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Molar ectopic pregnancy in a cesarean scar treated with methotrexate – a case report and review of the literature

Mimoděložní těhotenství v molární oblasti s jizvou po císařském řezu léčenou methotrexátem – kazuistika a přehled literatury

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Summary: **Objective:** To present a rare case of partial molar pregnancy implanted in a previous cesarean section scar and summarize the literature. **Case report:** A 33-year-old woman (gravida 5, two prior cesarean deliveries, two spontaneous abortions) presented with vaginal spotting and lower abdominal pain at 6 weeks of gestation. Transvaginal ultrasonography revealed a 28 × 19 mm gestational sac at the cesarean scar site, containing a 4mm fetus with positive cardiac activity. Vacuum curettage was performed under ultrasonographic guidance. Ten days postprocedure, rising serum beta-hCG levels and a persistent cystic mass on ultrasound raised suspicion for molar pregnancy. A single systemic dose of methotrexate was administered. Histopathology confirmed a partial molar pregnancy. The patient's serum beta-hCG levels normalized within 8 weeks. **Conclusion:** Partial molar pregnancy in a cesarean section scar is an extremely rare condition that may present with vaginal bleeding and pelvic pain. Early recognition through ultrasonography and laboratory evaluation, combined with timely intervention including curettage and methotrexate therapy, can lead to complete resolution. Clinicians should consider this diagnosis in patients with previous cesarean sections to ensure optimal outcomes.

Key words: cesarean section scar – molar pregnancy – partial hydatidiform mole – methotrexate – vacuum curettage

Souhrn: **Cíl:** Prezentovat vzácný případ částečného molárního těhotenství implantovaného do jizvy po předchozím císařském řezu a shrnout literaturu. **Kazuistika:** Žena, 33 let (gravida 5, dva předchozí císařské řezy, dva spontánní potraty), se dostavila s vaginálním špiněním a bolestmi v podbřišku v 6. týdnu těhotenství. Transvaginální ultrasonografie odhalila v místě jizvy po císařském řezu gestační váček o rozměrech 28 × 19 mm, který obsahoval 4-mm plod s pozitivní srdeční aktivitou. Pod ultrazvukovým dohledem byla provedena vakuová kyretáž. Deset dní po zákroku vzbudily rostoucí hladiny beta-hCG v séru a přetrvávající cystická masa na ultrazvuku podezření na molární těhotenství. Byla podána jednorázová systémová dávka methotrexátu. Histopatologie potvrdila částečné molární těhotenství. Hladiny beta-hCG v séru pacientky se normalizovaly do 8 týdnů. **Závěr:** Částečné molární těhotenství v jizvě po císařském řezu je extrémně vzácný stav, který se může projevovat vaginálním krvácením a bolestí v pánvi. Včasné rozpoznání pomocí ultrazvuku a laboratorního vyšetření v kombinaci s včasnou intervencí včetně kyretáže a léčby methotrexátem mohou vést k úplnému vyřešení. Lékaři by měli tuto diagnózu zvážit u pacientek s předchozími císařskými řezy, aby zajistili optimální výsledky.

Klíčová slova: jizva po císařském řezu – molární těhotenství – částečný hydatidiformní mol – methotrexát – vakuová kyretáž

Backgroud

Cesarean scar pregnancy (CSP) is a rare form of ectopic pregnancy in which the gestational sac implants into the myometrium at the site of a previous cesarean section scar. CSP was first described in the literature by Larsen and Solomon in 1978 [1]. As cesarean section rates have increased over the years, the incidence of CSP has also risen, currently estimated to occur in approximately 1 in 2,216 pregnancies [2].

