A Study to Assess Knowledge and Acceptability of Fetal Surgery for Spina Bifida Amongst Healthcare Professionals in the UK

Running title: Acceptability of Fetal Surgery in the UK

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Abstract

Fetal surgery to repair open spina bifida before birth improves infant motor function and reduces ventriculoperitoneal shunt requirement. Alongside the development of the first UK fetal surgery service, we surveyed the knowledge and acceptability of this treatment to healthcare professionals in the UK and Ireland via an electronic questionnaire.

Ninety-eight (98) responses were received: MFM clinicians (21), midwives (20), paediatric neurosurgeons (17), obstetricians (15), neonatologists (13), theatre nurses (11), commissioners (1). Overall 70% of responders agreed with the concept that fetal surgery improved neonatal outcome in selected cases, although only 41% of paediatric neurosurgeons agreed. A variety of concerns were expressed, the most common being the lack information regarding mid-to long term effects on the child and mother.

In offering this new service it is important that we are cognisant of healthcare professional concerns and address them by applying internationally accepted criteria for fetal surgery, emphasising patient choice and collecting long-term data.

Keywords:

Spina bifida, fetal surgery, acceptability, opinions, healthcare professionals

Impact statement

What is already known on this subject?

Fetal surgery is a potentially controversial intervention, which is increasing in availability globally. Fetal surgery is now available in the UK on a charitable-funding basis. Prior to starting a UK fetal surgery service we assessed the attitudes and knowledge of healthcare professionals in the UK towards this new surgery.

What do the results of this study add?

Overall the majority of healthcare professionals agree with the concept that fetal surgery improves neonatal outcome in selected cases, but a variety of concerns exist, the most common being the lack of information regarding mid-to long term effects on the child and mother. Other concerns included a lack of education, training and research; the specific risk of preterm birth following surgery; the evidence base for this procedure; effects on maternal choice and financial implications.

What are the implications of these findings for clinical practice and/or further research?

Those developing this new service should be mindful of the concerns expressed and address them by applying internationally accepted criteria for fetal surgery, emphasising patient choice and collecting long-term data.

Introduction

Fetal surgery is a potentially controversial intervention, with many ethical and social aspects to be considered (Deprest *et al*., 2011). The Management of Myelomeningocele Study (MOMS) (Adzick *et al.,* 2011) published in 2011 provided level 1 evidence that closure of open spina bifida (myelomeningocele, MMC) before birth can reduce the requirement for ventriculoperitoneal shunt placement and improve motor function at 12 and 30 months of age. Following this, a questionnaire study (Antiel *et al.*, 2017) of 670 maternal-fetal medicine (MFM) physicians, neonatologists and paediatric surgeons in the US showed that the majority believed prenatal surgery for fetal spina bifida to be favoured over postnatal surgery. A minority of respondents indicated that they were now less likely to recommend termination of pregnancy, with the majority reporting no change. Attitudes varied according to specialty, with neonatologists and paediatric surgeons more likely to recommend prenatal surgery. The availability of fetal surgery has rapidly expanded across the globe, with the majority of units offering open fetal spina bifida repair surgery over fetoscopic repair (Sacco *et al.*, 2018).

Our unit has established a clinical service offering open fetal surgery for spina bifida for the first time in the UK and NHS England has recently announced plans to fund the surgery through its Specialist Commissioning Services. Acceptability is a key consideration in the implementation of new healthcare interventions such as fetal surgery, for both providers and receivers. “Buy in” from healthcare professionals referring, treating or caring for patients is necessary to ensure that the intervention is offered and delivered as intended (Sekhon *et al.,* 2017). Whilst establishing our open fetal surgery service in the UK, we undertook a national survey of healthcare practitioners to understand current attitudes and concerns towards this intervention and plans for implementation. We aimed to include healthcare professionals not directly involved in fetal surgery as we wanted to seek a wider opinion given that this was the first service of its kind in the UK. We also were aware that patients considering this option come into contact with many healthcare professionals other than MFM doctors and paediatric surgeons.

