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Patient empowerment improves follow-up data collection after fetal surgery for spina bifida: institutional audit

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KEYWORDS: complications; data collection; fetal surgery; Maternal and Fetal Adverse Event Terminology; MFAET; patient empowerment; spina bifida

CONTRIBUTION

What are the novel findings of this work?
Within a fetal surgery program for spina bifida in which the referring unit provides the obstetric and neonatal care after discharge, spontaneous return of outcome data by referring centers was low, yet patient empowerment improved data collection. The Maternal and Fetal Adverse Event Terminology (MFAET) ranked surgical complications in a more clinically relevant way compared with the generic Clavien–Dindo classification.

What are the clinical implications of this work?
Involving patients allowed them to play an active role as coresearchers, which improved the acquisition of outcome data for this audit. The MFAET is a useful tool to report on complications after fetal surgery and should be considered in future reports.

ABSTRACT

Objectives *To define and grade fetal and maternal adverse events following fetal surgery for spina bifida and to report on the impact of engaging patients in collecting follow-up data.*

Methods *This prospective single-center audit included 100 consecutive patients undergoing fetal surgery for spina bifida between January 2012 and December 2021. In our setting, patients return to their referring unit for further pregnancy care and delivery. On discharge, referring hospitals were requested to return outcome data. For this audit, we prompted patients and referring hospitals to provide data in cases of missing outcomes. Outcomes were categorized as missing, returned spontaneously or returned following additional request, by the patient and/or referring center. Postoperative maternal and fetal complications until delivery were defined and graded according to Maternal and Fetal Adverse Event Terminology (MFAET) and the Clavien–Dindo classification.*

Results *There were no maternal deaths, but severe maternal complications occurred in seven women (anemia in pregnancy, postpartum hemorrhage, pulmonary edema, lung atelectasis, urinary tract obstruction and placental abruption). No cases of uterine rupture were reported. Perinatal death occurred in 3% of fetuses and other severe fetal complications in 15% (perioperative fetal bradycardia/cardiac dysfunction, fistula-related oligohydramnios, chorioamnionitis and preterm prelabor rupture of membranes (PPROM) before 32 weeks). PPROM occurred in 42% of patients and, overall, delivery took place at a median gestational age of 35.3 weeks (interquartile range, 34.0–36.6 weeks). Information provided following additional request, from both centers and patients but mainly from the latter, reduced missing data by 21% for gestational age at delivery, 56% for uterine-scar status at birth and 67% for shunt insertion at 12 months. Compared with the generic Clavien–Dindo classification, the*

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MFAET system ranked complications in a more clinically relevant way.

Conclusions *The nature and rate of severe complications following fetal surgery for spina bifida were similar to those reported in other large series. Spontaneous return of outcome data by referring centers was low, yet patient empowerment improved data collection.* © *2023 The Authors. Ultrasound in Obstetrics & Gynecology published by John Wiley & Sons Ltd on behalf of International Society of Ultrasound in Obstetrics and Gynecology.*

INTRODUCTION

Open spina bifida has a prevalence of approximately 4.9 and 3.2 per 10 000 births in Europe and the USA, respectively[1,2]. Patients suffer from variable degrees of lifelong neurodevelopmental delay, bowel and bladder dysfunction and orthopedic disability[3].

The Management of Myelomeningocele Study (MOMS) demonstrated that, in selected cases, repair of spina bifida in the fetus, rather than postnatally, reduces the need for a cerebrospinal fluid shunt at 12 months of age (40% *vs* 82%) and improves the composite score for mental development and motor function at 30 months[4,5], differences that persist into school age[6]. However, fetal surgery comes with substantial maternal and fetal risks. In the MOMS trial, the average gestational age at delivery following fetal surgery was 34.1 weeks, and 13% of women delivered before 30 weeks[4]. Uterine dehiscence, chorioamniotic membrane separation, placental abruption and preterm prelabor rupture of membranes (PPROM) are typical complications. Since the MOMS trial, several cohort studies have reported on complications of prenatal repair, some of which used the generic Clavien–Dindo classification for surgical interventions[7–12]. Recently, the Maternal and Fetal Adverse Event Terminology (MFAET)[13] has been proposed as a tool to aid classification of maternal and fetal adverse events, using a well-defined list of pregnancy-related complications. However, to our knowledge, this classification has not yet been applied clinically.

