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A LARGE AMNIOCELE WITH PROTRUDED RIGHT ARM AND HEMITHORAX: A CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT

Uterine rupture is a rare but serious peripartum complication associated with high rates of maternal and fetal death. Spontaneous silent rupture of the intact uterus is exceptional. Amniocele which is defined by the herniation of the amnion through a uterine defect is another extremely rare condition. We hereby report an uncommon case of an asymptomatic 38-year-old woman (gravida 3 para 1 with 1 live child) who presented with a large amniocele detected at 22th weeks of gestation by routine ultrasonography. After informing the patient and her husband about the potential risks of complete uterine rupture and eventually hysterectomy, it was decided by mutual agreement to continue the pregnancy in our facility. The herniated amniotic sac continued to increase as the pregnancy advanced. Consequently, a scheduled cesarean section was performed at the 29th week of gestation which allowed the cephalic extraction through the uterine defect of a male newborn, Apgar score at 1 and 5 minutes were 10 and 10 respectively, birth weight 1500g. Patient's recovery course was uncomplicated. Patient and newborn were discharged home on postoperative day-4. A review of the literature was then made and all cases of amniocele before 34 weeks of gestation were studied. The management of amniocele depends on several factors including the desire to continue the pregnancy of the family, term at the time of diagnosis, fetal viability, severity of symptoms, thickness and size of uterine defect. Therapeutic strategies can be classified into three categories: termination of pregnancy, surgical repair of uterine defect, and expectancy with strict supervision. Amniocele is an uncommon pathology whose management is complicated. Practitioners need to adopt the best course of action depending on the situation: surgical repair when possible remains the method that has shown the best results, but the expectant attitude with strict supervision can also be beneficial and preserve the pregnancy.

KEYWORDS: Amniocele, Hemithorax.

BACKGROUND

Practitioners are well aware of the risk of uterine rupture associated with labor after cesarean section or myomectomy especially in a multiparous patient. It is a rare but serious peripartum complication associated with high rates of maternal and fetal death. Spontaneous silent rupture of the intact uterus is exceptional. Amniocele which is defined by the herniation of the amnion through a uterine defect is another extremely rare condition. The following case of a silent rupture of an unscarred uterus associated with a large amniocele of incidental discovery at 22th weeks of gestation presented an interesting clinical problem but also therapeutic issues.

CASE REPORT

We hereby report an uncommon case of an asymptomatic 38-year-old woman (gravida 3 para 1 with 1 live child) who presented with a large amniocele detected at 22th weeks of gestation by routine ultrasonography. The patient had history of an uneventful vaginal delivery four years prior to the index pregnancy and a spontaneous miscarriage at 8 weeks of amenorrhea two years later. She had no history of intrauterine procedure such as curettage or hysteroscopy. The ultrasound performed at the 21st week of gestation showed an active monofetalous pregnancy in transverse position, fetal biometry concurring to the age of pregnancy with an anterior low-lying placenta, and an oligohydramnios. This ultrasound revealed a significant herniation of the amniotic sac through a defect of the

right uterine wall of 3 cm, extending into the right flank, coming into contact with the gallbladder with a height of 12.60 cm. (Figure 1) This amniocele was containing the right fetal arm. (Figure 2) MRI was then performed at 22 weeks of gestation and revealed a 3 cm rupture of the right uterine wall and a large amniocele that measured 16.9 cm by 16.1 cm by 10 cm and contained the right fetal arm. (Figure 3).

After informing the patient and her husband about the potential risks of complete uterine rupture and eventually hysterectomy, it was decided by mutual agreement to continue the pregnancy in our facility. Consequently, the patient was hospitalized with close monitoring of uterine contractions, bleeding, bi-weekly ultrasound monitoring and exclusive bed rest. The herniated amniotic sac continued to increase as the pregnancy advanced. At the 28th week of gestation the amniocele measured 19 cm by 16 cm by 11 cm and was containing the right fetal arm and its right hemithorax through a defect of the right uterine wall of 6 cm. (Figure 4) Estimated fetal weight was 1.6 kg at that time.