Methods

We designed an electronic questionnaire aimed at healthcare professionals in the UK to assess the respondents' background knowledge of spina bifida and fetal surgery, opinions about fetal surgery and concerns. The questionnaire was designed collaboratively between MFM and paediatric neurosurgeons taking into consideration the previous US questionnaire (Antiel *et al.*, 2017), and was piloted on members of our local team for construct validity and reliability before national distribution. We also collected demographic data and the respondents' level of training. In the questionnaire we described the scientific rationale for closure of MMC during fetal life, including earlier reversal of hindbrain herniation and the "two-hit" hypothesis - namely that the neurologic defects seen in spina bifida arise from both the primary neural tube defect and secondary in-utero damage following mechanical and chemical trauma to the exposed neural elements.

From October 2017 to March 2018 the questionnaire was distributed to healthcare professionals (obstetricians, maternal-fetal medicine clinicians and midwives, paediatric surgeons, neonatologists and theatre nurses) nationally through contacts (>120 participants) and via contacting the British Maternal Fetal Medicine Society (BMFMS), the British Association of Perinatal Medicine (BAPM) and by email via the British Paediatric Neurosurgery Group (BPNG). Responses were collected and analysed using questionnaire survey software (Google Forms, Alphabet, California, US).

For our sample size calculation we aimed to tolerate a 10% margin of error with a 95% certainty that our questionnaire results found the correct answer. We estimated that there was a population of around 2000 specialists from whom to choose our sample (1000 BAPM, 500 BMFMS, 400 BPNG, 100 contacts). We assumed that approximately two-thirds of respondents would agree that fetal surgery was acceptable, based on the previous US study (Antiel *et al.*, 2017) giving a recommended sample size of 82 participants. Assuming that only 50% of respondents would agree that fetal surgery was acceptable led to a recommended sample size of 92 participants.

Results

Demographics

Ninety-eight (98) responses were received. The survey was not directly sent to all professionals but posted on the websites of BMFMS and BAPM, so no denominator could be calculated. Demographics are shown in Table 1. The majority of responders were aged 41-60 years and from England. Maternal-fetal medicine clinicians, paediatric neurosurgeons, general obstetricians and neonatologists (all a mix of consultants and trainees), midwives and theatre staff (nurses and operating department practitioners), were the largest groups of respondents and the majority had over ten years' experience in their clinical role.

Knowledge and Opinions Regarding Spina Bifida

Results of questions pertaining to knowledge and opinions of responders regarding spina bifida are shown in Figure 1. Paediatric neurosurgeons were most likely to rate their knowledge of spina bifida as "expert"; MFM clinicians, obstetricians and neonatologists most commonly described their knowledge as "good" and midwives "limited". Paediatric neurosurgeons, MFM clinicians and neonatologists were most likely to be moderately or very familiar with managing patients with spina bifida; general obstetricians and midwives were "slightly" or "not at all" familiar with this. Most respondents agreed that spina bifida was associated with significant disability, and that babies having postnatal repair generally do not have a normal quality of life, although 19.4% of respondents were unsure regarding this latter point.

Knowledge and Opinions Regarding Fetal Surgery

Knowledge about fetal surgery was rated highest in MFM clinicians, with most other groups rating their knowledge as "limited" (Figure 2). Most respondents (72.4%) had no experience performing or counselling about fetal surgery. Familiarity with the MOMS trial was highest amongst MFM

clinicians and paediatric neurosurgeons and the “two-hit” hypothesis was generally well-known. Around 70% of respondents agreed with the concept that fetal surgery improved the outcome in selected cases, suggesting that this was generally acceptable as a treatment option, although this was lower in the group of paediatric neurosurgeons (41%).

Concerns Regarding Fetal Surgery

There were a variety of concerns raised regarding fetal surgery, the most common being the lack of mid-to long-term information regarding the effects on the child and mother. The ten commonest concerns are shown in Figure 3.