In 2012, the University Hospitals Leuven established a clinical fetal surgery program for spina bifida, following off- and on-site training[14]. In this program, subsequent obstetric and neonatal care are well defined (Appendix S1, Figure S1), but provided by the patient's referring unit after discharge rather than at the fetal surgery center. Such a decentralized fetal surgery program hampers the collection of follow-up data needed to audit a clinical service and ensure optimal care[14].

The primary objective of this study was to collect and assess data on maternal and fetal complications of patients undergoing fetal surgery for spina bifida in our unit, using the purpose-designed MFAET classification. Secondary objectives were to report on the rate of spontaneous return of follow-up data and the impact of engaging patients in collecting outcomes.

METHODS

This was a single-center study conducted at the University Hospitals Leuven, Leuven, Belgium, including the first 100 patients undergoing fetal surgery for spina bifida between January 2012 and December 2021. Auditing of the fetal surgery program for spina bifida was approved by the Ethics Committee for Clinical Studies (S63598) of UZ Leuven. All patients provided written informed consent for surgery as well as for data collection.

Our eligibility criteria for fetal surgery were largely in line with those of the MOMS trial, except for an increase in the upper limit of maternal body mass index from 35 to 40 kg/m^2 and the inclusion of women with well controlled diabetes or previous Cesarean section (Appendix S2)[15]. Fetal repair was done under general anesthesia, through laparotomy, uterine exposure and entry to the amniotic sac via an initial small hysterotomy with further expansion using a stapler after crushing of the myometrium[4]. Initially, a 6–8-cm stapled hysterotomy was made, which we were able to reduce to 3–4 cm over the years of the study. Through the hysterotomy, ideally, a three-layered repair (dura, myofascia, skin) was performed to provide watertight closure[16]. Eventually the uterus was closed in two watertight layers, the first running, the second interrupted and inverting. The first 10 patients received magnesium sulfate as tocolysis, the following 90 had atosiban. Details of postoperative management can be found in Appendix S1 and Figure S1. In December 2019, our center was assigned by the National Health Service England as a highly specialized commissioned service, and a modified leaflet with postoperative and postnatal instructions was added (Appendix S1).

Data collection

Available data were first retrieved from electronic clinical records. These were entered into a purpose-designed Research Electronic Data Capture (REDcap) database[17,18] and covered maternal demographics, pre-existing conditions, prenatal fetal findings, operative and postoperative complications and pregnancy and infant outcomes. In cases of missing data, we contacted patients and referring centers. For this audit, outcomes after discharge were again requested from patients and referring centers via e-mail through the REDcap platform. In case the e-mail failed to deliver or contact details were missing, patients and referring centers were contacted by telephone, and in the absence of a response, via letter, with the offer of telephone support.

Classification of outcomes

Complications were first graded according to the MFAET framework on a scale of Grade 1 to Grade 5, whereby Grade 1 represents mild complication, Grade 2

moderate complication, Grade 3 severe complication, Grade 4 life-threatening complication and Grade 5 fatal complication (Appendix S3)[13]. Maternal complications not included in the MFAET system were graded according to the Common Terminology Criteria for Adverse Events[19]. Complications were first assigned to categories by an experienced fetal surgeon (J.A.D.). Subsequently, they were recategorized by four physicians engaged in the fetal surgery program (S.V., J.A.D., E.V.d.E and S.M.-P.) and the lead authors of the MFAET publication (R.S. and A.D.) in a consensus meeting. Prior to that, all investigators were trained by grading 20 complications in five practice cases (Appendix S4). To enable comparison with earlier studies[12,20], complications were also categorized by two observers (S.V. and J.A.D.) according to the Clavien–Dindo classification (Table S1)[7]. Complications were reported as early postoperative (≤ 7 days) or late postoperative (> 7 days until delivery).

