Neurosurgeons' opinions on the prenatal management of myelomeningocele

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OBJECTIVE Improvements in imaging and surgical technological innovations have led to the increasing implementation of fetal surgical techniques. Open fetal surgery has demonstrated more favorable clinical outcomes in children born with open myelomeningocele (MMC) than those following postnatal repair. However, primarily because of maternal risks but also because of fetal risks, fetal surgery for MMC remains controversial. Here, the authors evaluated the contemporary management of MMC in the hope of identifying barriers and facilitators for neurosurgeons in providing fetal surgery for MMC.

METHODS An online survey was emailed to members of the Congress of Neurological Surgeons (CNS) and the International Society for Pediatric Neurosurgery (ISPN) in March 2019. The survey focused on 1) characteristics of the respondents, 2) the practice of counseling on and managing prenatally diagnosed MMC, and 3) barriers, facilitators, and expectations of fetal surgery for MMC. Reminders were sent to improve the response rate.

RESULTS A total of 446 respondents filled out the survey, most (59.2%) of whom specialized in pediatric neurosurgery. The respondents repaired an average of 9.6 MMC defects per year, regardless of technique. Regardless of the departments in which respondents were employed, 91.0% provided postnatal repair of MMC, 13.0% open fetal repair, and 4.9% fetoscopic repair. According to the surgeons, the most important objections to performing open fetal surgery were a lack of cases available to become proficient in the technique (33.8%), the risk of maternal complications (23.6%), and concern for fetal complications (15.2%). The most important facilitators according to advocates of prenatal closure are a decreased rate of shunt dependency (37.8%), a decreased rate of hindbrain herniation (27.0%), and an improved rate of motor function (18.9%). Of the respondents, only 16.9% agreed that open fetal surgery should be the standard of care.

CONCLUSIONS The survey results showed diversity in the management of patients with MMC. In addition, significant diversity remains regarding fetal surgery for MMC closure. Despite the apparent benefits of open fetal surgery in selected pregnancies, only a minority of centers and providers offer this technique. As a more technically demanding technique that requires multidisciplinary effort with less well-established long-term outcomes, fetoscopic surgery may face similar limited implementation, although the surgery may pose fewer maternal risks than open fetal surgery. Centralization of prenatal treatment to tertiary care referral centers, as well as the use of sophisticated training models, may help to augment the most commonly cited objection to the implementation of prenatal closure, which is the overall limited caseload.

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KEYWORDS spina bifida; survey; fetal surgery; meningomyelocele

ABBREVIATIONS CNS = Congress of Neurological Surgeons; ISPN = International Society for Pediatric Neurosurgery; MMC = myelomeningocele; MOMS = Management of Myelomeningocele Study.

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MYELOMENINGOCELE (MMC), characterized by extrusion of the spinal cord together with the meningeal membranes, is the most severe and common form of spina bifida.¹¹ Standard treatment is closure of the MMC defect within 48 hours of birth to minimize infectious and traumatic risk to the tissue.^{14,16} Since publication of the first case of surgical closure of an MMC defect in 1892, only limited progress has been made in surgical repair with the primary goals of creating a watertight closure over the open neural placode.⁶ Patients born with MMC can face lifelong disabilities such as neurological deficits below the level of the defect, hydrocephalus, hindbrain herniation, scoliosis, tethering of the spinal cord, urinary and fecal incontinence, and sexual dysfunction.

Early in vivo studies suggested that prenatal intervention could prevent secondary insults to the neural placode and the development of hindbrain herniation or hydrocephalus due to persistent intrauterine CSF leakage. Further surgical innovations and improvements in instrumentation refined this technique and improved mortality and morbidity rates.^{1,25} Initially, fetal surgery served as an option to treat potentially lethal conditions such as congenital diaphragmatic hernia, improving early mortality rates.^{19,28} With increasing knowledge and experience, the indications for fetal surgery have broadened toward treating nonlethal diseases such as MMC.^{1,8} At the end of the last century, early case studies of fetal MMC repair showed promising results and eventually led to a randomized controlled trial published in 2011.