Free Text Responses

Respondents were given the opportunity to leave free text comments in the questionnaire. These are reported below, grouped by topic.

Preterm birth

- ‘Some improvement in outcomes must be balanced against the risks of prematurity.’ - MFM Consultant
- ‘[Lack of] evidence as to the relative benefits of fetal surgery versus the potential long-term complications of pre-term birth on baby and family and maternal complications.’ – Fetal Medicine Midwife
- ‘The risk of preterm delivery following fetal surgery with its consequent co-morbidities need to be highlighted.’ - Neonatologist

Insufficient evidence base regarding surgical outcomes

- ‘Several studies in Europe now suggest expectant management is associated with better outcomes than the experimental arm in MOMS. In that respect MOMS was unusual as outcomes in the control group were worse than we would expect.’ – MFM Consultant
- ‘I am aware of a small number of surgeons offering in utero surgery for MMC in Europe. I know from colleagues who have treated some of these in-utero repair cases post-op that serious complications from in-utero surgery go unreported, and this sends a misleading message to expectant parents regarding the safety of in-utero surgery. My concern about a small number of lone practitioners doing in-utero surgery is that they have an inherent bias towards the procedure and become zealous proponents.’ - Paediatric Neurosurgeon
- ‘I take issue fundamentally with the concept that surgical repair in-utero can reduce lifelong disability, versus postnatal repair.’ - Paediatric Neurosurgeon
- ‘There is no evidence anywhere that laparoscopic surgery for spina bifida improves outcomes.’ – MFM Consultant

Maternal choice and information

- ‘Mothers may feel compelled to have a treatment that could adversely affect all their future pregnancies and result in net harm over the mother’s reproductive life course.’ – MFM Consultant
- ‘I would love to see more research into this technique and am pleased we are going to offer it in the UK.’ – Fetal Medicine Midwife
- ‘If the existence of …… service caused even a small number of parents to choose not to terminate (and have a healthy replacement baby) the net harm would be considerable.’ – MFM Consultant
- ‘[There is] evidence of historical poor or less than optimal consent processes for reproductive technologies both in research and clinical practice i.e. consenting in one consultation, not ensuring time for consideration and clarification.’ – Fetal Medicine Midwife
- ‘Both maternal and neonatal outcomes must be balanced.’ - Fetal Medicine Midwife

Financial Implications

- ‘In an NHS struggling to resource basic care, while this is exciting stuff, is the balance of needs being appropriately considered?’ – MFM Consultant
- ‘[I am concerned about] distribution of finances in difficult economic times.’ - General Obstetrician

Discussion

In this study we assessed the knowledge of and attitudes of healthcare professionals in the UK to fetal surgery for spina bifida. The level of knowledge and experience of spina bifida and fetal surgery was relatively high, reflecting the expertise of the professional groups that we surveyed. We targeted a range of healthcare workers who were likely to come into contact with women whose fetus has spina bifida and who might be considering fetal surgery. Familiarity with the "two-hit hypothesis" and the MOMS trial was also high and in general over two-thirds of respondents agreed with the concept that fetal surgery improved outcomes in selected cases. This is in keeping with a US study of healthcare worker acceptability for fetal surgery following publication of the MOMS trial (Antiel *et al.*, 2017). Fetal surgery acceptability was lower in paediatric neurosurgeons. It is possible that this is because they have more experience of the standard postnatal closure technique. There were concerns expressed regarding fetal surgery, particularly about the safety and efficacy long term for both the mother and her child, the risk of preterm birth, the implications for maternal choice and cost.

A strength of this study is that we sought opinions from a range of healthcare professionals involved in the care of patients considering fetal surgery, such as neonatologists and theatre staff, rather than concentrating only on MFM clinicians or paediatric neurosurgeons. We did this prior to implementation in order to use the information clinically in planning our service. As with any survey, responder bias is a limitation, as well as response rate. We tried to reduce selection bias for participants through directly contacting a large number (>120) and by approaching the remainder via society membership, but we cannot exclude this possibility. Although we received between 11 and 20 responses per healthcare group, we believe that the opinions expressed are likely to be similar to those held by other healthcare professionals as the same themes came up both within and between groups.