Information exchange

We compared the availability of data on three empirically chosen outcome measurements that we considered essential to assess the quality of care: (1) uterine-scar status at delivery; (2) gestational age at delivery; (3) shunt rate at 12 months. We recorded whether these data were returned spontaneously following the instructions issued at discharge or upon additional request, and whether this information was provided by the referring center and/or the patient.

Statistical analysis

Descriptive statistics were calculated using SPSS version 28 (IBM Software Inc., Armonk, NY, USA) and GraphPad Prism for Windows V.9.3.1 (GraphPad Software, San Diego, CA, USA) and are reported as *n* (%) and median (interquartile range (IQR)), as appropriate. The Pearson χ-square test was used to compare response rates. To assess the variability in reporting accuracy between patients and centers, we performed a Bland–Altman analysis of the continuous variable gestational age at birth; $P < 0.05$ was considered to indicate statistical significance.

RESULTS

Maternal and pregnancy characteristics and operative details are displayed in Table 1. All but three patients were from Europe; one in four patients were UK citizens referred as part of the NHS England Highly Specialised Commissioned Services.

Complications

We identified 266 maternal and 54 fetal complications. All complications were reviewed, and of the 24 classified initially as severe to life-threatening, 21% were recategorized (four downgraded, one upgraded). The recategorization rates for mild and moderate complications were 0% and 1%, respectively (two upgraded). Therefore, 2% of complications were recategorized at the review stage. Four types of complication (amnioperitoneal fistula, oligo- to anhydramnios, uterine dehiscence, PPROM Grade 1) lacked a precise MFAET definition. Hence, they were labeled as 'difficult to categorize' and may be revised in a future MFAET edition.

Taking only the highest graded complication for each woman into account, mothers had a variety of mild (15%), moderate (77%), severe (6%) and life-threatening (1%) complications; no lethal maternal complications were recorded. Along the same lines, in fetuses, mild complications were reported in 21% of cases, moderate in 10%, severe in 14%, life-threatening in 1% and lethal in 3%. There was no trend in the occurrence of complications over time (Table S2).

Early postoperative complications

Early maternal complications are detailed in Table 2. There were no maternal deaths (Grade 5). Five (5%)

Table 1 Demographic and operative characteristics and findings on presurgical ultrasound in 100 patients with spina bifida

Characteristic	*Value*
Maternal age (years)	31 (27–36)
BMI at first visit (kg/m^2)	25 (22–30)
Nulliparous	42 (42)
History of uterine surgery	
Cesarean section	13 (13)
Myomectomy	0 (0)
Country	
Belgium	10 (10)
Other European*	87 (87)
Non-European†	3 (3)
Findings in index fetus	
Ventriculomegaly (diameter ≥ 10.0 mm)	69 (69)
Type of lesion	
Myeloschisis	35 (35)
Myelomeningocele	65 (65)
Talipes	23 (23)
Anatomical level of lesion (ultrasound)	
T12 or higher	6 (6)
L1–L2	17 (17)
L3–L4	42 (42)
L5–S1	35 (35)
Lower limb movement‡	68/82 (83)
T12 or higher	3/6 (50)
L1–L2	15/15 (100)
L3–L4	27/32 (84)
L5–S1	23/29 (79)
Operative characteristics	
Gestational age at surgery (weeks)	25.3 (24.8–25.7)
Neurulation placode	41 (41)
Skin repair technique	
Primary closure	77 (77)
Skin substitute	23 (23)
Skin-to-skin time (min)§	200 (175–225)
Neurosurgery time (min)¶	74 (60–90)
Length of hospital stay (days)	6 (6–7)

Data are given as median (interquartile range), *n* (%) or *n*/*N* (%). *Excluding Belgium and including UK. †Patients were from USA ($n = 1$) or Australia ($n = 2$). ‡Preserved lower limb movement was defined as movements in hip, knee and ankle joints of both lower limbs. §Data available for 73 patients. ¶Data available for 79 patients. BMI, body mass index.

