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Cesarean scar ectopic partial molar pregnancy: A case report and a review of literature

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ABSTRACT

A scar ectopic pregnancy exhibiting hydatidiform features is an extremely rare and clinically challenging entity. Delayed diagnosis and failure to treat such cases promptly can lead to devastating consequences. In this report, we present a case of cesarean scar ectopic partial molar pregnancy in a 37-year-old woman who presented with complaints of vaginal discharge with streaks of blood and lower abdominal pain. Diagnostic laparoscopy revealed an abnormal mass of brown soft tissue in the anterior wall of the uterus, measuring 13.0 × 9.0 × 2.0 cm, raising suspicion (in the context of elevated serum human chorionic gonadotropin levels) of a scar ectopic pregnancy. Open laparotomy was performed, and the scar ectopic mass was successfully removed. The histologic examination of the tissue revealed a partial hydatidiform mole. The patient experienced a full recovery post-operatively, with serum human chorionic gonadotropin levels gradually declining to normal values. This report is unique in its presentation of the clinicopathological features of cesarean scar ectopic molar pregnancy and the successful management of the condition.

1. Introduction

Cesarean scar pregnancy (CSP) is an uncommon, potentially life-threatening variant of ectopic pregnancy. It is, by definition, a pregnancy that results from implantation of the embryo into the myometrial site of a cesarean section scar [1]. Although CSPs were once considered to be extremely rare, their incidence is increasing as a consequence of the rising rates at which cesarean sections are being performed. Furthermore, the use of high-resolution ultrasound scans is increasing the number of cases being detected and hence reported [2]. According to Ouyang et al., the overall estimated incidence of CSPs is 1 per 1688 pregnancies [3]. CSP accounts for 6% of all ectopic pregnancies in women with at least one previous lower uterine segment scar [4].

Molar pregnancy is a type of gestational trophoblastic disease (GTD), where abnormal trophoblasts with a neoplastic potential implant in the uterus with an incidence of 0.6–8 per 1000 pregnancies [5]. The condition compromises two closely related but genetically different types of abnormal pregnancies: complete moles and partial moles. Both carry the potential for malignant transformation, although the risk is significantly greater for complete moles [5]. Many malignant forms arise from molar pregnancy, such as placental-site trophoblastic tumor, gestational trophoblastic neoplasia (GTN), invasive mole, choriocarcinoma and epithelioid trophoblastic tumor [5].

The concomitant occurrence of CSP and molar pregnancy is extremely rare. We describe the laparoscopic management of a patient with partial mole implanted in a cesarean section scar.

2. Case Presentation

A 37-year-old Middle Eastern woman (para 3 + 1) who had had two spontaneous vaginal deliveries, one lower transverse cesarean section (LTCS), and one miscarriage, presented to the emergency department four months after a miscarriage complaining of persistent vaginal discharge with streaks of blood and lower abdominal pain. Her medical history was remarkable for one emergency LTCS four years prior to presentation due to late decelerations and fetal distress; the surgery was

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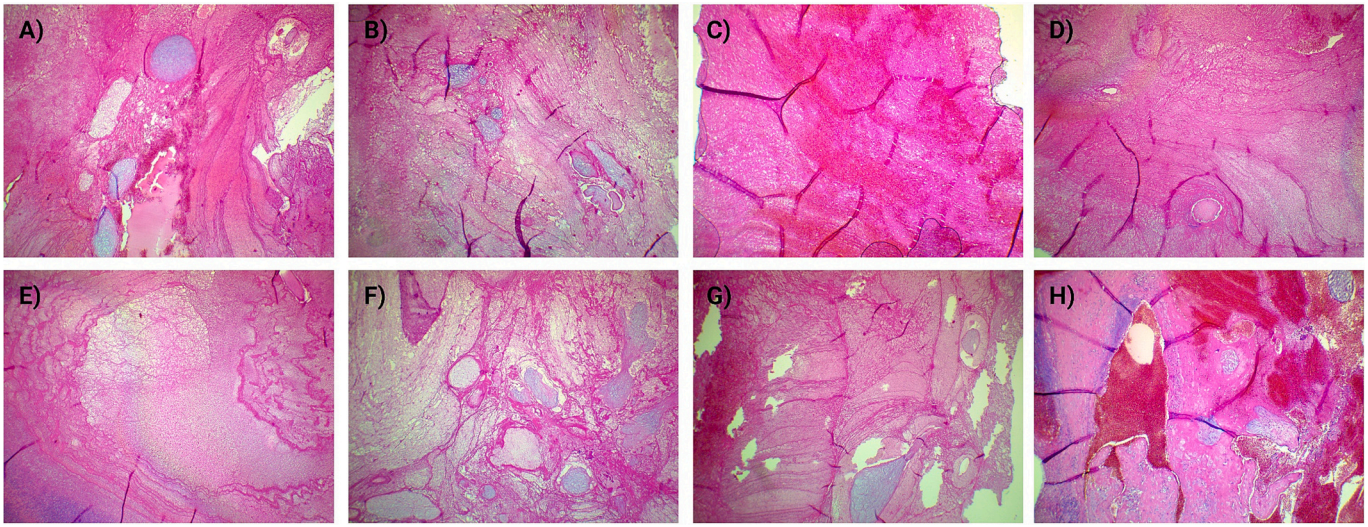

