by a professional transcription service, and all information will be anonymised so neither you nor your doctors can be recognised from the written transcript. All information about you will be kept confidential, following legal and ethical practice, unless there is serious risk of harm. No information will be disclosed outside the study without first discussing this with you.

### What will happen to information about me?

UCL is the sponsor for this study based in London, the United Kingdom. We will be using information from you and/or your medical records in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. UCL will keep identifiable information about you for 2 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

You can find out more about how we use your information at <https://www.ucl.ac.uk/legal-services/data-protection-overview>, or by e-mailing the UCL Data Protection Office [[data-protection@ucl.ac.uk](mailto:data-protection@ucl.ac.uk)].

Any information that can be used to identify you (name, address, telephone number) will be stored securely at UCL, the university that is sponsoring the study, either in a locked filing cabinet or on a securely protected computer. The recording of the interview will be given a code/identification number and will not include your name or contact details. The recording, a written version of the interview and the information we get from these will be stored securely at UCL on a password protected computer. Information about you may be looked at by authorised staff only: Dr Crombag and Professor Neil Marlow. Both have a duty of confidentiality to you. Your personal details will only be kept whilst we need to contact you, following which they will be securely destroyed. Recordings and the written versions will be kept for 2 years after the study ends. There will be nothing that identifies you in this material or in the results.

UCL will use your name and contact details to contact you about the research study, and make sure that relevant information about the study is recorded for your care, to oversee the quality of the study. Individuals from UCLH and regulatory organisations may look at your medical and research records to check the accuracy of the research study. UCLH will pass these details to UCL along with the information collected from you and/or your medical records. The only people in UCL who will have access to information that identifies you will be people who need to contact you to audit the data collection process. The people who receive the results of the study will not be able to identify you and will not be able to find out your name, NHS number or contact details.



Guided Instrumentation for  
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UCL will keep identifiable information about you from this study for 2 years after the study has finished.

### **What will happen if I don't want to carry on in the study?**

You can withdraw from the interview at any time by contacting the researcher.

### **What if I find the interview upsetting?**

You can contact our local Fetal Medicine Unit midwives or your GP. You may also find it useful to contact the following charities: Shine charity at [www.shinecharity.org.uk](http://www.shinecharity.org.uk) or 01733555988 or [firstcontact@shinecharity.org.uk](mailto:firstcontact@shinecharity.org.uk), ARC (Antenatal Results and Choices) at [www.arc-uk.org](http://www.arc-uk.org) or 0845 077 2290; BLISS at [www.bliss.org.uk](http://www.bliss.org.uk) or 0500 618140; SANDS at [www.uk-sands.org](http://www.uk-sands.org) or 020 7436 5881.

### **What if there is a problem?**

Please ask to speak to the researchers who will answer your questions ([neeltje.crombag@kuleuven.be](mailto:neeltje.crombag@kuleuven.be) or [a.david@ucl.ac.uk](mailto:a.david@ucl.ac.uk)). If your problem is not resolved or you want to speak to someone independent you can contact the UCLH Fetal Medicine Unit on 020 3447 6195. If you remain unhappy and wish to complain formally, you can contact Patient Advisory Liaison Services (PALS) on 020 3447 3042.

### **Who is organising and funding the research?**

The study is being run by the University College London Hospital, is sponsored by University College London and is funded by the Wellcome Trust and the Engineering and Physical Sciences Research Council (EPSRC).

### **Who has reviewed the study?**

All research in the NHS is looked at by two independent groups of people. All patient-based research is reviewed first by the Health Research Authority, and subsequently by one of their Research Ethics Committees, to protect your interests. This study has been given favourable opinion by South Central - Berkshire Research Ethics Committee.

### **What will happen to the results of the research study?**

The findings will be published in medical journals and a summary will be sent to you, if you so wish, and will be available on the GIFT-Surg website ([www.gift-surg.ac.uk](http://www.gift-surg.ac.uk)). You will not be identified in any report or publication, but we may include anonymous direct quotations from the interview.



## Interview guide

The aim of the interview is to assess how women (and their partners) perceive the acceptability of a fetal surgical intervention for MMC and CDH, with regards to the GIFT-surg project. Participants will be asked to share their thoughts, views, feelings and experiences with regards to their decision to participate in fetal surgery.