Table 2 Consensus categorization of adverse events from surgery until delivery, according to Maternal and Fetal Adverse Event Terminology (MFAET) classification[13]

	Early complications (postoperative day 0–7) (n = *100*)		*Late complications (postoperative day 8 to delivery)* (n = *95*)	
MFAET grade	*Maternal*	*Fetal*	*Maternal*	*Fetal*
Grade 5: death	—	Fetal bradycardia: non-labor ($n=1$ (1%))	—	Perinatal death ($n=2$ (2%))
Grade 4: life-threatening consequences needing urgent intervention	—	—	Placental abruption ($n=1$ (1%))	Chorioamnionitis ($n=1$ (1%))
Grade 3: severe, but not immediately life-threatening	Anemia in pregnancy (transfusion needed) ($n=2$ (2%)) Pulmonary edema ($n=1$ (1%)) Lung atelectasis ($n=1$ (1%)) Urinary tract obstruction ($n=1$ (1%))	Oligo- to anhydramnios due to fistula ($n=2$ (2%)) Fetal bradycardia: non-labor ($n=1$ (1%)) Fetal cardiac function abnormality ($n=1$ (1%))	Postpartum hemorrhage ($n=2$ (2%))	PPROM§ ($n=9$ (9%)) Oligo- to anhydramnios due to fistula ($n=1$ (1%))
Grade 2: moderate or needing local or non-invasive intervention	Oliguria ($n=24$ (24%)) Anemia in pregnancy (iron supplementation needed) ($n=14$ (14%)) Hypotension ($n=12$ (12%)) Infection* ($n=7$ (7%)) Premature labor ($n=6$ (6%)) Dysesthesia ($n=2$ (2%)) Bladder perforation ($n=1$ (1%)) Wound complication† ($n=1$ (1%)) Amnioperitoneal fistula ($n=1$ (1%) Cerebrospinal fluid leakage ($n=1$ (1%))	Fetal bradycardia: non-labor ($n=1$ (1%))	PPROM§ ($n=40$ (42%)) Premature labor ($n=28$ (29%)) Uterine dehiscence¶ ($n=11$ (12%)) Postpartum hemorrhage ($n=5$ (5%)) Wound complication** ($n=4$ (4%)) Amnioperitoneal fistula ($n=1$ (1%)) Chorioamnionitis ($n=1$ (1%)) Hemorrhage in pregnancy ($n=1$ (1%))	PPROM§ ($n=10$ (11%))
Grade 1: no or mild symptoms not leading to intervention	Anemia in pregnancy ($n=64$ (64%)) Maculopapular rash ($n=3$ (3%)) Wound complication‡ ($n=2$ (2%)) Hemorrhage in pregnancy ($n=1$ (1%)) Amnioperitoneal fistula ($n=1$ (1%)) Lethargy ($n=1$ (1%)) Hallucinations ($n=1$ (1%))	PPROM§ ($n=4$ (4%))	Postpartum hemorrhage ($n=10$ (11%)) Uterine dehiscence¶ ($n=8$ (8%)) Wound complication†† ($n=5$ (5%)) Chorioamnionitis ($n=1$ (1%)) Hemorrhage in pregnancy ($n=1$ (1%))	PPROM§ ($n=21$ (22%))