Fig. 1. Histopathological features of the Cesarian section scar ectopic molar pregnancy (A-H) shows hyalinized hydropic chorionic villi, few of which show eccentrically trophoblastic proliferation with mild atypia, favoring the diagnosis of a partial mole.

uncomplicated and delivery of the baby was successful.

The patient was vitally stable. Speculum exam was unremarkable and showed a normal-looking, healthy vulva, vagina, and cervix. The serum human chorionic gonadotropin level (β-hCG) was high (43 IU/L), and the patient was anemic with a hemoglobin of 8 g/dL, a mean cell volume of 76 μm^3, and a hematocrit of 23.8%. An ultrasound (U/S) scan was significant for a bulky uterus, with multiple fibroids, mild free fluid, with no abnormalities detected in the adnexa. However, the endometrial lining was not assessed.

The patient was started on amoxicillin/clavulanic acid and admitted for diagnostic laparoscopy (DL) for suspected ectopic versus molar pregnancy. At DL, the pelvic cavity was healthy, and both tubes and

Table 1
Demographics, symptoms, diagnostic tools, and management plan of reported Cesarian section ectopic molar pregnancies.

Study ID	Age	Symptoms	Diagnostic tools	U/S findings (β-hCG levels IU/L)	Management	HM type
Wu et al. [13]	31	Vaginal bleeding after a miscarriage and lower abdominal pain	Transvaginal U/S and β-hCG	An irregular gestational sac, encircled by a placenta with multi-cystic spaces (61,798)	D&C under U/S guidance	Incomplete HM
Michener and Dickinson [14]	30	Not reported	U/S, and β-hCG	Not reported (9800)	Intragestational sac and systemic methotrexate initially, followed by emergency hysterectomy due to life-threatening hemorrhage	Not reported
Potdar et al. [15]	40	Asymptomatic	U/S and β-hCG	Gestational sac at the site of Cesarian scar (106,500)	Shirodkar's cervical suture followed by U/S guided surgical evacuation	Incomplete HM
Jin et al. [16]	44	One month of irregular vaginal bleeding and lower abdominal pain	Transvaginal U/S and β-hCG	Irregular gestational sac, encircled by a placenta with multi-cystic spaces (94,724)	D&C under U/S guidance	Incomplete HM
Ko et al. [17]	34	Unresolved symptoms of pregnancy for three weeks following surgical termination of pregnancy	U/S and β-hCG	A 23 × 22 mm mass near the Cesarian section scar (21,925)	D&C under U/S guidance followed by bimanual compression, oxytocin, and UAE due to bleeding	Incomplete HM
EG Dağdeviren et al. [18]	34	Asymptomatic	Transvaginal U/S and β-hCG	A 28 × 24 mm mass in a Cesarian section scar, reaching the uterine serosa and bulging from the anterior wall of the uterus (59.705)	Laparotomy	Complete HM
Ling et al. [19]	28	Amenorrhea and three days of irregular vaginal bleeding	Transvaginal U/S, β-hCG, and pelvic MRI	A 1.2 × 1.4 × 1.5 cm gestational sac near a Cesarian section scar in the anterior uterine wall (7894)	Bilateral UAE with suction evacuation	Incomplete HM
Liu et al. [20]	35	Irregular vaginal bleeding one month after evacuation of an intrauterine molar pregnancy	Transvaginal U/S and β-hCG	A mass with mixed echogenicity resembling a honeycomb, 26 × 29 mm in size which was bulging slightly outside the uterus (19,600)	Bilateral UAE with intraoperative infusion of methotrexate, followed by D&C under U/S guidance. Uterine balloon compression device post-operative due to unresolved bleeding.	Not reported.
Jiang et al. [21]	35	Vaginal bleeding for more than one month and amenorrhea for more than two months	U/S, β-hCG, and MRI	A 11.5 × 7.5 cm mass at the uterine anterior lower wall, with an anechoic 5.1 × 2.8 cm area inside (1,512,540)	U/S guided suction evacuation, UAE and chemotherapy.	Incomplete HM
Daggez et al. [22]	25	Abdominal pain, three days vaginal bleeding, and amenorrhea for 40 days	U/S, β-hCG, and MRI	A 23 × 25 mm mass with multi-cystic spaces, implanted on the Cesarian section scar (41,616)	Suction curettage with pre- and post-operative intravenous oxytocin	Incomplete HM
Current case	37	Vaginal discharge with streaks of blood and lower abdominal pain	U/S, β-hCG and DL	A bulky uterus with multiple fibroids with changes and mild free fluid (43)	DL proceeded to open laparotomy due to bleeding	Incomplete HM