A scheduled cesarean section was then performed at the 29th week of gestation which allowed the cephalic extraction through the uterine defect of a male newborn, Apgar score at 1 and 5 minutes were 10 and 10 respectively, birth weight 1500g. (Figure 5) Directed delivery of the placenta was easy and the patient received only 5 IU of oxytocin. Inspection of the uterus didn't reveal any scar nor adhesion. The corporal hysterorraphy was performed in a single layer. (Figure 6) Being well aware of the risks of recurrence and future complications that may arise from another pregnancy, the patient requested a tubal section ligation that was performed. Remainder of the surgery was completed in the usual fashion. The blood loss was estimated at 500cc and the operative time was 55 minutes. The postoperative hemoglobin was 11.5 g/dL. Patient's recovery course was uncomplicated. She had normal amount of vaginal bleeding postpartum. Patient and newborn were discharged home on postoperative day-4.

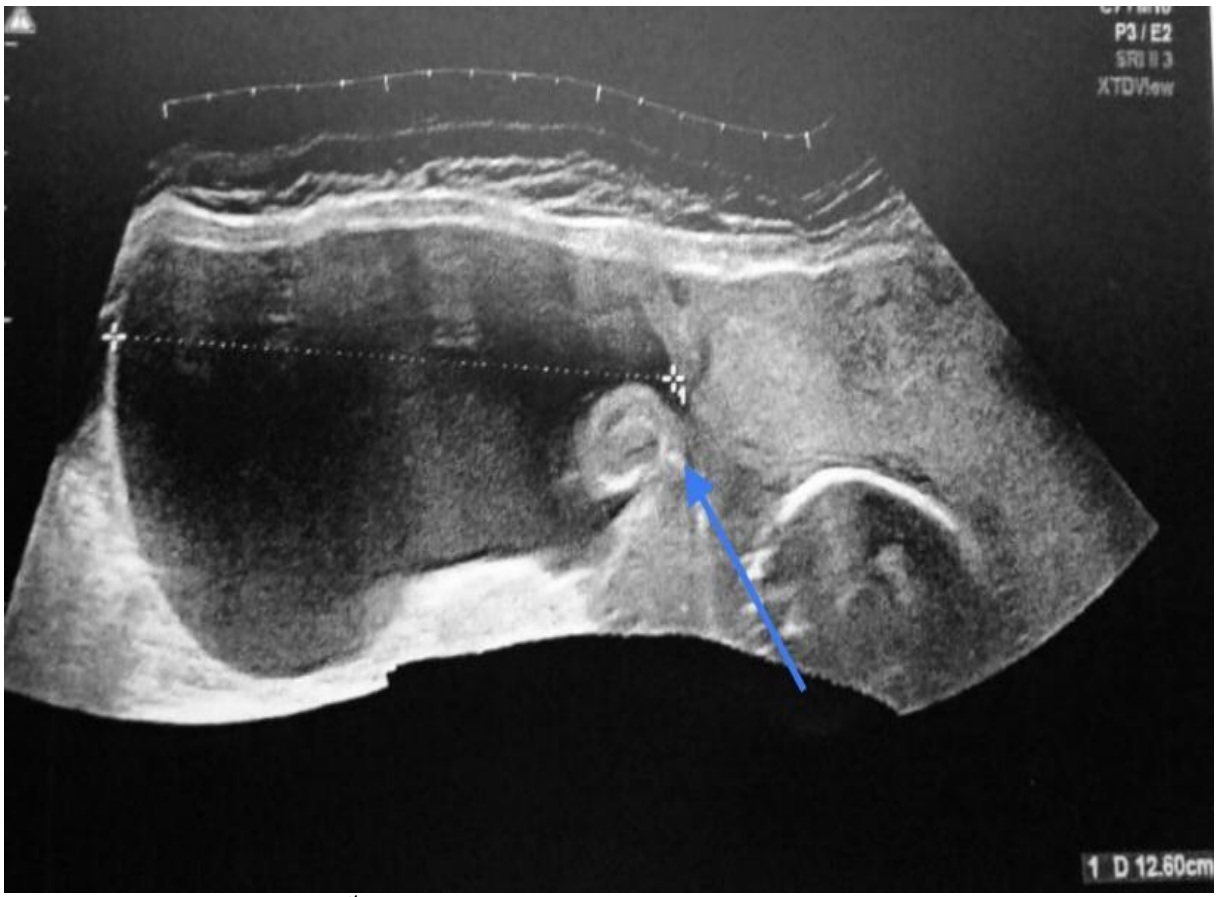

Figure 1: Ultrasound image at 21st weeks of gestation showing a significant herniation of the amniotic sac through a defect of the right uterine wall with a height of 12.60 cm.