In the Management of Myelomeningocele Study (MOMS), women diagnosed with a fetus having MMC were randomized to undergo either open fetal surgery before 26 weeks of gestation or conventional postnatal surgical repair.² This trial was stopped prematurely given the efficacy of prenatal surgery. Results showed that only 40% of the prenatal surgery group needed placement of a shunt versus 82% in the postnatal surgery group at 1 year. In addition, children with MMC in the prenatal surgery group were less likely to have hindbrain herniation and were more likely to have a better level of motor functioning and to walk independently. However, these promising fetal outcomes came at the expense of maternal morbidity with higher rates of preterm delivery and a higher risk of maternal complications such as placental abruption and chorioamniotic membrane separation. Fetal surgery also carries risks for the fetus such as the need for transfusions and a lower birth weight. Furthermore, the MOMS had strict inclusion criteria, reflected by the fact that of the 1087 women who had undergone preliminary screening, only 183 underwent randomization.

Fetoscopic techniques were developed in the hopes of lowering maternal morbidity both in the current pregnancy and in future pregnancies. Developments and refinements have further improved fetoscopic techniques.^{9,22} Unlike open prenatal repair, fetoscopic surgery was less well received by the neurosurgical community because of the complexity of the procedure and less-established outcomes.^{12,13}

With the rise of fetal surgery and the increasing evidence of its risks and benefits in treating selected pregnancies with MMC, neurosurgeons may increasingly face ethical dilemmas, such as weighing maternal risks against

potential fetal benefits. An expected paradigm shift in the treatment of MMC since publication of the MOMS results has led to an interest in knowing about the current management and application of fetal surgery by our colleagues. We aimed to identify the barriers and facilitators of fetal closure perceived by the neurosurgeons providing current techniques for MMC repair. Therefore, we conducted a survey on the management of MMC by neurosurgeons to evaluate the clinical practice worldwide.

Methods

Based on the literature, a first concept survey was developed.^{2–4,15,18,27} This survey was reviewed by two pediatric neurosurgeons experienced in treating MMC as well as a maternal-fetal medicine specialist before being finalized. The survey consists of 17 questions and can be divided into three sections: 1) characteristics of the respondents, such as subspecialization, tenure, and country of employment; 2) the practice of counseling on and managing MMC, such as the items discussed during counseling of parents, number of MMC defects closed yearly, and the availability of prenatal surgery to treat MMC; and 3) barriers, facilitators, and expectations of fetal surgery for MMC. The survey can be found in the Supplemental Material: Survey. The institutional review board waived the need for study approval as this research does not involve patients.

Members of the Congress of Neurological Surgeons (CNS) and the International Society for Pediatric Neurosurgery (ISPN) were approached by an email invitation containing a cover letter asking them to fill out a survey. The CNS was founded in 1951 and represents more than 9000 international members, who include residents, neurosurgeons, and medical students, among others. The ISPN consists of more than 350 pediatric neurosurgeons with the aim of improving the neurosurgical care of children. In March 2019 the survey was distributed by email, using SurveyMonkey. To increase the response rate, reminders were sent. By filling out a survey, respondents gave informed consent. Only responses from neurosurgeons and neurosurgery residents were included.

Data were analyzed using the IBM SPSS Statistics, version 21.0 for Windows (IBM Corp.). Descriptive statistics were used to present data in frequencies and percentages. Categorical data were analyzed using the chi-square test. A *p* value < 0.05 was considered to be statistically significant.

Results

Characteristics of the Respondents

A total of 448 respondents filled out the survey. Two responses were not from neurosurgeons or neurosurgery residents and were therefore excluded. Eighty-four percent of the respondents were from the CNS, while the remaining 16% were from the ISPN. Respondents were employed in a total of 58 countries, with the US (56.3%), Brazil (5.2%), and India (4.9%) having the most respondents (Fig. 1). Africa was the least represented (2.7%; Table 1).

Respondents had a mean clinical experience of 21.6 ± 12.9 years (mean ± standard deviation), with residents

FIG. 1. A geographic overview of the working locations of the respondents. Countries marked with a *star* have one or more respondents providing prenatal closure techniques. Created with mapchart.net©. CC BY-SA 4.0 (<https://creativecommons.org/licenses/by-sa/4.0/>).

having a mean clinical experience of 5.5 years (Table 1). Most of the respondents had pediatric neurosurgery as their subspecialization (59.2%). Spine (29.6%) and neurooncology (28.3%) followed as the most frequent subspecializations.