Due to the nature of the interview, the interview style to be used is called 'responsive' interviewing(1) This interview style emphasizes on the importance of building a relationship of trust between interviewer and interviewee. Therefore, it is essential to invest on building on this relationship. This is mainly done by creating a quiet and calm environment, an empathically neutral position of the interviewer and an open approach by using both verbal and non-verbal communication (2). The interview will not be set-up as a question-answer setting, but as a setting in which the respondent feels free to share their story, to get an understanding of the respondent's perceptions, feelings, thoughts, views and experiences. In other words, the respondent needs to do most of the talking. The interviewer will guide her through the key-themes.

### *Stages of the interview*

Before recording the interview, the interviewer will introduce her/himself to the interviewee. This must be seen as a normal introduction, when two (or three people) first meet. These first few minutes are crucial for establishing a good *rapport* (2)(3). Building *rapport* means that the interviewer shows to the respondent that she/ he is sincerely interested in her/ their story (STAGE 1). When both interviewer and respondent(s) are at ease, the interviewer will introduce the research and explain the purpose of the interview. It will be emphasized that participating is voluntary, and arrangements of confidentiality will be set out. There is no wrong or right answer, the interviewer is interested the respondent's perspective, in their own words. The respondents will be informed that if they do not wish to speak about certain areas, they do not have to (STAGE 2). When both interviewer and respondent(s) are at ease, the interviewer will empathically reflect on the personal situation of the respondent(s) and the past few weeks and days in which they have received the diagnosis and all the different options. First, this is to collect some information on their history and context (the interviewer is informed on major details), second this is invite the respondent to reflect on what happened to them. The interviewer will not record this, but will make notes if necessary. This first phase is also used to collect the *contextual questions*, if not already collected (STAGE 3). The actual interview will start then with an open question: "I would like to learn more on how you have come to the decision for the treatment of your baby: fetal surgery". Respondents will be encouraged to share their thoughts, views, feelings and experiences regarding the decision to participate in fetal surgery. By using probing questions, the interviewer further explores the different levels of this decision. The seven components of the *theoretically framework of acceptability* will be used as *key themes* or *prompts* (affective attitude, burden, perceived effectiveness, ethicality, intervention coherence, opportunity costs, and self-efficacy (Table 1)), but any other related themes emerging from the respondent, to seek breadth and depth of coverage (STAGE 4). Shortly before the end of the interview (5 to 10 minutes), the interviewer will signal the approach of the end of the interview, to encourage the respondents to raise anything important. This can be done by simply asking for final thoughts or comments (STAGE 5). The interview will be finished by thanking the respondent for her/his participation, and emphasize on the value of their participation. This is also the time give contact details for further questions, or support services. This is also the moment to discuss how and when we will contact the respondent for the next interview.

Thoughts and emotions regarding taking part in the intervention	<i>Affective attitude</i>
The perceived amount of effort that is required to participate in the intervention (to much cognitive effort, too much risks, too expensive, too much time)	<i>Burden</i>
How is the intervention judged by a person's individual feelings and values? (morally good or correct)	<i>Ethicality</i>
What is the participant's understanding of the intervention?	<i>Intervention coherence</i>
What benefits, profits or values must be given up to engage in the intervention, from the participant's point of view	<i>Opportunity costs</i>
What are chances of the intervention being able to cure or improve the condition being treated as perceived by the participant	<i>Perceived effectiveness</i>
To which extent the participant feels confident to be able to motivate oneself(ves) to participate and to adapt to the behavioral changes required by the intervention	<i>Self-efficacy</i>

*Table 1 Key themes based on components of theoretical framework of acceptability*



Interview identification code: .....

Initials: .....

*Demographic variables*

Age: .....

Parity: .....

Education: .....

Marital status: .....

Number of living children: .....

Country of origin: .....

*Variables on fetus/ neonate*

Fetal diagnosis: .....

Chosen treatment option: .....

Gestational age of the fetus/ neonate at interview X: .....wks.

Preference to receive study results yes/no

**References**

1. Rubin, H; Rubin I. Qualitative interviewing. The art of hearing data. Thousand Oaks: Sage; 2005.
2. Evers J. Het kwalitatieve interview: kenmerken, typen en voorbereiding. In: Evers J, editor. Kwalitatief interviewen: kunst en kunde. 2nd ed. Amsterdam: Boomlemma; 2015.
3. Yeo, A; Legard, R; Keegan, J; Ward, K; McNaughton Nicholls, C; Lewis J. In-depth interviews. In: Ritchie,J; Lewis, J; McNaughton Nicholls, C; Ormston R, editor. Qualitative Research Practice. Second. London: Sage; 2014.