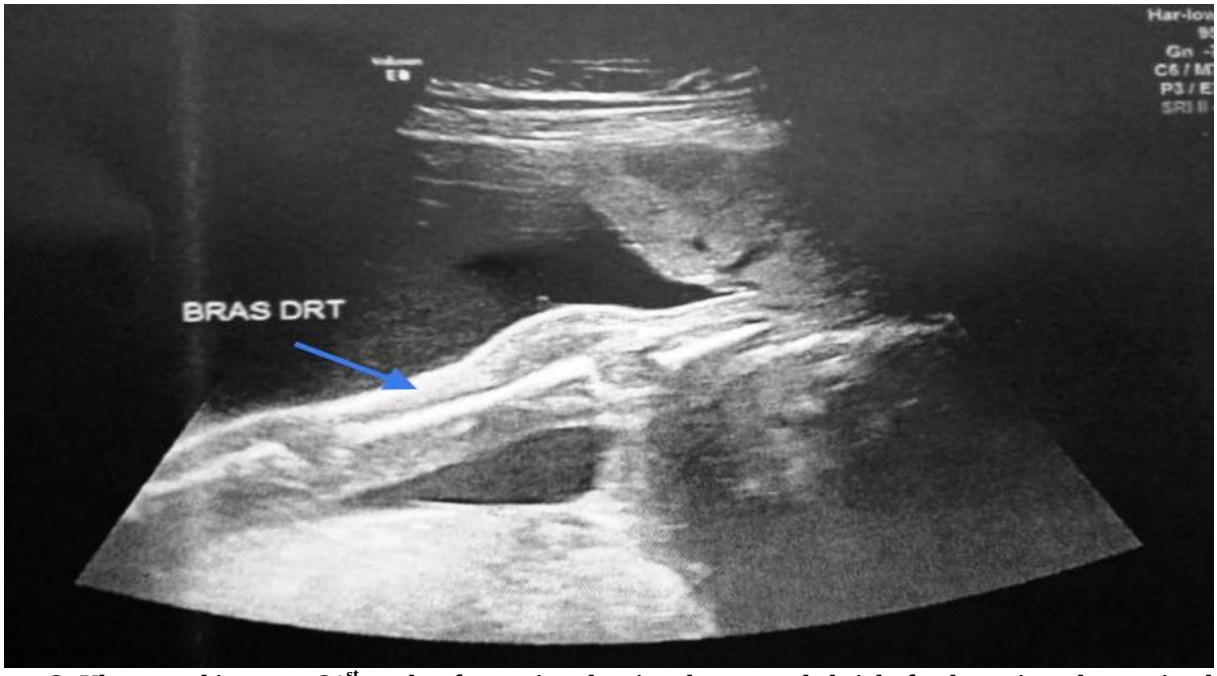

Figure 2: Ultrasound image at 21st weeks of gestation showing the protruded right fetal arm into the amniocele.