Among the respondents, 81.0% treated patients with MMC. Neurosurgeons subspecialized in pediatric neurosurgery were more likely to treat patients with MMC ($p < 0.001$). Among neurosurgeons not subspecialized in pediatric neurosurgery, 54.8% treated patients with MMC. Neurosurgeons performing closure of MMC defects operated on a mean of 9.6 ± 17.9 patients annually.

Counseling and Management of MMC

Three-quarters (74.5%) of the neurosurgeons and 50% of the residents deemed themselves sufficiently informed to counsel parents ($p = 0.008$), and 72.2% of the respondents actually counsel potential parents with children suspected to have MMC. Of those neurosurgeons who deemed themselves insufficiently informed to counsel, 37.2% still counsel potential parents. Table 2 gives an overview of several items discussed by respondents during the counseling of potential parents. All items differed statistically significantly between pediatric and nonpediatric neurosurgeons. Surgical closure of the MMC defect, the need for CSF shunting, and the expected level of motor function were always discussed by 83.1%, 75.3% and 69.4% of respondents, respectively. Termination of the pregnancy and the option of fetal surgery were never discussed by 35.6% and 23.7% of the neurosurgeons, respectively.

TABLE 1. Characteristics of the 446 respondents

Variable	No. (%)
Function	
Neurosurgeon	394 (88.3%)
Neurosurgeon in training	52 (11.7%)
Specialty	
Epilepsy	39 (8.7%)
Functional	28 (6.3%)
Peripheral nerve	25 (5.6%)
Pediatrics	264 (59.2%)
Neurooncology	126 (28.3%)
Neurovascular	83 (18.6%)
Neurotrauma	93 (20.9%)
Spine	132 (29.6%)
Other	47 (10.5%)
Yrs of clinical experience	21.6 ± 12.9
Continent of respondent	
Africa	12 (2.7%)
Asia & Oceania	67 (15.0%)
Europe	45 (10.1%)
North America	280 (62.8%)
South America	42 (9.4%)
Department provides	
Postnatal surgical closure of MMC	406 (91.0%)
Open fetal surgery	58 (13.0%)
Fetoscopic surgery	22 (4.9%)

TABLE 2. Items discussed during counseling by pediatric versus nonpediatric neurosurgeon respondents

Item	Pediatric Neurosurgeons	Nonpediatric Neurosurgeons	p Value
No. of respondents	265	181	
Termination of pregnancy			<0.001
Always	22.0%	12.2%	
Often	8.2%	3.8%	
Sometimes	17.1%	13.7%	
Rarely	25.7%	18.3%	
Never	26.9%	51.9%	
Repair of defect			<0.001
Always	92.7%	65.7%	
Often	4.1%	10.4%	
Sometimes	1.2%	4.5%	
Rarely	0.4%	3.0%	
Never	1.6%	16.4%	
Need for CSF shunting			<0.001
Always	85.4%	57.0%	
Often	11.4%	18.5%	
Sometimes	2.0%	6.7%	
Rarely	0	2.2%	
Never	1.2%	15.6%	
Option of fetal surgery			<0.001
Always	43.4%	15.2%	
Often	13.1%	10.6%	
Sometimes	14.8%	17.4%	
Rarely	13.5%	17.4%	
Never	15.2%	39.4%	
Hindbrain herniation			<0.001
Always	66.9%	36.6%	
Often	18.4%	15.7%	
Sometimes	9.0%	17.2%	
Rarely	3.7%	11.9%	
Never	2.0%	18.7%	
Risk of spinal cord untethering			<0.001
Always	68.2%	44.8%	
Often	15.9%	20.1%	
Sometimes	10.2%	14.2%	
Rarely	4.1%	3.7%	
Never	1.6%	17.2%	
Expected level of motor function			<0.001
Always	76.8%	55.6%	
Often	15.9%	17.3%	
Sometimes	3.7%	7.5%	
Rarely	0.8%	2.3%	
Never	2.8%	17.3%	
Urological management			<0.001
Always	80.1%	48.9%	
Often	11.4%	16.5%	
Sometimes	4.5%	12.0%	
Rarely	1.2%	3.0%	
Never	2.8%	19.5%	