Theme	Example quote
Ethicality (E)	Q1 "If it doesn't work, if he needs a shunt and he never walks, well at least we can say we did everything we could at the time that was available. And you can look him in the eyes and say, 'Look, we tried anyway.'" <i>Father12</i>
	Q2 "But it was kind of a progression thing, that first meeting was awful and you know that second meeting was more beneficial, we were talking to more specialized individuals, that's when we talked to the neurosurgeon and then the specialist in the delivery." <i>Father2</i>
Affective attitude (AA)	Q3 "But we've come to the decision now and it's not something that, yes, I'm terrified of it, but I wouldn't go back on it, at all. You know, and if in the morning, if they told me I couldn't do it, I think it would devastate me, because this is the decision we've come to." <i>Mother17</i>
Perceived effectiveness (PE)	Q4 "It sounds a bit strange, but psychologically for me, just to know every day you're kind of waking up waiting for the kicks, "Is that a kick? Is that a punch? [...] So if this back is closed from twenty-six weeks, just to be able to relax a little. I'm not saying I'm going to enjoy the pregnancy but just to know that, right, no more damage can be done, that's a big thing." <i>Mother12</i>
Burden (B)	Q5 "I think a couple of times we kind of went 'do we have enough [money]?' But I suppose in the long run, it's worth it. Yeah, I mean, as [mother] said, we've got family support, so if we do need money, they're willing to give it." <i>Father16</i>

Table 1 Prospective acceptability, example quotes

Theme	Example quote
Affective attitude (AA)	Q6 “Well, it's actually all about the children you've got and then the child that you're trying to protect at that time. So, there's not really a right one to go by for a mother.” <i>Mother24</i>
	Q7 “That's one time where – I forgot about that – that is the one time where I thought I was going to lose it is when she was in prenatal surgery and nobody told me that she was out of prenatal surgery.” <i>Father2</i>
Burden (B)	Q8 “And I think it's a good thing to have, to be honest, because I think it's healthy where I was just like, ‘why have I done this? I feel awful. I don't ever want to do this again. I never want — I'm just suffering so much, why put myself through this? Why couldn't I just let it go to natural causes and just see what happens?’ There was definitely that element there for a day, definitely, which was my lowest point.” <i>Mother10</i>
Affective attitude (AA)	Q9 “I could turn around to somebody now, honestly, hand on my heart, and say, ‘Do you know what? If I had to do it again, I would.” <i>Mother15</i>

Table 2 Concurrent acceptability, example quotes

Theme	Example quote
Self-efficacy (SE)	Q10 “Every time we were going for check-ups there was a different doctor checking her. And different doctors were putting different notes. And like towards the end, maybe a month before the c-section, there was a doctor and she wrote the abbreviation for natural birth: NB.” <i>Father22</i>
Affective attitude (AA)	Q11 “Because any delivery in general pregnancy is not easy, even if it’s normal. Every woman has something else. But this was – I’m not saying I’m the only one with a child with spina bifida with the surgery and everything, but it’s not a normal pregnancy, it’s hard on its own, you know.” <i>Mother22</i>
Burden (B)	Q12 “The week before I had her, I actually thought we were both going to die, I was in that much pain [...] Yeah, they underestimated how I felt. But it was that bad I didn’t think we were going to make it. Yeah, it was horrible.” <i>Mother11</i>
Ethicality (E)	Q13 “I wanted [to go] home because you felt like you were playing doctor as well as mummy, when really all you needed was to be playing mum with a wee bit of support. [...] I don’t know if it’s because his surgery was done [elsewhere] before birth, so when he was born, they just weren’t sure where he slotted in on the timeline of what should be happening when. [...] Because he was kind of nine weeks past his surgery, they were like, what do you do? What do you? Does he need all this?” <i>Mother15</i>
Perceived Effectiveness (PE)	Q14 “Yeah, I wouldn’t say I regret the surgery, I think it’s just disappointing that she does have the hip problem because we weren’t expecting it.” <i>Mother5</i>
Intervention coherence (IC)	Q15 “... we’ll just have to see, but because we’re so happy with her and we’re in a bubble, we don’t think there’s anything wrong so I guess if one day they said, you know, she’s going to need a wheelchair, she’ll need crutches, of course it will be heart-breaking but we – these are the good years because we do everything for her.” <i>Mother20</i>
Ethicality (E)	Q16 “Then it hits that you that at one point we were sort of heading towards her not being here. So that hits you quite often.” <i>Mother3</i>
	Q17 “They said to us, [at diagnosis], yes, your child is handicapped, it will be in a wheelchair and [euh] she may have brain damage, and those things. It was brought as a very dark outlook. [...] but now, if, in hindsight, if I think, no, how have I ever been able to question this decision [of fetal surgery]. I should not have questioned it...But it is a lack of knowledge.” <i>Mother19</i>
	Q18 “Then once she came out, obviously her legs were kicking and then she wee’d everywhere. They were like, ‘Oh, she’s not going to do any of this,’ and I was like, do you know what? Stuff the world. I said, ‘Look, she’s come out doing everything they said she wouldn’t, and their idea of an abortion is her.’” <i>Mother7</i>
Burden (B)	Q19 “Yes, it does, and then when everything starts to settle and you’ve had time in the house and time to actually think about things, you realise look what we’ve just been through. You know, it hits you then like a train and you’re like oh my goodness.” <i>Mother15</i>
	Q20 “It happened at the 20 weeks anomaly scan [diagnosis of spina bifida]. Then I really thought this baby is not going to come.....And then, after she was born, they called me to get down to the neonatal ward, again I thought [name of child] is going to die, you know? Those moments were just very intense....and at a certain moment....I envisioned that [first child] was in the water, drowned. All those things. Just an enormous fear to lose both my kids.” <i>Mother19</i>