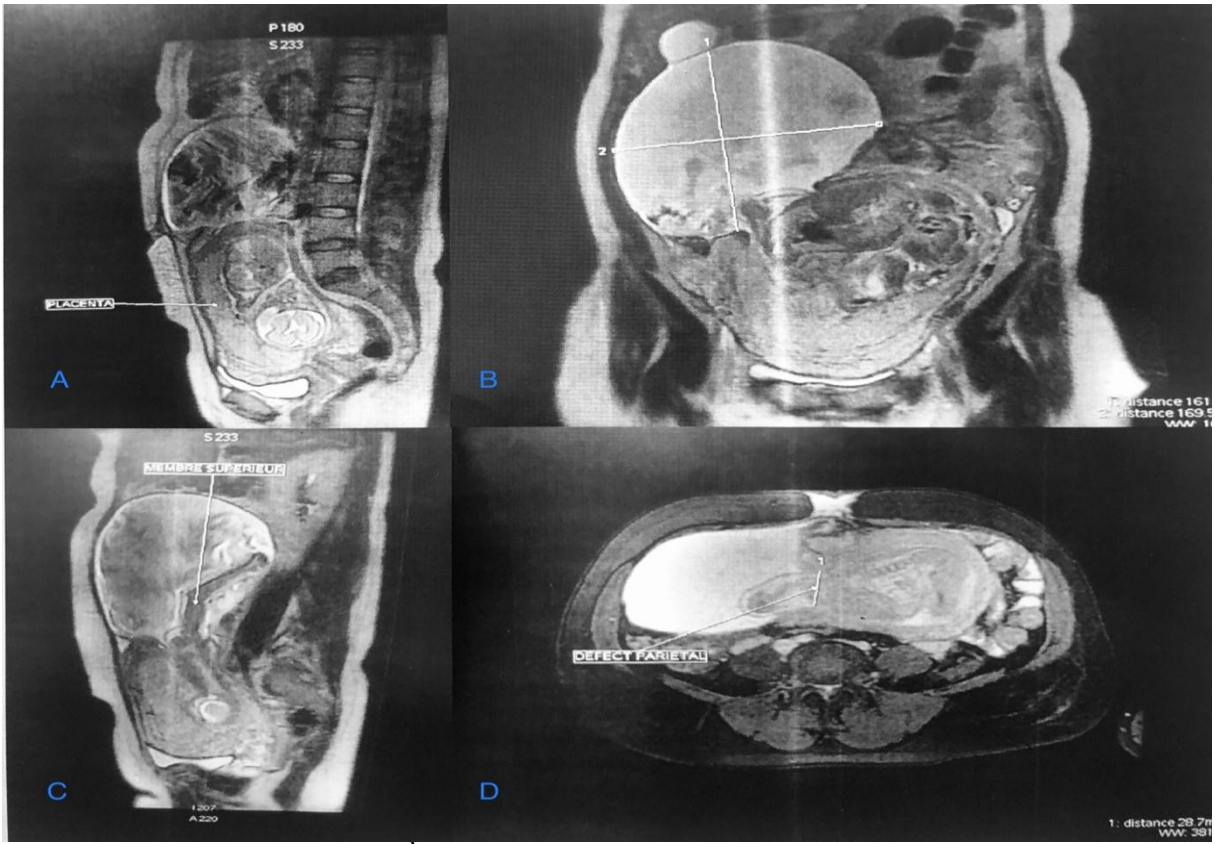

Figure 3: Abdominal-pelvic MRI at 22th weeks of gestation showing
A: Anterior Low-lying placenta
B: Amniocele of 161mm by 169mm extending into the right flank
C: Protruded fetal right arm into the amniocele
D: Right wall uterine defect of 29.7mm

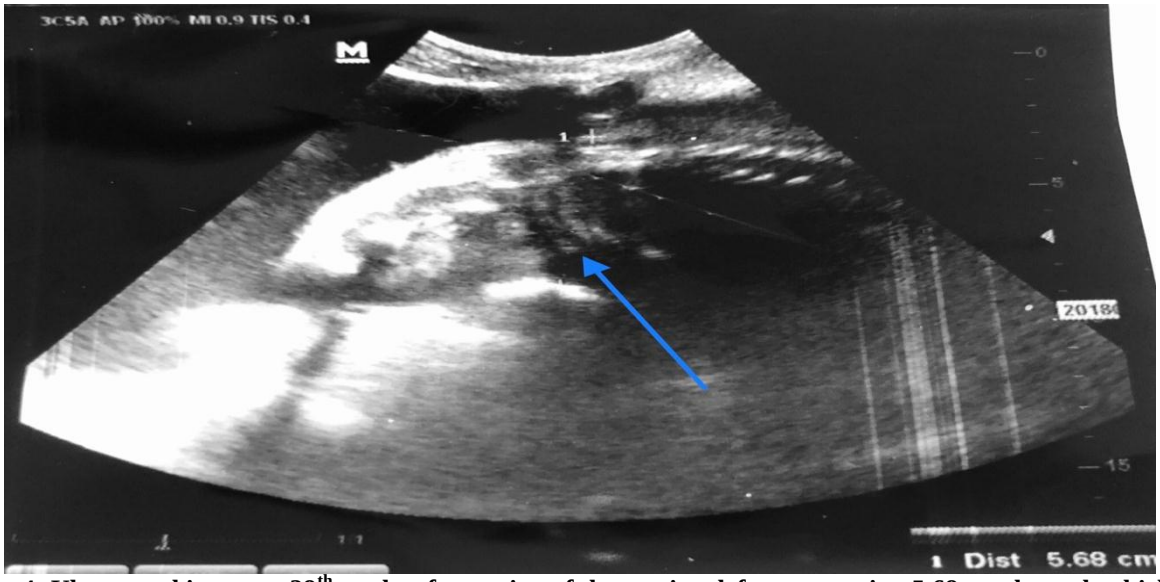

Figure 4: Ultrasound image at 28th weeks of gestation of the uterine defect measuring 5.68 cm through which protrudes the right arm and hemithorax of the fetus.