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TABLE 2. Items discussed during counseling by pediatric versus nonpediatric neurosurgeon respondents

Item	Pediatric Neurosurgeons	Nonpediatric Neurosurgeons	p Value
Orthopedic management			<0.001
Always	58.4%	31.6%	
Often	22.9%	18.0%	
Sometimes	11.4%	22.6%	
Rarely	4.5%	6.8%	
Never	2.9%	21.1%	

Ninety-one percent of the departments in which the neurosurgeons are employed provide conventional postnatal surgical closure of the MMC defect. Open fetal and fetoscopic procedures are provided by 13.0% and 4.9% of the centers, respectively. Four out of all centers provided fetal surgery but did not perform conventional postnatal surgery. Performing more surgical closures annually was not associated with providing fetal surgery ($p > 0.05$).

Table 3 gives an overview of the disciplines involved in the treatment of (unborn) patients with MMC. As regards clinical management by the respondents, neurosurgery (78.0%), obstetrics (77.4%), and pediatrics (59.9%) were always involved in the care of MMC patients. Rehabilitation medicine, orthopedics, and neurology were never involved according to 27.5%, 26.6%, and 23.4% of respondents, respectively. Pediatric neurosurgeons were less likely than nonpediatric neurosurgeons ($p = 0.002$) to involve pediatrics.

Barriers, Facilitators, and Expectations of Fetal Surgery for MMC

When asked to rank the most important objections to providing open fetal surgery to treat MMC, 33.8% ranked “too few cases available to become proficient in the technique” as number one (Fig. 2). The risk of maternal complications and the risk of fetal complications were ranked second and third, respectively, by 23.6% and 15.2%. Costs and ethical concerns were ranked last by 32.4% and 27.0%, respectively.

The most important reason to provide open fetal surgery according to respondents was a decreased rate of shunt dependency (37.8% of respondents; Fig. 3), followed by a decreased rate of hindbrain herniation (27.0% of respondents) and an improved rate of motor function (18.9% of respondents). Ranked last was the motivation to give parents a treatment option during gestation (18.2% of respondents).

When comparing postnatal surgery with open fetal surgery and fetoscopic surgery, respondents expected the risk of maternal complications to be the highest for open fetal surgery (89.2% of respondents), closely followed by fetoscopic surgery (79.9% of respondents; Table 4). Conventional postnatal closure was expected to have the lowest risk (92.5% of respondents). Open fetal surgery (70.6% of respondents) and fetoscopic surgery (66.8% of respondents) were expected to have the highest risks for perioper-

ative fetal complications. No significant differences could be found in the expectations between countries providing fetal surgery and countries that do not. Furthermore, no significant differences could be identified between residents and neurosurgeons regarding barriers, facilitators, and expectations of fetal surgery (Fig. 3).

Table 5 gives an overview of respondent answers regarding the outcomes of open prenatal surgery. Among all respondents, 62.6% assumed that fetal surgery would lead to a higher risk of neonatal death, which contrasts with what the MOMS showed.

Table 6 gives an overview of respondent opinions regarding the position of fetal and fetoscopic surgery in the management of MMC. More than half of the respondents (54.4%) disagreed or strongly disagreed that open fetal surgery should be the standard of care. Similarly, more than half of the respondents (58.1%) disagreed or strongly disagreed that potentially damaging women as a result of fetal surgery is acceptable. Pediatric neurosurgeons were more likely than nonpediatric neurosurgeons to disagree on both of these items ($p < 0.001$ and $p = 0.003$, respectively). Reactions to the statement “denying potential improvement in the quality of life of MMC patients by not offering fetal surgery is acceptable” were more divided with almost a third (strongly) agreeing, a third being neutral, and almost a third (strongly) disagreeing. Slightly more than half (50.7%) of the respondents strongly disagreed or disagreed with the statement that providing fetoscopic surgery without performing a randomized controlled trial is acceptable.