Table 3 Retrospective acceptability, example quotes

	Maternal-fetal surgery		Termination of pregnancy	
	Mothers n (%)	Fathers n (%)	Mothers n (%)	Fathers n (%)
	24	22*	5	3
Age				
20-29 years	9 (38%)	4(18%)	1(20%)	-
30-39 years	15(62%)	14(64%)	3(60%)	3(100%)
>40	0 (0%)	4(18%)	1(20%)	-
Level of education				
Low	4(17%)	5(23%)	-	-
Intermediate	3(12%)	6(27%)	1(20%)	1(33%)
High	17(71%)	11(50%)	4(80%)	2(67%)
Parity				
Nulliparity	7(29%)		2(40%)	
Multiparity	17(71%)		3(60%)	
Marital status				
Partner	23(96%)		5(100%)	
Single	1(4%)		-	
Country of origin mother				
United Kingdom and Crown Dependencies	12(50%)		-	
United States of America	1(4%)		-	
Ireland	3(13%)		-	
The Netherlands	3(13%)		1(20%)	
France	1(4%)		1(20%)	
Croatia	1(4%)		-	
Hungary	1(4%)		-	
Hong Kong	1(4%)		-	
Finland	1(4%)		-	
Belgium	-		3(60%)	
Country currently resident				
United Kingdom and Crown Dependencies	15(63%)		-	
Ireland	5(21%)		-	
The Netherlands	4(17%)		1(20%)	
Belgium	-		4(80%)	
Treatment centre				
Belgium (UZ Leuven)	19(79%)		5(100%)	
United Kingdom (UCLH)	5(21%)		-	
Gestational age at birth				
<24 weeks			5(100%)	
25-28 weeks	1(4%)		-	
29-32 weeks	1(4%)		-	
33-36 weeks	13(54%)		-	
> 36	9(38%)		-	

Table 4 Socio-demographic characteristics of study participants (data from one father missing)