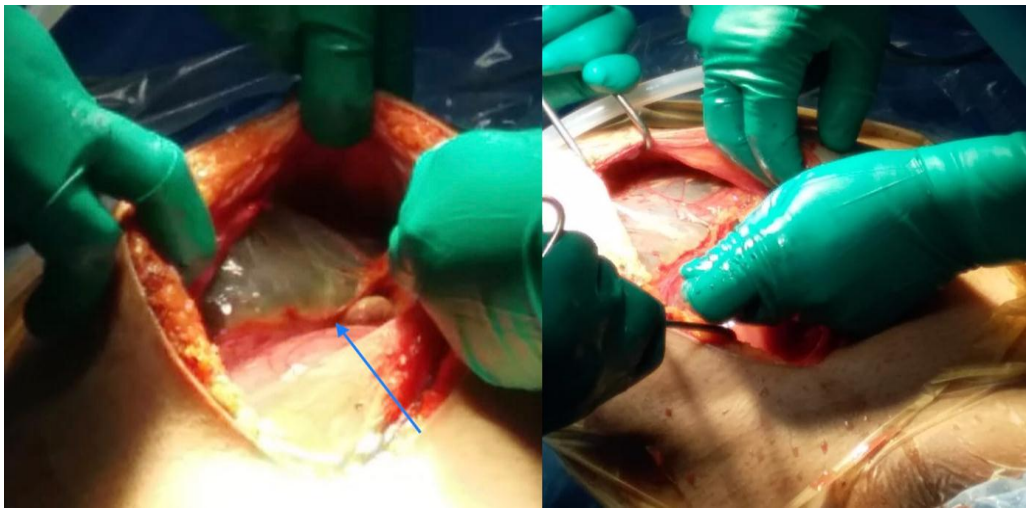

Figure 5: Uterine rupture with extruded amniotic sac.

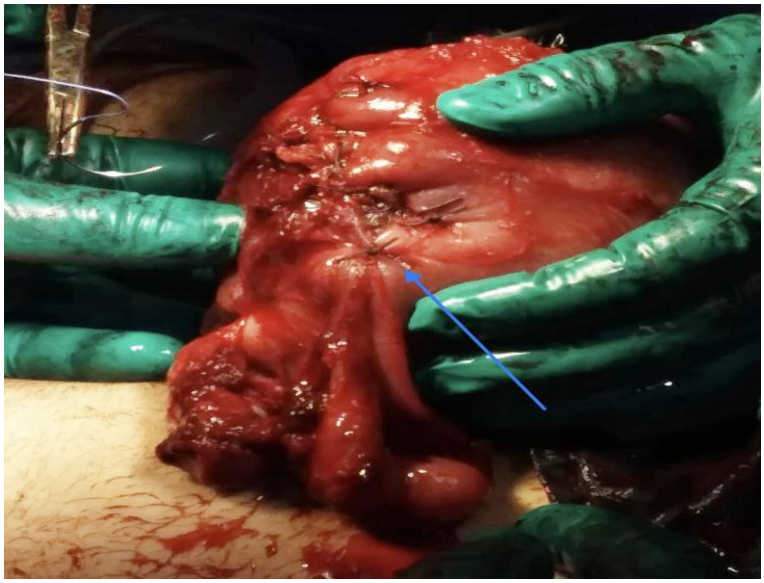

Figure 6: Hysterorrhaphy of the uterine defect.

Table 1: Literature review of amniocele before 34 weeks of gestation.