A recent transcript of a scientific conference of patients and healthcare professionals convened by SHINE, a UK spina bifida charity, had similar consensus findings after a review of the literature

concerning fetal surgery for spina bifida (Shanmuganathan *et al.*, 2018). Attendees agreed that further data was required regarding long-term neonatal outcomes following open prenatal surgery (currently being collected as part of the MOMS2 trial), and the long-term maternal outcome including the impact on future pregnancies. A recent study showed no impact of open fetal surgery on future fertility (Wilson *et al.*, 2010). There was also a call for standardised reporting of postnatal surgery outcomes to allow comparison with prenatal surgery. Finally there was agreement that pregnant women whose fetus is known to have spina bifida should be counselled that prenatal surgery is an option in specific circumstances.

The legality and ethics of fetal surgery, particularly regarding the status of the mother and the fetus, have been debated extensively (Rodrigues *et al.*, 2013) (Cao *et al.*, 2018) (Chervenak and McCullough, 2017). Some countries have now adopted guidelines to support providing information to parents on the option of prenatal MMC repair and prognosis if there are no maternal or fetal contraindications for prenatal repair at 20–26 weeks' gestation (Wilson *et al.*, 2014).

The findings and concerns expressed in this study and the above conference have guided the establishment of the first open fetal surgery clinical service in the UK, and have informed the recently approved NHS Specialist Commissioning Service specification. Collecting data on both short and long-term outcomes is an absolute requisite to enable the quality of a new clinical service to be evaluated. This will require outcome data to be provided not only by MFM clinicians and neonatologists who manage the care of the mother and neonate in the short term, but also long term engagement from the paediatric neurosurgical and neurological teams who manage the child growing up. As well as careful data collection, further work should explore acceptability to patients and family which we are currently exploring in our patient population.

As knowledge about fetal surgery for spina bifida increases amongst patients and healthcare professionals, aided by media reports and social media, the provision of a UK clinical service in a strictly regulated and transparent manner will provide NHS patients with the best possible care.

Conclusion

This study highlights the opinions of healthcare professionals in the UK regarding fetal surgery for spina bifida. The majority of respondents agree with the concept of fetal surgery but have concerns, particularly regarding long-term effects. In offering a new clinical service it is important that healthcare professionals are cognisant of existing concerns and address them as far as possible by applying internationally accepted criteria for fetal surgery, emphasising patient choice and collecting long-term data to evaluate outcomes.