Discussion

This study gives an overview of the current management of MMC and opinions on fetal surgery by neurosurgeons worldwide. Open fetal surgery (13.0%) and especially fetoscopic surgery (4.9%) are provided by only a minority of the centers in which the respondents are employed. Despite the well-documented decreased rates of shunt dependency and hindbrain herniation and improved rate of motor function in strictly selected pregnancies, only 16.9% of respondents agreed that open fetal surgery should be the standard of care. The most important objections to the procedure are a limited caseload available to become proficient in the technique, the risk of maternal complications, and the risk of fetal complications. The bind in the trade-off between maternal health and the health and quality of life of patients with MMC is illus-

TABLE 3. Disciplines involved in the management of MMC

Discipline	Pediatric Neurosurgeons	Nonpediatric Neurosurgeons	p Value
Pediatrics			0.002
Always	55.6%	71.3%	
Often	12.5%	10.3%	
Sometimes	8.6%	6.9%	
Rarely	16.8%	1.1%	
Never	6.5%	10.3%	
Genetics			NS
Always	22.6%	25.3%	
Often	23.9%	26.6%	
Sometimes	27.4%	22.8%	
Rarely	16.1%	10.1%	
Never	10.0%	15.2%	
Neurosurgery			NS
Always	78.4%	77.0%	
Often	15.2%	8.0%	
Sometimes	4.0%	6.9%	
Rarely	1.6%	4.6%	
Never	0.8%	3.4%	
Orthopedics			NS
Always	21.7%	16.7%	
Often	14.7%	11.1%	
Sometimes	14.3%	25.0%	
Rarely	23.5%	18.1%	
Never	25.8%	29.2%	
Neurology			NS
Always	17.0%	9.6%	
Often	18.8%	17.8%	
Sometimes	20.6%	35.6%	
Rarely	20.6%	12.3%	
Never	22.9%	24.7%	
Obstetrics			NS
Always	78.5%	75.0%	
Often	9.8%	8.3%	
Sometimes	4.9%	9.5%	
Rarely	4.5%	3.6%	
Never	2.4%	3.6%	
Rehabilitation medicine			0.001
Always	33.5%	31.6%	
Often	9.3%	18.4%	
Sometimes	8.4%	21.1%	
Rarely	19.5%	6.6%	
Never	29.3%	22.4%	
Social work			NS
Always	38.7%	46.2%	
Often	24.8%	17.9%	
Sometimes	12.2%	9.0%	
Rarely	12.2%	10.3%	
Never	12.2%	16.7%	

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TABLE 3. Disciplines involved in the management of MMC

Discipline	Pediatric Neurosurgeons	Nonpediatric Neurosurgeons	p Value
Urology			NS
Always	33.2%	32.9%	
Often	13.2%	16.5%	
Sometimes	12.3%	19.0%	
Rarely	20.5%	8.9%	
Never	20.9%	22.8%	

NS = not significant.

trated by the equally divided opinions regarding the statement that “denying potential improvement in the quality of life of MMC patients by not offering fetal surgery is acceptable.”

Comparison With Other Studies

When the results of the MOMS were published in 2011, a paradigm shift was expected.² However, such a shift was not reflected in our results: 23.7% of respondents never discuss the option of fetal surgery during counseling, and only 16.9% agree that open fetal surgery should be standard care. Moreover, only 13.0% of respondents offer open fetal surgery, while 4.9% offer fetoscopic surgery. An explanation for these low rates may be the generalizability of the MOMS, which included pregnancies carrying fetuses with MMC based on strict inclusion criteria. Results of fetal surgery for other pregnancies are less well-documented and warrant prospective studies. Another explanation may be the maternal comorbidity and the risk for future pregnancies. In the literature, a maternal complication rate of 6.2% for fetoscopic surgery and a rate of 20.9% for open

fetal surgery have been reported.²⁹ When taking into account only serious maternal complications such as those requiring ICU care or surgical intervention, risk rates of 1.7% and 4.5% have been reported for fetoscopic and open procedures, respectively.