Gestational trophoblastic disease encompasses a broad spectrum of benign and malignant disorders arising from the trophoblastic cells of the placenta. While partial hydatidiform mole and complete hydatidiform mole represent the

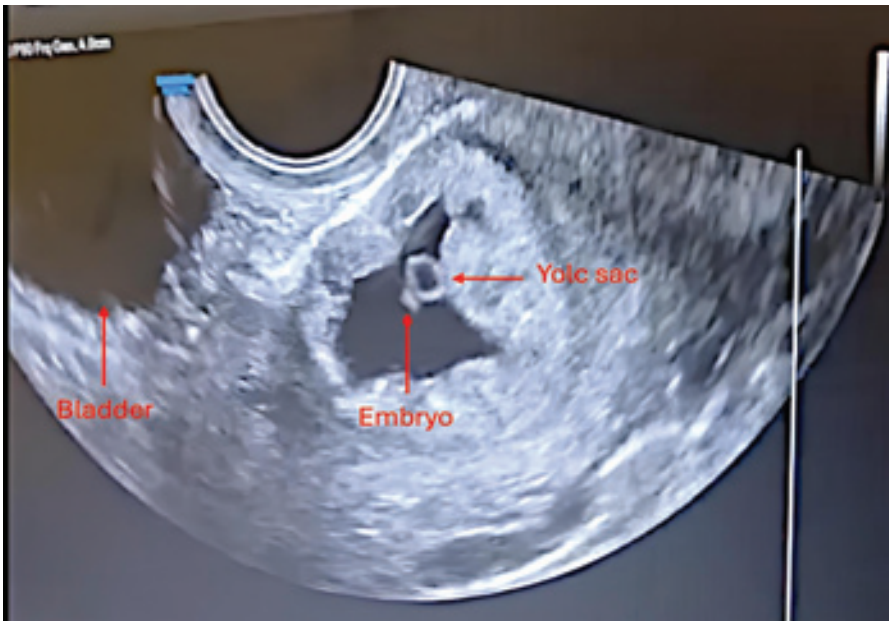

Fig. 1. Transvaginal ultrasound showing a gestational sac adjacent to the previous cesarean section scar, with crown–rump length measurement and fetal cardiac activity, consistent with a partial molar pregnancy.
Obr. 1. Transvaginální ultrazvuk zobrazující gestační váček sousedící s jizvou po předchozím císařském řezu s měřením délky temeno–kostrč a srdeční aktivitou plodu, což odpovídá částečné molární graviditě.

benign forms, the malignant variants include invasive mole, gestational choriocarcinoma, placental site trophoblastic tumor, and epithelioid trophoblastic tumor [3]. The incidence of hydatidiform mole ranges from 0.04–0.9%, depending on the population's ethnic background [4–6]. The presence of a hydatidiform mole within a cesarean scar, representing the coexistence of two rare conditions, constitutes an extremely uncommon clinical entity.

Here, we report a rare case of molar pregnancy occurring at the site of a previous cesarean section scar.

Own observation

A 33-year-old woman presented with vaginal spotting and lower abdominal pain following approximately 2 months of amenorrhea. Based on her last menstrual period, she was estimated to be 6 weeks and 4 days pregnant. This was her 5th pregnancy; her obstetric history included two prior cesarean deliveries and two spontaneous abortions. On sterile speculum examination, the vagina appeared normal, the cervix had a nulliparous appearance, and there was spotting-type bleeding. Transvaginal ultrasonography revealed an anteverted uterus with an endometrial thickness of 11 mm at the fundal level. A gestational sac measuring 28 × 19 mm was observed below the previous cesarean scar, containing a fetus with a crown-rump length (CRL) of 4 mm and positive fetal cardiac activity, corresponding to a gestational age of 6 weeks and 1 day. The cervix was closed, and the cervical length measured 31 mm (Fig. 1). Both ovaries appeared normal on ultrasound.

As fetal cardiac activity was observed, a serum beta-hCG test was not requested. Based on the patient's clinical and ultrasonographic findings, a diagnosis of cesarean scar ectopic pregnancy was established. Following detailed counseling and the acquisition of written informed consent from the patient and her husband, dilatation and curettage were performed under general anesthesia with suprapubic ultrasonographic guidance for CSP termination. Following curettage, a Foley catheter was inserted into the uterine cavity to prevent potential hemorrhage from the cesarean scar, and the balloon was inflated with 30 mL at the scar level. The catheter was removed appropriately one day after the procedure. The patient, who experienced no postoperative bleeding, was discharged in stable condition.