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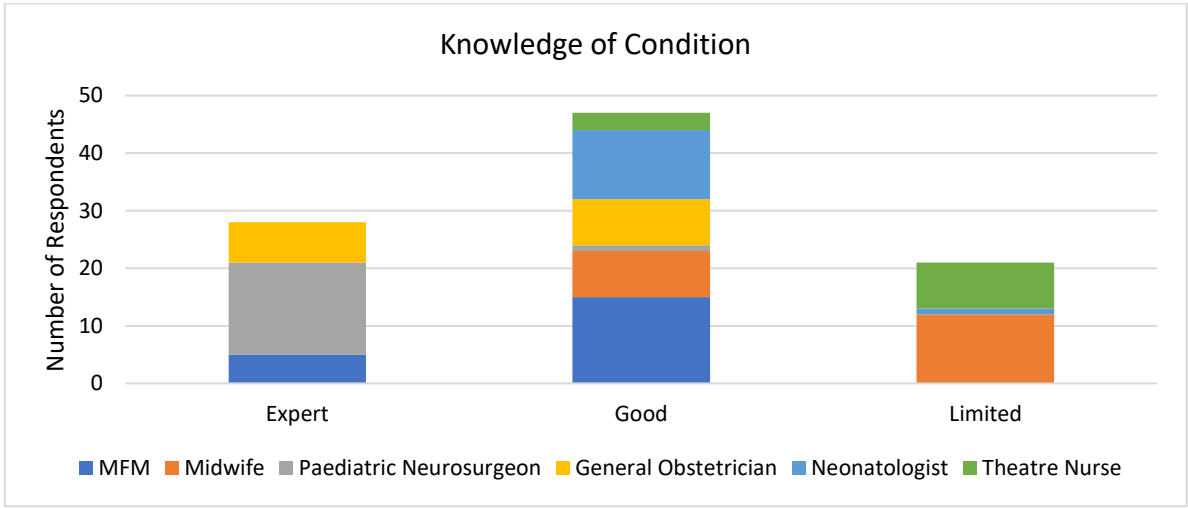
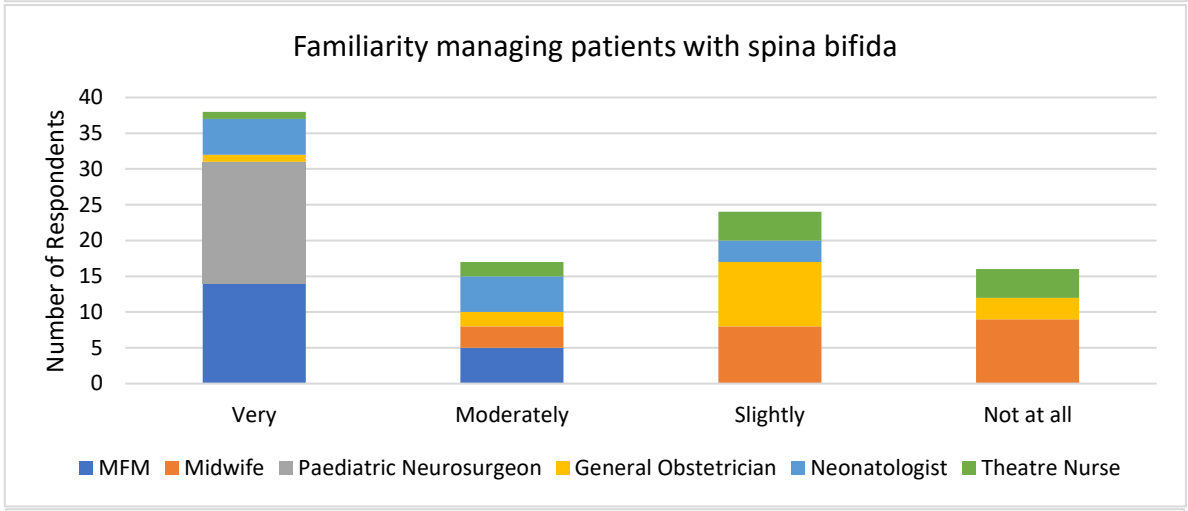
Declaration of Interest