ovaries were healthy with no bleeding, but an abnormal mass of brown soft tissue was detected in the anterior wall of the uterus measuring 13.0 × 9.0 × 2.0 cm. The picture was suspicious for scar ectopic pregnancy. Dissection of the mass was attempted with resultant moderate bleeding, and a decision to proceed with open laparotomy was then taken. During the laparotomy procedure, the bladder was successfully dissected and pushed down, both ureters were checked, and the scar ectopic mass was removed completely. The uterus was then closed in two layers, hemostasis secured, and a drain was inserted. The patient was estimated to have lost 2 L of blood; she was given intraoperatively two units of packed red blood cells and two units fresh frozen plasma.

Biopsy of the removed tissue (Fig. 1) showed hyalinized hydropic chorionic villi, few of which showed eccentrically trophoblastic proliferation with mild atypia (these features favor the diagnosis of a partial mole). A molar CSP diagnosis was established. Recovery after the operation was uneventful, and follow-up with serial β-hCG levels was implemented. A significant decline in β-hCG levels to less than 0.1 IU/L was observed one month after the operation.

Due to the rarity of our case and the lack of evidence in the literature, the patient was followed every 3–6 months after the normalization of β-HCG levels. Six months after the initial presentation, the patient's follow-up did not yield any significant emerging events.

3. Discussion

The exact pathophysiology behind the development of ectopic cesarean scar pregnancies is yet to be fully understood. However, a possible mechanism is that damage to the myometrium inflicted by the cesarean incision creates microscopic tracts through which an implanting blastocyst pathologically invades [6]. In keeping with this, it was originally believed that cesarean incisions usually heal without any complications, yet, recently, some authors have described defects in cesarean scars picked up on trans-vaginal U/S that imply impaired wound healing. These are typically seen as anechoic, triangular areas. It is worth noting that 24%–88% of women with previous cesarian section have these defects and are mostly asymptomatic [7]. Despite the indefinite pathogenesis-related data, it is suggested that a short time interval between a cesarean section and a subsequent pregnancy can impede scar healing and thus contribute to scar implantation [6].

Along with the rise in the use of cesarean section in the last decade from 5 to 30% of deliveries, an increase in maternal and neonatal short- and long-term complications has been documented [8]. One of these is CSP, defined as a rare complication, where the gestational sac or trophoblasts are implanted within or on top of the cesarean scar niche, estimated to complicate 1/1800–1/2500 cesarean deliveries [9]. According to Timor-Tritsch et al., different approaches to treating CSP have been described. In their case series they adopted a conservative approach, with a combination of both intragestational sac injection and systemic methotrexate, which was deemed to be successful in the treatment of early CSP [10]. Less conservative approaches were associated with a high risk of complications, the highest being with dilatation and suction curettage (D&C), systemic methotrexate, and uterine arterial embolization (UAE). Özcan et al. reported on 50 cases of CSP, 39 of which were treated with a transabdominal U/S-guided suction curettage, and 11 using a hysterotomy; 38 and 10, women, respectively, were treated successfully, but one suffered from uterine rupture in the transabdominal U/S-guided suction curettage group, and one suffered from bladder injury in the hysterotomy group [11]. Due to the intricate nature of CSP and the individualism of each case, an agreement on a first-line treatment is yet to be reached.