Case Nº	Author (Reference)	Year	Age (years)	Gravity Parity	Onset of symptoms (weeks)	Symptom at diagnosis	Underlying cause	Location of uterine defect	Management	Latency period (weeks)	Outcome
1	Kushnir O[7]	1990	29	G5 P1	28	Suprapubic pelvic pain	High transverse CS	Anterior wall	Termination	0	CS due to variable fetal heart decelerations (28w)
2	Avrech OM[8]	1994	26	G3 P1	31	Acute upper abdominal pain	Classical CS	Lower uterine segment	Termination	0	CS (31w)
3	Hamrick-Turner JE[9]	1995	27	G5 P3	13	Lower abdominal cramping and vaginal bleeding	Three previous CS	Lower uterine segment	Termination	2	Hysterectomy (15w)
4	Van Alphen M[10]	1995	34	G7 P3	28	Vague abdominal pain	Previous cornual uterine rupture following salpingectomy for ectopic pregnancy	Fundus	Termination	0	CS (28w) and subsequent supravaginal hysterectomy
5	Markos F[11]	1995	30	G6 P3	33	Decreased fetal mouvement, and intermittent abdominal pain	Uterine perforation following curettage	Fundus	Termination	0	CS (33w)
6	Catanzarite VA[12]	1996	N/A	G5 P3	33	Abdominal cramps and upper abdominal pain	Previous traumatic transverse fundal uterine rupture, twin gestation	Fundus	Termination	0	CS (33w)
7	Hasbargen U[13]	2002	30	G1 P0	29	Lower abdominal pain	Laparoscopic myomectomy	Posterior wall	Termination	0	CS due to increasing abdominal pain and a low-grade temperature of 38ºC (29w)
8	Youngs DJ[14]	2004	late 20s	G3 P1	26	Acute abdominal pain	Previous CS	Lower uterine segment	Termination	0	CS due to worsening pain and decelerations in fetal heart rate (26w)
9	Chou MM[15]	2007	29	G2 P0	21	None	Previous laparoscopic myomectomy, and twin pregnancy	Fundus	Termination	0	Termination (21w)
10	Zuckerwise LC[16]	2011	27	G5 P2	19	None	Two previous CS	Lower uterine segment	Termination	2	Hysterectomy (21w)
11	Jo YS[17]	2012	30	G3 P2	23	Abdominal discomfort	Amniocentesis (22w)	Fundus	Termination	0	CS due to vaginal bleeding and progressive lower abdominal pain (23w)
12	Mishina M[18]	2014	36	G1 P0	32	Severe abdominal pain and reduced fetal movement	Amniocentesis (16w)	Fundus	Termination	0	CS (32w)
13	Kwon J[19]	2016	30	G1 P0	24	Acute abdominal pain and amniotic fluid leaquage	Laparoscopic myomectomy	Fundus	Termination	0	CS (24w)
14	Cheng PJ[20]	2003	32	G2 P1	6	Uterine cramping	Uterine perforation following curettage	Fundus	Surgical repair (9w)	27	CS due to preterm labour (33w)
15	Chen FP[21]	2007	29	G1 P0	26	Abdominal pain, nausea and vomiting	None	Fundus	Surgical repair (26w)	11	CS (37w)
16	Gorthi S[22]	2009	31	G3 P1	22	None	Previous CS	Lower uterine segment	Surgical repair (26w)	6	CS due to spontaneous labour (28w)
17	Fujii T[23]	2000	30	G3 P1	19	None	Uterine perforation following curettage	Fundus	Surgical repair (20w)	17	CS (36w)
18	Liao C-Y[24]	2009	20	G7 P0	13	Sudden abdominal pain and vaginal bleeding	Previous laparoscopic cornual resection for an intestinal pregnancy, and twin pregnancy	Fundus	Surgical repair (13w)	17	CS due to preterm labour (30w)
19	Hamar BD[25]	2003	32	G4 P2	20	Vaginal bleeding and uterine cramps	Previous CS	Lower uterine segment	Expectant	11	CS due to variable decelerations in fetal heart rate (31w)
20	Cotton DB[5]	1982	34	G4 P3	20	Abdominal pain	None	Fundus	Expectant	9	CS due to severe variable decelerations in fetal heart rate (29w)
21	Oyelese Y[26]	2003	39	G4 P3	17	Sudden lower abdominal pain	Uterine perforation following curettage	Fundus	Expectant	16	CS due to fetal tachycardia with recurrent deep variable decelerations (33w)
22	Taipale P[27]	2005	30	G3 P2	25	Lower abdominal pain	Two previous CS	Lower uterine segment	Expectant	7	CS due to acurate suprapubic pain and decelerations in fetal heart rate (32w)
23	Taipale P[27]	2005	N/A	G2P1	33	Lower abdominal pain	Previous CS	Lower uterine segment	Expectant	1	CS due to increased pain (34w)
24	Rabinowitz R[28]	2006	25	G5 P3	27	Abdominal pain	Previous CS (26w)	Anterior wall	Expectant	3	CS due to uterine rupture (30w)

