Case Report

A Molar Pregnancy as a Cesarean Scar Pregnancy: About the Ninth Case and Literature Review

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Abstract

Ectopic pregnancy and molar pregnancy are not rare events. However, cesarean-scar pregnancy is a rare form of ectopic pregnancy and the combination of cesarean scar pregnancy and molar pregnancy is an extremely rare event. Only a small number of cases have been reported in the literature. This is the ninth case in the world.

Here a case report about a molar pregnancy on cesarean section scars ectopic localisation and a review of the literature.

Keywords: Cesarean-scar pregnancy; Molar pregnancy; Magnetic resonance imaging; Hysterctomy

Introduction

Ectopic pregnancy and molar pregnancy are not rare events. However, cesarean-scar pregnancy is a rare form of ectopic pregnancy and the combination of cesarean scar pregnancy and molar pregnancy is an extremely rare event. Only a small number of cases have been reported in the literature. This is the ninth case in the world. Here a case report about a molar pregnancy on cesarean section scars ectopic localisation and a review of the literature.

Case Presentation

We report the case of S.R, 43 -years-old lady, g5p3. Her past medical history was significant for obesity (bmi 30) and essential hypertension under medication. Her past obstetrical history included previous left tubal ectopic pregnancy, one previous uncomplicated vaginal delivery, and two previous cesarean sections nine and six years ago. Her past surgical history was remarkable for a left salpingectomy for the ectopic pregnancy and umbilical hernia repair.

She had been diagnosed with a spontaneous abortion one month previously after a vaginal bleeding, and an initial value of HCG level at 80600 mUI/ml with a significant decrease the next day to 7380 mUI/ ml. The patient was referred by her treating doctor with a history of persistent bleeding after a spontaneous miscarriage and a pathology result showing an incomplete hydatidiform mole.

On admission to our unit, the patient had a slight vaginal bleeding. She was hemodynamically stable. Physical examination was unremarkable. Transvaginal ultrasonography showed: The uterus was normal in size, with a thin endometrial echo and no evidence of an

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*Corresponding author: Inès Hfaiedh, University Hospital Habib Bougatfa Bizerte, 7000 Tunisia intrauterine gestational sac. There were a 37.4 mm \times 25.5 mm complex heterogeneity echo conglomeration with multiple cystic dark areas, multiple anechoic areas and multiple liquid dark areas. There were abundant blood flow signals inside the lesions. These lesions were located in the uterine isthmus with a doubt about the involvement of cesarean section scar (Figure 1). The findings were suspicious for a molar pregnancy at the cesarean scar site. The patient's HCG level at this time was 27300 mIU/ml (Figure 2).

In our case, MRI was performed to further evaluate the mass and blood flow near the cesarean scar. The result showed a group of abnormal cystic components signal on the anterior wall of the uterine isthmus incision, which was suspected to be a CSP.

During the first day of hospitalization, she presented an important vaginal bleeding. Careful dilatation and suction curettage under Ultrasound guidance was performed; abundant grape-like tissue mixed with blood clots was shaved out. Bleeding was initially brisk but decreased substantially by the time all adherent placental tissue was removed and intrauterine tamponade balloon by Foley bulb



Figure 1: Magnetic Resonance Imaging (MRI) demonstrated an irregular mass in the proximal isthmus. A: The lesion was hypointense to hyperintense on T1-weighted image; B: The lesion was hypointense to hyperintense on T2-weighted image; C: Heterogeneous hyperintensity was seen on diffusion-weighted image; D: The lesion was hypointense on apparent diffusion coefficient maps; E: Gadolinium-enhanced magnetic resonance imaging indicated that the edge of the mass was enhanced in the arterial phase; F and G: The edge of the mass was persistently enhanced in the venous and delayed phases; H: Sagittal T2-weighted image showing localized widening of the upper cervical canal

was set up for 24 hours. Her hemodynamic status remained stable during and after the procedure. After 06 hours, the patient presented a recurrent important vaginal bleeding; she underwent an emergency hysterectomy because we don't dispose of emergency uterine arterial embolization in our department.

During the operation, 40 mm \times 30 mm of pregnancy material was observed in the old cesarean section. After dissecting the bladder from the peritoneum (Figure 3). The final pathology revealed atypical chorionic villi in keeping with a partial hydatidiform molar pregnancy in a uterine scar. The HCG level was 530 mIU/mL on the second postoperative day. The patient was discharged with HCG follow-up according to molar pregnancy follow-up protocols during six months. Weekly monitoring of HCG levels noted stagnant levels making the diagnosis of persistent trophoblastic disease; a FIGO score is equal to one. Two cycles of methotrexate-based chemotherapy resulted in HCG level navigation. The patient then received two more cycles of chemotherapy for consolidation.



Figure 2: Intraoperative image of scar ectopic pregnancy with impending rupture and bulging scar seen in the lower segment.



Figure 3: Final Diagnosis Histological examination confirmed the diagnosis of hydatidiform mole in a uterine scar.

Outcome and follow-up

This patient will be monitored clinically at 3, 6 and 12 months after HCG navigation. Follow-up of HCG levels every 15 days for 16 weeks and then every 2 months for 12 months for our patient (low risk according to the FIGO score).