Patients could report multiple complications. *Infection other than wound infection, requiring antibiotic treatment: urinary tract infection ($n=2$), vaginal infection ($n=1$), *e causa ignota* ($n=4$). †Wound infection ($n=1$). ‡Seroma ($n=1$), hematoma ($n=1$). §Preterm prelabor rupture of membranes (PPROM) was categorized as maternal complication (Grade 2) as well as fetal complication, which was further subdivided (Grade 1, chorioamniotic membrane separation without confirmed rupture of membranes; Grade 2, PPROM between 32+0 and 33+6 weeks; Grade 3, PPROM between 22+0 and 32+0 weeks; Grade 4, PPROM before 22 weeks). ¶Definition of uterine dehiscence was agreed upon during consensus meeting as myometrial defect covered by peritoneum (Grade 1, asymptomatic scar thinning without repair at delivery; Grade 2, asymptomatic partial scar dehiscence or scar thinning which was repaired at delivery; Grade 3, scar dehiscence associated with symptoms, yet not perceived as life-threatening; Grade 4, life-threatening symptomatic scar dehiscence). **Wound infection ($n=4$). ††Seroma ($n=4$), dehiscence ($n=1$).

patients had severe complications, including two women who received postoperative blood transfusions owing to a low postoperative hemoglobin level but without an obvious active bleeding site. Both patients already had a low hemoglobin level (< 10 g/dL) preoperatively. One patient developed pulmonary edema, which resolved after a single course of furosemide. One dyspneic patient was diagnosed with lung atelectasis on X-ray, which improved after oxygen supplementation and physiotherapy. One patient had flank pain on day 4. On ultrasound scan, she had unilateral hydronephrosis without demonstrable urolithiasis. A similar pain episode had occurred before the index pregnancy. She received a double-J stent on postoperative day 6, which was removed 3 months later.

Eighty-three patients (83%) had mild or moderate early maternal complications. Of note, in two patients there was severe oligohydramnios on postoperative day 2 and 7, respectively, and an amnioperitoneal fistula was confirmed on magnetic resonance imaging (MRI). The first patient, who was one of our earlier cases, was managed conservatively as an inpatient for 55 days (Grade 2). The second patient was managed as an outpatient (Grade 1). One patient developed acute lower abdominal pain on day 4 and, because of an initial epoch of hematuria, we suspected and eventually confirmed on MRI urinary leakage at the bladder dome. An indwelling catheter was inserted for 10 days, which resolved the symptoms (Grade 2). Two of our first 10 patients, who received magnesium sulfate rather than atosiban for tocolysis, experienced lethargy or hallucinations.

Five (5%) Grade ≥ 3 early fetal complications occurred. One fetus developed bradycardia and decreased cardiac contractility during surgery, requiring resuscitation. Although the fetus recovered, intrauterine demise was diagnosed the following day (Grade 5). Two additional fetuses required intraoperative resuscitation: one for bradycardia (heart rate < 100 bpm) without hypocontractility, and one for hypocontractility without bradycardia. Both recovered after a single dose of atropine. For the two mothers with amnioperitoneal fistulas, the associated oligohydramnios was categorized as a Grade-3 fetal complication. Five (5%) fetuses had mild-to-moderate complications, including one with bradycardia < 100 bpm, which resolved by maternal repositioning.

Late postoperative complications

We obtained postdischarge outcome details for 95 patients. Delivery occurred at a median gestational age of 35.3 weeks (IQR, 34.0–36.6 weeks). Twenty-one (22%) women delivered prior to 34 weeks' gestation, including six (6%) who delivered prior to 30 weeks. Late Grade ≥ 3 complications were present in three (3%) women, including one case of placental abruption at 35 weeks (Grade 4) and two cases in which blood transfusion was required for postpartum hemorrhage (Grade 3). Mild or moderate complications developed in 73 (77%) patients. The majority were instances of PPROM before 37 weeks, occurring in 40 (42%) women at a median gestational age of 34.1 weeks (IQR, 32.3–35.5 weeks). The median interval from PPROM to delivery was 0 days (IQR, 0–3 days). Of the 17 women who had PPROM before 34 weeks, two delivered before 30 weeks, 13 between 30 and 34 weeks and two after 34 weeks. There were no instances of uterine rupture, yet nine (9%) patients were diagnosed with scar dehiscence without symptoms. At Cesarean section, 11 (12%) women underwent uterine scar repair, including two of the 10 patients who were described as having a thin scar. One patient presented with anhydramnios at 31 weeks, went into preterm labor at 33 weeks and an amnioperitoneal fistula was diagnosed at the time of birth. One patient presented with contractions at 26 weeks, with elevated C-reactive protein, and amniocentesis demonstrated chorioamnionitis, prompting preterm delivery (maternal Grade 2; fetal Grade 4). Both mother and baby recovered following administration of antibiotics, and at the time of writing the child was a healthy 5-year-old. Chorioamnionitis was demonstrated on pathology in one asymptomatic patient who delivered at 29 weeks (Grade 1).