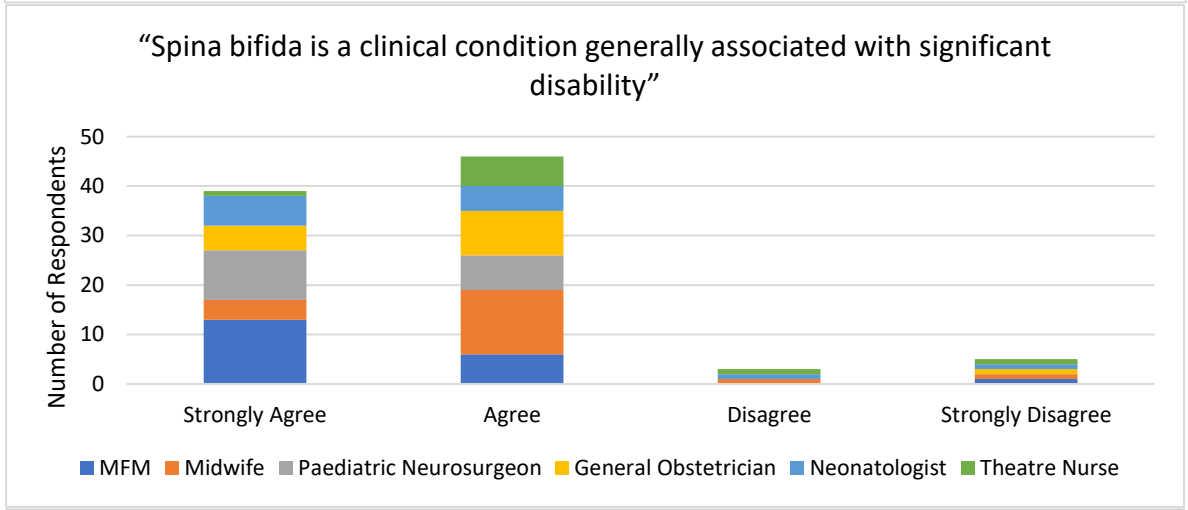
All authors report no conflicts of interest.

		Number (n = 98)	Percentage (%)
Gender	Female	56	57.1
	Male	40	40.8
	Unanswered	2	2.0
Age (years)	26-30	7	7.1
	31-40	21	21.4
	41-50	32	32.7
	51-60	37	37.8
	61-70	0	0
	Unanswered	1	1.0
Professional Role	Maternal Fetal Medicine Clinician	21	21.4
	Midwife (Fetal Medicine)	20	20.4
	Paediatric Neurosurgeon	17	17.3
	Obstetrician (general)	15	15.3
	Neonatologist	13	13.3
	Theatre/ Operating Department Nurse	11	11.2
	NHS Commissioner	1	1.0
Years of Experience in Role	0-2	8	8.2
	2-5	17	17.3
	5-10	21	21.4
	>10	51	52.0
	Unanswered	1	1.0
Country of Practice	England	80	81.6
	Channel Islands	6	6.1
	Republic of Ireland	4	4.1
	Scotland	4	4.1
	Wales	4	4.1