The patient's serum beta-hCG level measured 6 days after the procedure was 10,746 mIU/mL, and ten days later, it had risen to 11,469 mIU/mL. A transvaginal ultrasound performed ten days post-procedure revealed a 27 × 30 mm heterogeneous cystic mass extending outward from the cesarean scar site, with detectable blood flow on Doppler imaging (Fig. 2). The combination of rising serum beta-hCG levels and ultrasound findings raised suspicion for a molar pregnancy. The patient and her husband were informed of the clinical findings. Laboratory investigations and a chest X-ray were performed, all of which were within normal limits. The hospital's pathology department was contacted to expedite the pathological evaluation. Meanwhile, the patient was administered a single dose of methotrexate (50 mg/m^2). Seventeen days after the procedure, the pathology report confirmed the diagnosis of a partial molar pregnancy (Fig. 3). During follow-up, the patient's serum beta-hCG level became negative approximately 8 weeks after the procedure. The heterogeneous cystic appearance previously observed at the cesarean scar site on ultrasound resolved by approximately 8 weeks.

Discussion

CSP and GTD are rare conditions; our case of cesarean scar molar pregnancy, which combines these two pathologies, is therefore extremely rare. In cases where timely diagnosis is not established and the appropriate treatment procedure is not applied, the risk of

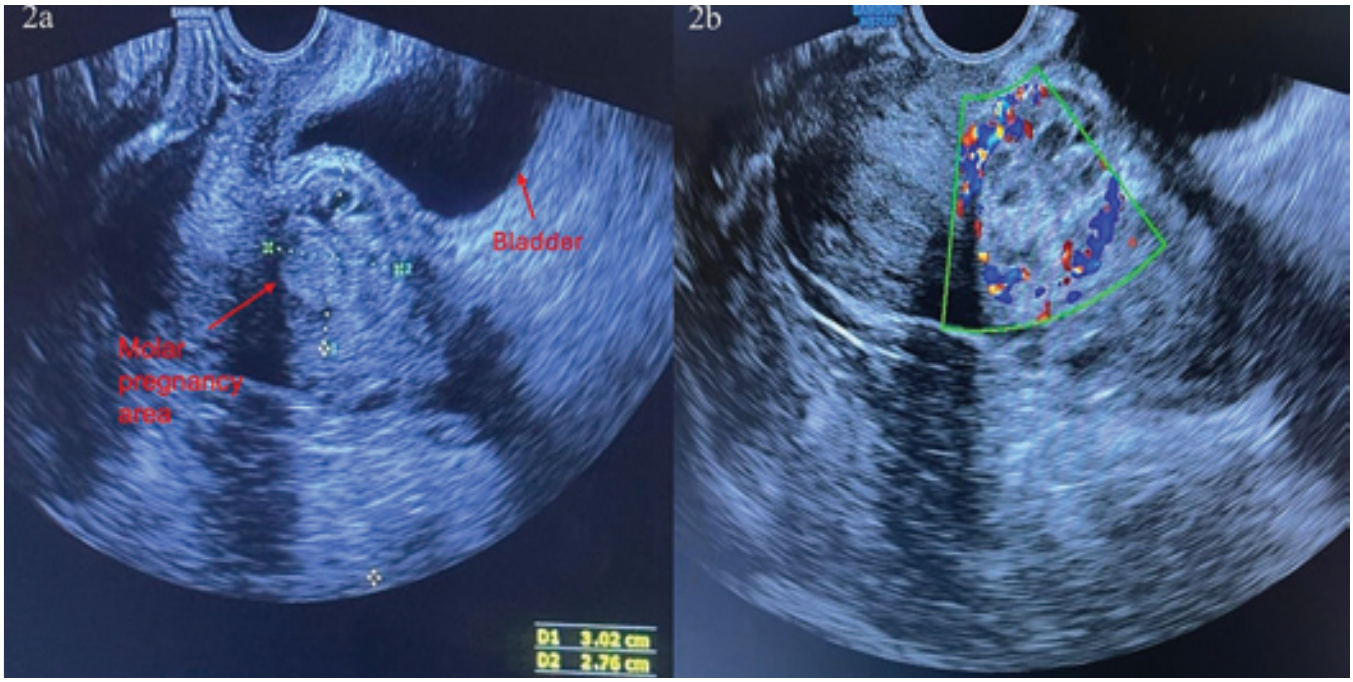

Fig. 2a) After revision curettage, a 30 × 27 mm heterogeneous molar pregnancy lesion protruding toward the serosa was detected at the site of the previous cesarean section scar. The arrow indicates the uterine bulge toward the bladder, considered a pathognomonic feature of cesarean scar ectopic pregnancies. 2b) Colour Doppler ultrasound demonstrating vascularity within the molar pregnancy lesion.