Regarding fetal complications, we observed two perinatal deaths; one was an intrauterine demise at 29 weeks with an umbilical cord knot shown on postmortem examination, the other was a neonatal death from complications of prematurity following delivery at 29 weeks. Other fetal complications are displayed in Table 2.

Clavien–Dindo classification

Complications were also categorized using the Clavien–Dindo classification system (Table S1). We categorized three maternal complications as severe, of which one was early (urinary-tract obstruction) and two were late (placental abruption and chorioamnionitis). Five fetal complications were classified as severe (two cases of fetal bradycardia, two of perinatal death and one of fetal cardiac dysfunction).

Shunt rate at 12 months

At the close of the audit, 69 children had reached the age of 12 months. Of those, 32 (46%) had a ventriculoperitoneal shunt or endoscopic ventriculostomy ($n = 29$ and $n = 3$, respectively). Fetuses with no, mild to moderate (10–15 mm) or severe (> 15 mm) ventriculomegaly on preoperative ultrasound were shunted eventually in 32% (6/19), 47% (17/36) and 64% (9/14) of cases, respectively.

Information exchange

Two (2%) patients declined to provide further follow-up information. Seventy (70%) patients returned follow-up data upon additional request, as did referring centers in 46 (46%) cases. The response rate among patients operated on before or after the start of the NHS-commissioned service was comparable (36/56 *vs* 34/44; $P = 0.15$), but the response rate among referring centers increased (18/56

vs 28/44; $P = 0.002$). Additional feedback provided following additional request, from both centers and patients but mainly from the latter, reduced the rate of missing information on gestational age at delivery from 26% to 5%, uterine-scar status from 61% to 5% and shunt rate at 12 months from 89% to 22% (Table 3, Figure 1).

In 32 patients, both the local center and the patient reported on outcomes at delivery. Data provided by the center and patient were identical in 94% (30/32) of these cases for gestational age and scar status. In two patients, there was a difference in gestational age at delivery of 1 day. According to the Bland–Altman method, the limits of agreement within a 95% CI was 1 day. In two other patients, a discrepancy arose in scar status. Of the 31 replies on the shunt rate at 12 months, all were concordant.

DISCUSSION

Main findings

In 100 consecutive patients undergoing fetal surgery for spina bifida, severe complications (MFAET Grade 3–5) occurred in seven mothers and in 18 fetuses. Using the Clavien–Dindo classification, only three (3%) maternal complications would be considered severe. Reporting of spontaneous outcome was poor, particularly for outcomes to be reported at 1 year (11%). Direct engagement with patients and repeated requests to the referring centers resulted in the acquisition of additional information. However, despite all our efforts, we still lacked information on essential outcomes at the time of writing, including uterine-scar status in 5% and shunt rate in 22%.

Interpretation

One goal of this study was to compare the MFAET and Clavien–Dindo systems of classification, which differ in some respects. MFAET adds a specific dimension of pregnancy-related complications and discriminates between fetal and maternal complications. For instance, MFAET assesses the maternal and fetal impact of complications separately, such as in the case of chorioamnionitis and PPROM. Also, in our dataset, MFAET ranked certain adverse events, including pulmonary edema, lung atelectasis, urinary tract obstruction and postpartum hemorrhage, as more severe compared with the Clavien–Dindo system, reflecting their clinical impact. MFAET was found to have low interobserver variability for moderate and mild complications (only 1% reclassification), but higher variability for severe complications (20% reclassification). Therefore, some MFAET definitions may need further refinement. Finally, some of the typical complications of fetal spina bifida surgery were not captured by the MFAET system, such as uterine-scar status, the occurrence of an amnioperitoneal fistula and the presence of oligo- or anhydramnios.