Table 1: Demographics of Questionnaire Respondents







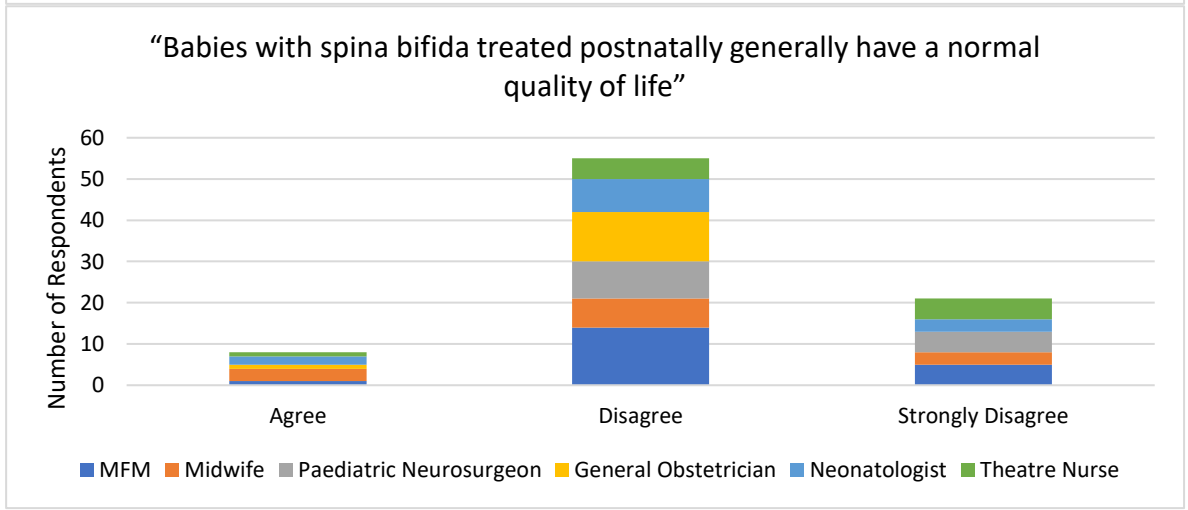


Figure 1: Knowledge and Opinions Regarding Spina Bifida

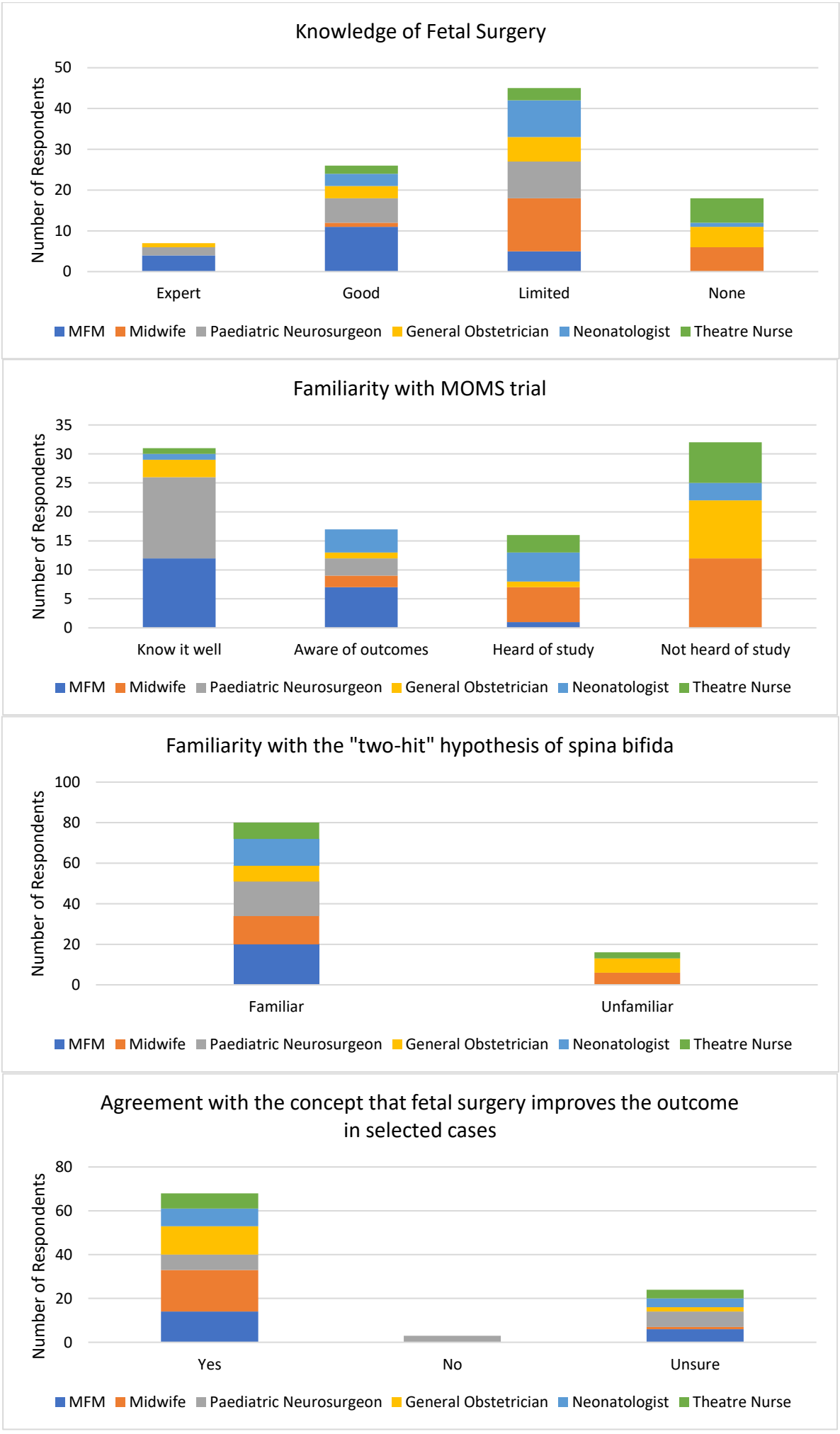