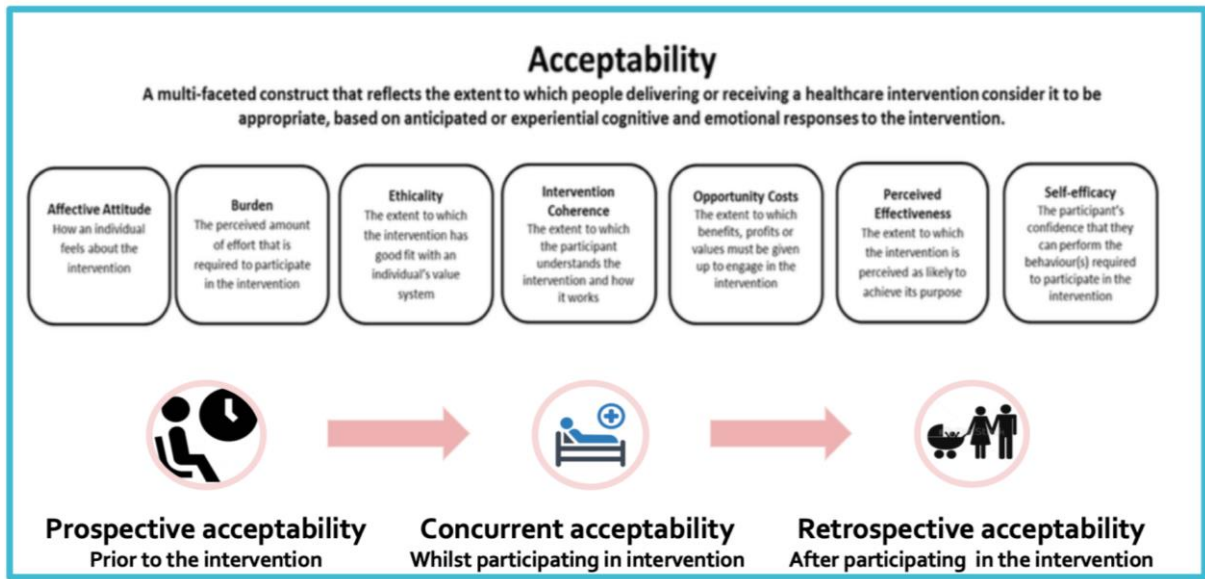


Figure 1 The framework of acceptability (adapted from the framework of acceptability by Sekhon et al<sup>12</sup>).

Figure 2 Identified themes of acceptability in temporal order, organized according to the Theoretical Framework of Acceptability

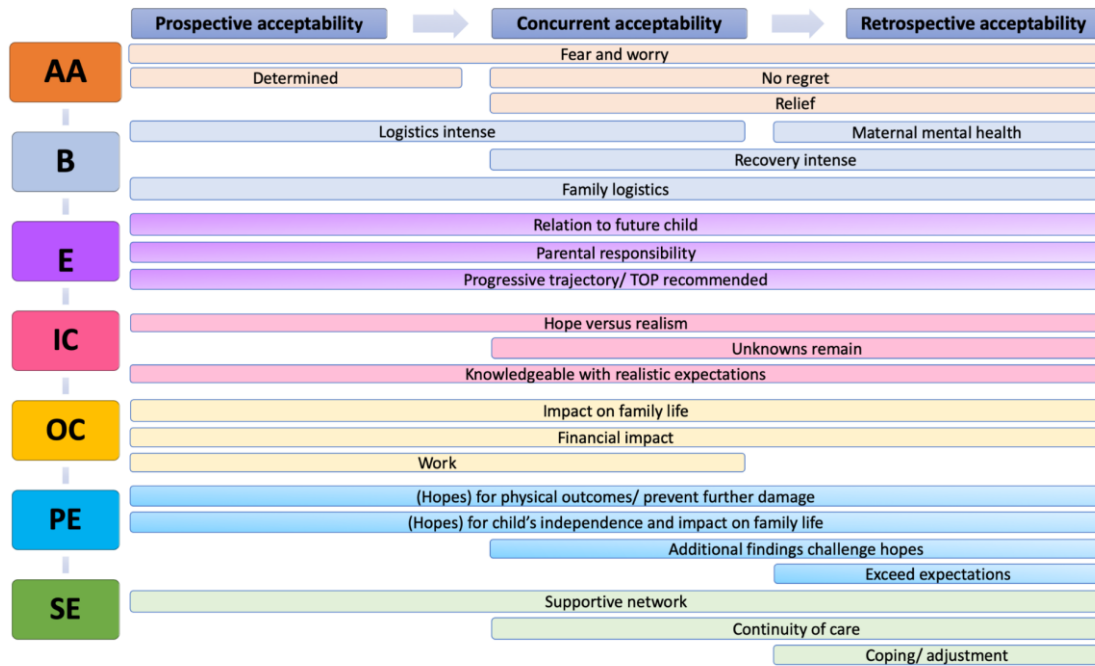


Figure 3 Assessed, eligible and included parents, per round of interviews (FS-fetal surgery; TOP-termination of pregnancy; EM-expectant management)

\* Unable to speak either Dutch or English