Our results showed that 62.6% of the respondents assume that open fetal surgery carries a higher risk of neonatal death. Moreover, almost 40% wrongly assume a higher maternal mortality rate. These data may imply that a factor contributing to the nonoccurrence of the paradigm shift may be a lack of knowledge—on the one hand, a lack of knowledge because the outcomes reported in the literature may not be known among respondents, and on the other hand, because the results reported in the literature (e.g., MOMS) may not be generalizable to all pregnancies carrying fetuses with MMC.

Previous studies have shown that fetal surgery, especially fetoscopic surgery, is technically demanding and requires dedicated training.^{12,13,20} This may be reflected by the fact that only 4.9% of the departments surveyed provide fetoscopic surgery. A number of studies have been conduct-

FIG. 2. The most important objections to providing open fetal surgery for the treatment of MMC. The y-axis represents the percentage of respondents. The x-axis represents the rank chosen for each objection.

FIG. 3. The most important reasons to provide open fetal surgery for MMC. The y-axis represents the percentage of respondents. The x-axis represents the rank chosen for each expectation.

ed to assess the relationship between hospital and surgeon volume and surgical mortality and morbidity.^{7,24} These studies have shown that a high surgeon volume results in superior outcomes for patients. Paradoxically, the majority of our respondents also agree that performing only one case of fetal surgery per month is acceptable, which mirrors the respondents' current practice of approximately 9.6 MMC repairs per year.

In light of the high case volume, the Maternal-Fetal Management Task Force released a position statement in which minimum criteria are recommended to ensure optimal maternal and fetal outcomes.¹⁰ In this statement, an initial experience of 5 cases is required, as is an adequate annual volume of cases to maintain proficiency. The exact number of cases meant by "adequate" is subject to debate,

however. A recent meta-analysis on the learning curves of fetal surgery estimated that competence for standard hysterotomy is reached after 35 cases and that competence for minimally invasive procedures is reached after 56 cases.²⁰

In this context, a smaller number of centers dedicated to fetal surgery, in which surgeons perform larger volumes of cases, is more desirable than a more disseminated offering of fetal surgery. A remedy toward achieving and maintaining proficiency may be adequate training models.²⁶ Assisted nowadays by 3D-printed models, it is possible to create case-specific models to prepare for complex procedures such as fetoscopic surgery.

TABLE 4. Expectations regarding the risks of maternal and fetal complications of three techniques to close MMCs

Complication & Procedure	Level of Risk		
	Most	Moderate	Least
Maternal complication			
Postnatal surgery	2.7%	4.8%	92.5%
Open fetal surgery	89.2%	8.8%	2.0%
Fetoscopic surgery	79.9%	13.6%	9.5%
Fetal complication			
Postnatal surgery	16.3%	21.4%	62.2%
Open fetal surgery	70.6%	19.9%	9.5%
Fetoscopic surgery	66.8%	22.7%	10.5%

TABLE 5. Respondent knowledge about open prenatal surgery performed before 26 weeks of gestation

Statement	Respondent Answer	
	True	False
Fetal surgery leads to a higher risk of maternal death	39.4%	60.6%
Fetal surgery leads to a higher risk of maternal complications such as placental abruption	90.6%	9.4%
Fetal surgery leads to a higher risk of neonatal death	62.6%	37.4%
Fetal surgery halves the neonatal CSF shunt rate	68.2%	31.8%
Children who underwent prenatal surgery have a lower level of mental & motor function	11.5%	88.5%

Answers in boldface type indicate results of the MOMS.

TABLE 6. Overview of respondent opinions regarding the place of fetal surgery in the management of MMC

Statement	Pediatric Neurosurgeons	Nonpediatric Neurosurgeons	p Value
Providing prenatal fetoscopic surgery w/o performing an RCT is acceptable			NS
Strongly agree	5.5%	8.3%	
Agree	14.0%	18.8%	
Neutral	25.5%	31.3%	
Disagree	34.0%	28.1%	
Strongly disagree	21.0%	13.5%	
Potentially damaging women due to fetal surgery is acceptable			0.003
Strongly agree	2.5%	5.2%	
Agree	17.5%	11.5%	
Neutral	17.5%	34.4%	
Disagree	37.5%	36.5%	
Strongly disagree	25.0%	12.5%	
Open fetal surgery should be standard care			<0.001
Strongly agree	5.0%	4.2%	
Agree	12.0%	12.5%	
Neutral	21.5%	43.8%	
Disagree	36.0%	27.1%	
Strongly disagree	25.5%	12.5%	
A neurosurgeon performing only 1 fetal surgery case/mo is acceptable			0.006
Strongly agree	25.0%	10.5%	
Agree	43.0%	37.9%	
Neutral	13.5%	25.3%	
Disagree	10.0%	15.8%	
Strongly disagree	8.5%	10.5%	
Denying potential improvement in QOL of MMC patient by not offering fetal surgery is acceptable			NS
Strongly agree	9.5%	7.3%	
Agree	23.5%	14.6%	
Neutral	31.0%	41.7%	
Disagree	22.0%	27.1%	
Strongly disagree	14.0%	9.4%	