For benchmarking our outcomes, we compared the rate and nature of complications, including PPROM and gestational age at delivery, with that in other series[4,8,10,21]. As in previous trials, there were no symptomatic uterine ruptures in the index pregnancy[4,8,10]. Unfortunately, there

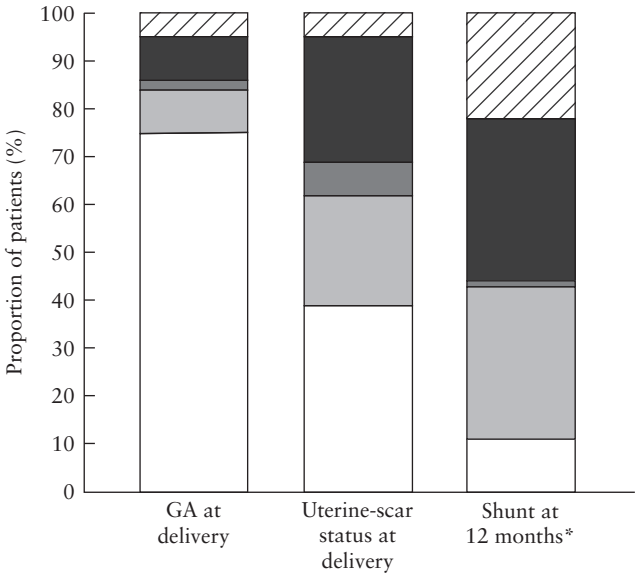

Figure 1 Source of follow-up data regarding post-discharge outcomes in 98 patients who underwent spina bifida repair. *Data for 88 patients, owing to three perinatal deaths and seven cases aged < 12 months at time of writing. □, data reported spontaneously; ▫ data provided only by patient; ■, data provided only by referring center; ■, data provided by both patient and referring center; ▨, persistent missing data. GA, gestational age.

Table 3 Missing data before and after information request

Reported outcome	*Missing data prior to invite*	*Response by patients following invite*	*Response by centers following invite*	*Persistent missing data*
GA at delivery ($n = 98$)	25/98 (26)	18/25 (72)	11/25 (44)	5/98 (5)
2012–2019 ($n = 55$)	11/55 (20)	6/11 (55)	1/11 (9)	4/55 (7)
2020–2021 ($n = 43$)	14/43 (33)	12/14 (86)	10/14 (71)	1/43 (2)
Uterine-scar status at delivery ($n = 98$)	60/98 (61)	48/60 (80)	32/60 (53)	5/98 (5)
2012–2019 ($n = 55$)	33/55 (60)	24/33 (73)	11/33 (33)	4/55 (7)
2020–2021 ($n = 43$)	27/43 (63)	24/27 (89)	21/27 (78)	1/43 (2)
Shunt at 12 months ($n = 88$)*	78/88 (89)	58/78 (74)	31/78 (40)	19/88 (22)
2012–2019 ($n = 53$)	45/53 (85)	29/45 (64)	12/45 (27)	15/53 (28)
2020–2021 ($n = 35$)	33/35 (94)	29/33 (88)	19/33 (58)	4/35 (11)

Data are given as *n*/*N* (%). *Data missing for 10 patients owing to three perinatal deaths and seven cases aged < 12 months at time of writing. GA, gestational age.