25	Hunter TJ[29]	2009	27	G3 P1	24	None	Previous CS (inverted T incision) with a subsequent curettage for retained placenta	Fundus	Expectant	3	CS due to significant uterine tenderness (27w) and subsequent hysterectomy for massive haemorrhage dur to placental increta
26	Deka D[30]	2011	25	G3 P0	10	Abdominal pain	Uterine perforation following curettage and subsequent uterine rupture	Fundus	Expectant	22	CS due to acute abdominal pain (32 w) and subsequent caesarean hysterectomy for placenta percreta
27	Iemura A[6]	2013	38	G1 P0	18	None	Previous myomectomy through a vertical incision from anterior to posterior wall via uterine fundus	Fundus	Expectant	12	CS due to lower uterine segment thinning and a vague lower abdominal discomfort (30w)
28	Bouet PE[31]	2016	33	G6 P5	22	None	Five previous CS	Lower uterine segment	Expectant	8	CS (30w)
29	Aiyekomogbon JO[32]	2017	35	G4 P3	21	Lower abdominal pain	Curettage for retained placenta	Fundus	Expectant	10	CS due to ruptured herniated sac with fetal distress (31w)
30	De Sá RAM[33]	2018	28	G5 P3	28	None	Three previous CS	Low uterine segment	Expectant	6	CS due to abdominal pain conistent with peritoneal irritation (34w) and subsequent subtotal hysterectomy for placenta increta
31	Present case	2018	38	G3 P1	22	None	None	Fundus	Expectant	7	CS due to increase of defect size up to 6 cm and with an estimated fetal weight greater than 1500g (29w)

DISCUSSION

Uterine rupture is a potentially catastrophic childbirth complication that is accompanied by high maternal and perinatal morbidity.[1] An Indian study recently identified major risk factors for uterine rupture: multiparity, scarred uterus and less than 18 months duration from last cesarean section.[2] The authors found that unscarred uterine rupture occurred in only 13% of cases with an incidence rate of 0.07%.[2] In another population-based cohort study in the Netherlands, the scarred uterus and the use of uterotonic agents (oxytocin/prostaglandin) were identified as major risk factors; but again 13% of uterine ruptures happened in unscarred uteri with an incidence rate of 0.008%.[3] Furthermore, Golan et al.[4] described uterine rupture as a complication associated with acute symptoms and / or blood loss. The authors also noticed that rupture of an unscarred uterus tend to be a more dramatic event.[4] The case presented is all the more uncommon since the uterine rupture was asymptomatic, the patient didn't show any sign of labor and her uterus was unscarred.

An amniocele is a herniation of the amniotic sac through a uterine defect and it was first defined by Cotton et al. in 1982.[5] To date, only 30 cases of amniocele were described in the literature, which makes our case the 31st.[5–33] The cases reported have greatly influenced us in our care in particular by informing us about the complications encountered but also the possibilities of management, especially the literature review of Iemura et al.,[6] it is therefore natural to continue their work and try to help practitioners who will face the same pathology in the future. It is from this perspective that we have recovered all cases of amniocele before 34 weeks of gestation reported in the literature and analyzed the pathogenesis but also the chosen management and their outcomes.

The patients aged from 20 to 39 years old,[24,26] the majority of cases (84%) were multiparous with only 5 primiparous (16%).[6,13,18,19,21] This is explained by the fact that obstetric events from previous pregnancies are the leading cause (71%) of amniocele: we can cite as supposed underlining cause previous caesarean sections (39%).[7–10,14,16,22,25,27,28,31,33] followed by a history of curettage (23%).[11,20,23,26,29,30,32] then a history of uterine rupture (9%).[10,12,24] We can add to that a history of myomectomy found in 4 patients of 31 i.e. 12.9%.[6,13,15,19] including 3 primiparous women but also amniocentesis that benefited 2 patients of 31 ie 6.5%.[17,18] It can therefore be affirmed that any obstetrical or gynecological event that can damage the integrity of the uterine wall can lead to its weakness and explain the appearance of a uterine defect from which herniation of the amniotic sac can exceptionally occur. It is important to note that in almost 10% of the cases, including ours, no underlining cause could be identified.[5,21] The localization of the uterine defect seemed to be correlated to its origin, thus fundic localization was the most frequently found (58%) and might be related to a history of curettage, myomectomy or uterine rupture followed by lower uterine segment localization (39%) that might be related to previous caesarean section. The history of iatrogenic uterine lesion could not be the only cause of the appearance of an amniocele. Indeed, as suggested by Iemura et al.,[6] diffuse uterine leiomyomatosis or diffuse adenomyosis could decrease the stretching capacities of the uterine musculature, therefore to compensate, the uterus will stretch the known areas of weakness: the lower segment and the uterine fundus, which is in line with the most frequently found localization in our literature review.