QOL = quality of life; RCT = randomized controlled trial.

Another debatable subject is the position of fetoscopic surgery. In a recent meta-analysis, outcomes of fetoscopic and open repair of MMC defects, published after MOMS, were analyzed and compared.²¹ A total of 11 studies were included, 4 retrospective and 7 prospective, comprising 436 patients. Fetoscopic MMC repair (percutaneous or laparotomy access combined) was associated with a lower rate of uterine dehiscence; however, this came with a higher rate of dehiscence or CSF leakage of the MMC defect, requiring postnatal revision. There were no between-group differences regarding fetal and postnatal mortality, rate of shunt placement, reversal of hindbrain herniation, motor response, gestational age before birth, chorioamniotic membrane separation, or placental abruption. Expectations of our respondents were similar with 89.2% expecting open fetal surgery to have the highest risk for maternal complications. Authors of another meta-analysis concluded similarly, suggesting that before performing a

randomized study, endoscopic techniques should be improved first.⁵

To date, no randomized controlled studies have compared the two techniques of fetoscopic and open postnatal repair. Only a minority of our respondents, 21.9% (strongly) believe it is acceptable to provide prenatal fetoscopic surgery without performing a randomized controlled trial. Fetoscopic surgery faces multiple barriers to implementation, as indicated by the reluctance of neurosurgeons to offer open fetal repair for MMC with only 13.0% of centers in our respondent pool offering the procedure.

Study Strengths and Limitations

Some limitations of this study must be acknowledged. First, the response rate could be a limitation. The survey was mailed to 7671 functional email addresses of CNS members. Three hundred seventy-four responses were from CNS members leading to an estimated response rate

of 4.9%. Of note, some of the email addresses may have belonged to medical students or other allied health professionals who may not have been eligible to fill out the survey. The importance of the response rate when judging the quality of cross-sectional research, however, is subject to debate.^{17,23} Given that 446 neurosurgeons with variable tenure and representing 58 countries responded to our survey, we expect a limited risk of nonresponse bias. However, we assume that MMC-dedicated centers, which also offer fetal surgery, may have been more inclined to respond.

Another limitation may be the unfamiliarity of the respondents with either the care of patients with MMC or prenatal surgery. However, 81.0% stated that they treat patients with MMC and 91.0% worked at a department where either postnatal or prenatal surgery is provided.

Conclusions

The survey results show the diversity in the management of patients with MMC. In addition, significant diversity remains regarding fetal surgery for MMC closure. Despite the apparent benefits of open fetal surgery in selected pregnancies, only a minority of centers and providers offer this technique. As a more technically demanding technique that requires multidisciplinary effort with less-well-established long-term outcomes, fetoscopic surgery may face similar limited implementation, although the surgery may pose fewer maternal risks than open fetal surgery. Centralization of prenatal treatment to tertiary care referral centers, as well as the use of sophisticated training models, may help to augment the most commonly cited objection to the implementation of prenatal closure, which is the overall limited caseload.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Gadjradj, Spoor, Eggink. Acquisition of data: Gadjradj, de Jong. Analysis and interpretation of data: Gadjradj, Spoor, DeKoninck. Drafting the article: Gadjradj. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Gadjradj. Statistical analysis: Gadjradj. Administrative/technical/material support: Gadjradj. Study supervision: Gadjradj, Wijnen, Miller, Rosner, Groves, Harhangi, Baschat, van Veelen, de Jong.

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