Figure 2: Knowledge and Opinions Regarding Fetal Surgery

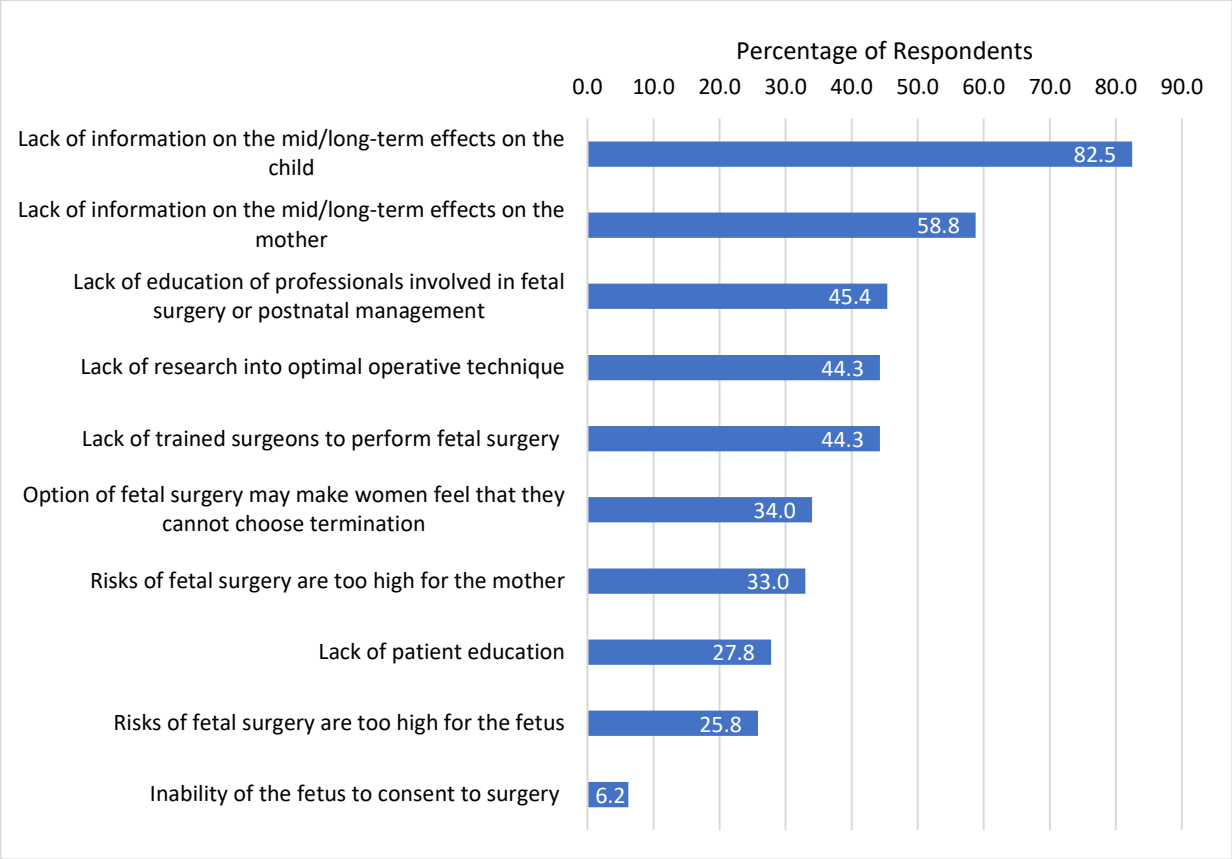


Figure 3: Respondents Concerns Regarding Fetal Surgery

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