Discussion

Caesarean scar pregnancy is one of the rarest forms of ectopic pregnancy whose frequency may increase due to changes in obstetric practices and especially the continuous increasing incidence of caesarean section worldwide. Cesarean scar pregnancy occurs into 1800-2200 pregnancies and accounts for 6% of all ectopic pregnancies in women who have previously had a cesarean delivery [1]. On the other hand, an ectopic pregnancy and a molar pregnancy has been described in medical journals less than 300 times [2,3]. The combination of both events is exceedingly rare. Only eight cases of molar pregnancy found in a cesarean scar have been found in the literature and we report the ninth case (Table 1).

The myometrium of the uterus usually thins and merges with the thin and fibrous scar after caesarean section, so hydatidiform moles in the uterine scar may have severe complications, such as uterine rupture, hemorrhage, hysterectomy, and serious maternal morbidity. A caesarean scar molar pregnancy engages the vital prognosis either by hemorrhage or by early uterine rupture [4]. Among the previous eight cases reported, there was one case of cesarean scar invasive molar pregnancy [5], five cases of partial molar pregnancy [6-10], one case of complete molar pregnancy [11], and one case in which the author did not specify the type [12].

Clinicopathological features of the nine cases, including our case, are listed in Table 1. The median maternal age was 34 years (range from 28 to 44 years). The clinical manifestations were not fully identical, but the most common symptom was vaginal bleeding for more than 1 mo (7/9 cases, 77%). Other presentations included symptoms of pregnancy [11], abdominal pain [7,9], and only one patient was asymptomatic on admission [11]. Trans-vaginal ultrasound scan plays a major role in diagnosing this condition. Only in two cases (two out of nine) the ultrasound didn't show the scar pregnancy and the diagnosis was made by laparotomy [11,12].

To date, MRI has not been used very often in the diagnosis of hydatidiform mole embedded in the scar on the uterus. Only in three out nine of cases. We performed a literature review and found that MRI was only rarely used in the diagnosis of hydatidiform moles. The use of this technique was not considered to add value in the diagnosis of CSP according to a new paper published in 2022 [13].

Gestational Trophoblastic Disease (GTD) encompasses a spectrum of interrelated tumors, including complete and partial hydatidiform mole, placental-site trophoblastic tumor and choriocarcinoma, which have varying propensities for local invasion and metastasis. Here, we report a patient with a partial hydatidiform mole embedded in cesarean scar tissue. Thus, molar pregnancy is confirmed, molar pregnancy should be managed accordingly to avoid Gestational Trophoblastic Neoplasia (GTN) which can arise following an ectopic molar pregnancy as it was seen with our patient. Histopathology should be sought and HCG levels should be monitored after surgical treatment [3].

Conclusion

Molar pregnancy on a scar remains an exceptional event. Differentiating molar cesarean section pregnancy from normal cesarean section pregnancy is difficult because of common symptoms that are why misdiagnosis rate and missed diagnosis rate of gestational trophoblastic diseases on CSP are high. The early diagnosis may reduce the bleeding and avoid hysterectomy. The diagnosis must be made by a good ultrasound evaluation and the anatomopathological

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	Publication	Cases	Age	Number OF Previous Cesarean Deliveries	Symptoms	Diagnosed By	Management	Histologic examination	Beta hCG Resolution (weeks)
Wu et al. [4]	2006	1	31	l (4 years ago)	Persistent symptoms after dilatation and curettage for suspected partial mole	Ultrasound and pathology	Careful dilatation and suction curettage under ultrasound guidance	incomplete hydatidiform mole	Not mentioned
Michener and Dickinson [5]	2009	1	33	2	Vaginal hemorrage	Pathology	Intragestational sac methotrexate injection (50 mg). Then Systemic methotrexate two months later for a plateau in β hCG levels decline, then ten months after initial management she comes with life-threatening haemorrhage and required an emergency hysterectomy	persistent molar tissue in the scar	Not mentioned
Jin et al. [6]			44	2	Vaginal bleeding Lower abdominal pain	Ultrasound and pathology	ultrasound-guided suction and curettage	a partial hydatiform mole	Not mentioned
Ko et al. [7]	2012	1	34	2	Persistent symptoms after medical termination	Ultrasound and pathology	Suction evacuation under ultrasound guidance then a uterine artery embolization was performed to control the bleeding	a partial hydatiform mole	7weeks
Kaluarachchi et al. [8]	2013	1	40	2	Asymptomatic PUL	Laparotomy & Pathology	Hysterectomy	complete hydatidiform mole	Not mentioned
Vimercati et	2016	1	34	1	Vaginal	Ultrasound and	Local And Systemic MTX	Hydatidiform	Not
Ling et al. [10]	2018	1	28	1 (one year ago)	Vaginal bleeding and abdominal pain	Ultrasonography MRI & Pathology	Uterine arterial embolization & careful suction evacuation under ultrasound guidance	incomplete hydatidiform mole	9 weeks
Hao ru Jiang [11]	2020	1	35	1	Vaginal bleeding	Ultrasonography MRI & Pathology	Suction evacuation Uterine arterial embolization chemotherapy	Hydatidiform mole	Not mentioned
Present case	2022	1	43	2 (6 years ago)	Persistent vaginal bleeding after spontaneous miscarriages	Ultrasonography MRI & Pathology	Suction evacuation under ultrasound guidance and intrauterine tamponade balloon by Foley bulb then Emergency hysterectomy chemotherapy	incomplete hydatidiform mole	9weeks

Table 1: A summary of all the reported cases of cesarean scar molar pregnancy in the literature.

result. A regular and disciplined follow-up of the patient is necessary. On one hand, the management remains uncodified due to the rarity of the cases. On the other hand, the incidence of pregnancy on scar is clearly rising, given the current obstetrical practices.

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