is no precise definition of uterine dehiscence and thinning, which may need to be addressed in future editions of the MFAET classification. Herein, we refer to dehiscence as a myometrial defect that was covered by the peritoneum. We also reported on 92 (92%) mothers experiencing mild-to-moderate complications (MFAET Grade 1–2). At first glance, this is dramatically higher than the 16% documented in our systematic review[12]. For that analysis, however, the Clavien–Dindo classification was used, which, again, ranks complications as less severe, and most included studies were retrospective chart reviews. Rather, the rate we observed is akin to that reported by Vonzun *et al.*[21] in a similar prospective single-center audit in Zurich, Switzerland. However, the nature of the complications differed. In the Swiss audit, seroma formation and chorioamniotic membrane separation were more frequent, whereas we observed a greater number of patients with anemia, oliguria and hypotension. The clinical impact of these complications and the differences between both cohorts are limited.

We observed nine (9%) patients with early fetal complications. The nature and rate of complications, as well as the shunt rate at 12 months of age (46%), are comparable with those in previous reports[4,8–10].

According to the American College of Obstetricians and Gynecologists, fetal surgery units must collect and benchmark their outcomes[14]. In a decentralized program, we observed a strikingly low rate of return of outcomes despite precise instructions on discharge. Delivery data were missing for 26% for gestational age at delivery and 61% for uterine-scar status, and the rate of missing infant outcome data was as high as 89% for shunt placement at 12 months. Additional requests to patients and centers significantly reduced the amount of missing data. Although good reference data are lacking, the problem of missing outcomes for patients not delivering at the fetal surgery center is not new[4,8,9]. The second observation was that, logically, the amount of missing data increased with increasing time after fetal surgery, up to 22% for the 1-year shunt rate. Finally, patients were more responsive to our additional requests than were referring centers (approximately 75% *vs* 45%) and patients' answers aligned remarkably well with those of the centers for the three selected key parameters (94–100%). Although discrepancies may occur more frequently for other, more sophisticated neurologic outcomes, our experience shows that patient involvement increases data acquisition in a decentralized setting. To improve data quality and accuracy, one could combine efforts and ask patients to urge caregivers at the follow-up unit to fill out standardized reports. Involving patients in research moves them from a passive to an active role and expands their role as coresearchers, thereby promoting patient empowerment[22,23].

Strengths and limitations

This study reports 'real-world' data on a reasonably sized consecutive case series of patients managed in a standardized way. We maximized data completeness through contact with patients and referring centers. Our study is one of the few to quantify the impact of patient engagement on data collection and assess concordance with hospital-provided data, which we found to be high. Finally, we first used the MFAET classification to categorize complications of a complex fetomaternal intervention. Our study was limited by its sample size, which precluded reliable estimation of the rate of rare complications. Furthermore, we do not have complete outcomes for all cases, including the two women who declined to participate in the follow-up study. This means that benchmarking outcomes remains difficult and complications may have been underestimated. Although there was good concordance between patient- and hospital-provided data, the delay in providing data and the self-reported nature of patient data may limit their accuracy and quality.

Conclusions

In a cohort of 100 consecutive patients undergoing fetal surgery for spina bifida, the rates and types of severe maternal and fetal complications observed were similar to those reported in other large series. The MFAET framework was first applied and helped in the clinically relevant classification of maternal and fetal complications. While the rate of spontaneous return of outcome data by referring centers was low, patient empowerment improved data collection.

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SUPPORTING INFORMATION ON THE INTERNET

The following supporting information may be found in the online version of this article:

Appendix S1 Instruction leaflets handed to patients at discharge

Appendix S2 Inclusion and exclusion criteria to assess eligibility for prenatal spina bifida surgery

Appendix S3 Maternal and Fetal Adverse Event Terminology (MFAET) classification system

Appendix S4 Practice cases for Maternal and Fetal Adverse Event Terminology (MFAET) classification validation meeting

Figure S1 Details of postnatal management pathway.

Table S1 Maternal and fetal adverse events from surgery until delivery categorized using Clavien–Dindo classification system

Table S2 Frequency of maternal and fetal complications in patients recruited at different times over study period, according to Maternal and Fetal Adverse Event Terminology classification system