In the literature, the discovery of the amniocele was possible with a routine ultrasonographic examination in 9 of the 31 cases (29%) including ours,[6,15,16,22,23,29,31,33] but in most cases (71%), its discovery was symptomatic with abdominal pain as the master symptom (61%).[5,7–14,17–19,21,26–28,30,32] followed by a sensation of uterine cramp (12.9%).[9,12,20,25] or vaginal bleeding (9.7%).[9,25] These mild to moderate symptoms contrast with the severity of uterine rupture described in the literature, which confers amniocele the character of relatively silent uterine rupture.

The management of amniocele depends on several factors including the desire to continue the pregnancy of the family, term at the time of diagnosis, fetal viability, severity of symptoms, thickness and size of uterine defect. Therapeutic strategies can be classified into three categories: termination of pregnancy, surgical repair of uterine defect, and expectancy with strict supervision. To date, there is no consensus on the therapeutic strategy to adopt to the discovery of an amniocele, analysis of cases reported takes all its interest.

The termination of pregnancy was chosen in 13 out of the 31 cases (41%).[7–19] The diagnosis of amniocele was, on average, discovered lately compared to all the cases: 26 ± 6.0 weeks of gestation versus 23 ± 6.7 for all the cases. In 4 out of 13,[8,11,12,18] the term of pregnancy greater than 31 weeks of gestation allowed fetal extraction in emergency to avoid more serious complications. In 2 cases,[7,14] signs of fetal distress detected at fetal heart rate recording indicated immediate fetal extraction. For the remaining cases, the practitioners felt that the excessive uterine defect represented an imminent risk of complete uterine rupture and emergency cesarean section was required despite the gestational age.

Surgical repair of the uterine defect aimed to reintroduce the amniotic sac inside the uterine cavity and suture the gap. This technique was confronted with two main difficulties: the amniotic sac could be adherent to the uterine defect or could be too important in volume making its reintroduction impossible as for the case of Aiyekomogbon Jo et al.,[32] but there is also a risk of preterm labour that can lead to its failure as for the case of Gorthi S et al.,[22] In the 5 cases,[20–24] where the

operation was carried out, the amniocele was discovered early at 17,2 ± 7,9 weeks of gestation compared with all the cases (23 ± 6.7). It has significantly lengthened the pregnancy with a latency period (interval between diagnosis and delivery) of 15.6 ± 7.9 weeks compared to the cases where the expectancy management was chosen with an average latency period of 8.8 ± 5.7 weeks.

Finally, expectancy with hospitalization for a strict clinical and radiological follow-up was chosen in 13 out of 31 cases (41%) including ours.[5,6,25–33] It was imperative to warn the family of the possible risks of such a choice, in particular of a fetal distress found in 5 out of 13 cases (38%),[5,25–27,32] of placental accretisation (23%).[29,30,33] but also and above all a complete uterine rupture (7.7%),[28] or even a rupture of the amniotic sac (7.7%).[32]

There is another very important point to raise: which pathway to use for fetal extraction. This is a point that has sucked many debates within our team. We believe that making an incision through the usually fibrotic seat defect, decreases the risk of uterine inertia.

CONCLUSION

Amniocele is an uncommon pathology whose management is complicated. Practitioners need to adopt the best course of action depending on the situation: surgical repair when possible remains the method that has shown the best results, but the expectant attitude with strict supervision can also be beneficial and preserve the pregnancy.

Abbreviations

MRI: Magnetic Resonance Imaging

Declarations

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Availability of data and materials

Supporting material is available if further analysis is needed.

Competing interests

The authors declare that they have no competing interests.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Ethics approval and consent to participate

Ethics approval has been obtained to proceed with the current study. Written informed consent was obtained from the patient for participation in this publication.

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