Obr. 2a) Po revizní kyretáži byla v místě jizvy po předchozím císařském řezu detekována heterogenní těhotenská léze moláru o rozměrech 30 × 27 mm, vyčnívající směrem k seróze. Šipka označuje děložní vyboulení směrem k močovému měchýři, které je považováno za patognomický rys mimoděložních těhotenství po císařském řezu. 2b) Barevný Dopplerovský ultrazvuk prokazující vaskularitu uvnitř těhotenské léze moláru.

hemorrhage and uterine rupture is high. Despite these risks, data regarding the optimal management of cesarean scar molar pregnancy remain limited.

We conducted a literature review to identify cases similar to this pathology, which has a low incidence. To the best of our knowledge, only 13 cases have been reported in the literature (Tab. 1) [7–19]. The first documented case was reported in 2006 by Wu et al. In the case reported by Wu et al., molar pregnancy was suspected based on ultrasound findings and β-hCG levels, and suction curettage was performed. A second vacuum aspiration was performed for the patient with persistent vaginal bleeding [7]. Since this case, various cases of cesarean scar molar pregnancy have been described, in which different diagnostic methods and treatment modalities were applied. The most common presenting symptom is vaginal bleeding, which is usually

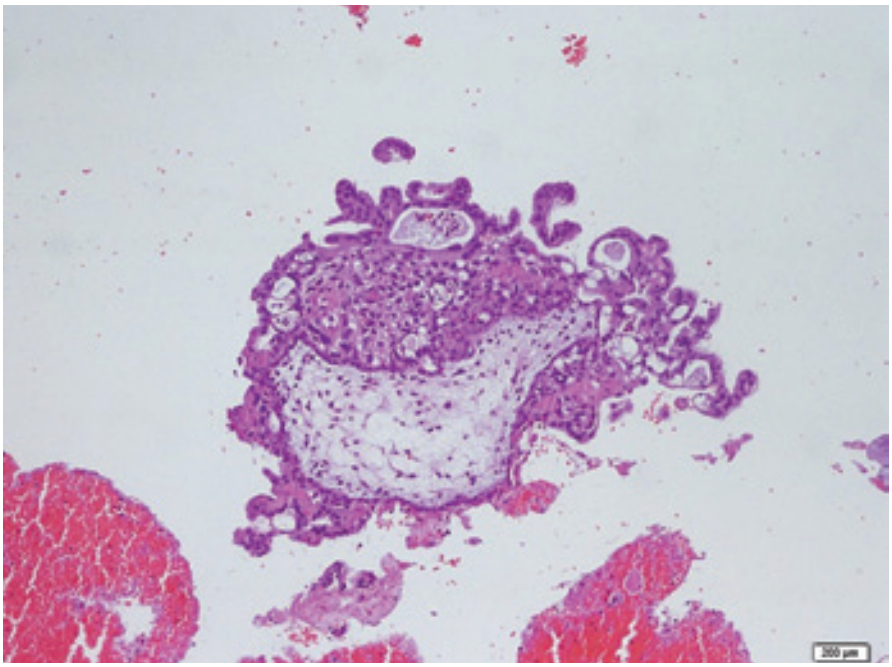

Fig. 3. Circumferential trophoblastic hyperplasia (hematoxylin and eosin stain, 100×).

Obr. 3. Cirkumferenční trofoblastická hyperplazie (barvení hematoxylinem a eosinem, 100×).

Tab. 1. Summary of reported cases of molar pregnancy in a previous cesarean section scar.
Tab. 1. Souhrn hlášených případů molárního těhotenství v jizvě po předchozím císařském řezu.