GTD is a group of rare diseases related to the process of conception characterized by implantation of abnormal trophoblasts. Most GTDs are benign, but some can be malignant. Hydatidiform mole (HM or molar pregnancy) is the benign form with two types. A complete mole constitutes the majority of HM, and is the result of fertilization of an empty ovum by a haploid sperm, with a resultant karyotype of 46, XX or less commonly a 46, XY, and no fetus formation. A partial mole is the product of fertilization of a normal ovum by a sperm with double the number of paternal genetic material or by two normal sperms, with a karyotype of either 69, XXX, 69, XXY, or, rarely, a karyotype of 69, XYY, and only a part of the fetus forms [12]. Both CSP and GTD are rare, and our case combines the two in one presentation.

Literature review identified ten cases similar cases (Table 1). The first case documented was back in 2006 by Wu et al. The patient was presumed to have a partial mole by U/S and serum β-hCG findings and a decision to perform a D&C under U/S guidance was taken. However, the tissue near the scar was missed and another D&C had to be done [13]. Multiple cases have been described since then, with different presentations, diagnostic methods and approaches to management. Presenting symptoms were abnormal vaginal bleeding (7 out of the 11 cases, 63%), lower abdominal pain (4 out of 11, 36%), amenorrhea (3 out of 11, 27%), and unresolving symptoms of pregnancy following a miscarriage; two patients were asymptomatic and in one case symptoms were not specified. For the diagnosis, there was an agreement on the important role played by transvaginal U/S and β-hCG levels. Some proceeded to a magnetic resonance imaging (MRI) scan to confirm the diagnosis. Management has varied between different case reports. Some cases were managed with a D&C, others with an U/S surgical suction evacuation, and some with open surgical interventions [13–22]. In contrast, our case was the only one with a DL approach, yielding better visualization and preparedness to treat the patient's condition. Michener and Dickinson used a different approach: they started with an intragestational sac methotrexate injection, and two months later systemic methotrexate was used for plateauing of β-hCG levels. Due to unresolved molar tissue, a hysterectomy was performed ten months after the initial presentation. [14] Bleeding control techniques prior to interventions have been suggested in a number of these case studies. Ko et al. counseled their patient about the risks of bleeding and the possibility of a UAE or a hysterectomy. They used U/S guided D&C which ended with one liter of blood loss and the need for a UAE [17]. C Ling et al. also considered the high vascularity of their patient's lesion and a UAE was performed. U/S was done to confirm decreased vascularity prior to the D&C [19]. In our case, with the laparoscopic visualization of the pelvic cavity, the CSP was easily identified; however, due to the vascularity of the mass and the resultant hemorrhage after attempted dissection, a decision to proceed with an open laparotomy was then taken and successful removal of the mass was performed.

4. Conclusion

In this report, we present the clinicopathological features of cesarian scar ectopic molar pregnancy and its successful surgical management. Physicians should be alert to the possibility of an ectopic molar pregnancy implanted on a cesarian scar, regardless of risk factors for HM. More research is needed to establish a more comprehensive approach to the diagnosis and management of these rare cases.

Contributors

Rania Al-Bataineh contributed to drafting the manuscript and undertaking the literature review.

Shireen Rawashdeh contributed to drafting the manuscript and undertaking the literature review.

Leen N. Lataifeh contributed to drafting the manuscript and undertaking the literature review.

Saja M. Alzghoul contributed to drafting the manuscript and revising the article critically for important intellectual content.

Ahmed H. Al Sharie contributed to drafting the manuscript and revising the article critically for important intellectual content.

Rawan Obeidat contributed to patient care, conception of the case report, acquiring and interpreting the data, drafting the manuscript and revising the article critically for important intellectual content.

Omar F Altal contributed to patient care, conception of the case report, acquiring and interpreting the data, drafting the manuscript and revising the article critically for important intellectual content.

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Patient consent

Written informed consent was obtained from the patient for publication of this report and associated images.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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