Reference	Age	Presentation	Gravida/ parity	Serum β-hCG (IU/L)	Diagnostic method	Treatment
Wu et al., 2006 [7]	31	vaginal bleeding, lower abdominal pain	8.1	61,798	ultrasound, pathology	dilation and curettage
Michener et al., 2009 [8]	33	vaginal bleeding	5.2	161	emergency hysterectomy	intragestational sac MTX injection
Jin et al., 2011 [9]	44	vaginal bleeding, lower abdominal pain	2.2	94,724	transvaginal ultrasound, pathology	suction curettage
Ko et al., 2012 [10]	34	asymptomatic	4.2	21,925	ultrasound, pathology	suction evacuation, uterine arterial embolization
Kaluarachchi et al., 2013 [11]	40	asymptomatic	4.2	6,743	laparotomy	hysterectomy
Dağdeviren et al., 2017 [12]	34	asymptomatic	3.2	59,705	transvaginal, ultrasound, pathology	partial resection
Ling et al., 2018 [13]	28	abdominal pain, vaginal bleeding	3.1	7,984	ultrasound, MRI, pathology	uterine arterial embolization, suction evacuation
Jiang et al., 2020 [14]	35	vaginal bleeding	4.1	1,512,540	ultrasound, laboratory tests	uterine arterial embolization, suction evacuation
Daggez et al., 2021 [15]	25	vaginal bleeding	2.1	41,616	laboratory tests, MRI, ultrasound	suction evacuation
Kriplani et al., 2022 [16]	–	vaginal bleeding	–	32,678	intraoperative diagnosis	intramyometrial injection of vasopressin, partial resection
Al-Bataineh et al., 2023 [17]	37	vaginal bleeding, abdominal pain	4.3	43	diagnostic laparoscopy	partial resection
Hosseinimousa et al., 2024 [18]	**Case 1:** 34 **Case 2:** 40	**Case 1:** hypogastric pain, nausea **Case 2:** vaginal bleeding	**Case 1:** 1/1 **Case 2:** 2/2	**Case 1: 27,633** **Case 2: 225,000**	in both cases, the ultrasound and laboratory tests	**Case 1:** suction evacuation and partial resection **Case 2:** suction evacuation and emergency laparotomy (resection of cesarean scar tissue)
Raine et al., 2025 [19]	43	vaginal bleeding	3.2	78,424	transvaginal ultrasound, computed tomography	hysterectomy
Present case	**33**	**vaginal bleeding, abdominal pain**	**5.2**	**11,469**	**transvaginal ultrasound, laboratory tests, and pathology**	**suction evacuation, MTX**

hCG – human chorionic gonadotropin, MRI – magnetic resonance imaging, MTX – methotrexate

followed by pelvic pain. Of the 13 cases reported in the literature, 10 presented to the hospital with vaginal bleeding, similar to our case. The other symptoms include inguinal pain and unexplained pregnancy-related signs. Three patients were reported to be asymptomatic.

In case reports, diagnostic methods show that ultrasound is the most frequently used modality. Laboratory results follow this. Other diagnostic procedures include histopathological examination, magnetic resonance imaging (MRI), computed tomography (CT), and, in some cases, surgical exploration. In our case, the diagnosis was established based on laboratory tests, ultrasound, and pathological examination. Curettage was performed in 8 of the 13 patients, uterine artery embolization in 3, partial resection in 4, and hysterectomy in 2. Michener and Dickinson applied a different treatment procedure; they injected methotrexate into the intragestational sac and subsequently administered systemic methotrexate to induce a plateau in β-hCG levels. During follow-up, an emergency hysterectomy was performed ten months later due to the occurrence of bleeding [8].

In our case, the patient initially underwent vacuum curettage. Ten days later,

molar pregnancy was suspected, and a single systemic dose of methotrexate was administered, resulting in complete recovery. Histopathological examination confirmed the diagnosis of partial molar pregnancy.

Conclusion

This case report presents the clinicopathological features and successful management of an ectopic molar pregnancy in a cesarean scar that resulted in cure. Clinicians should maintain a high index of suspicion for this rare pathology. This case report, supported by a literature review, summarizes the diagnostic and therapeutic approaches to cesarean scar molar pregnancy, emphasizing the necessity of patient-specific, multidisciplinary treatment plans.

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