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Commentary

Commentary on: Hydatidiform mole in the scar of uterus: a case report

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We read with great interest the paper published by Jiang et al. [1]. They illustrated a case of cesarean scar molar pregnancy in which the patient complained of vaginal bleeding and amenorrhea, and the concentration of human chorionic gonadotropin (HCG) increased abnormally and eventually recovered following comprehensive therapy including suction evacuation, uterine arterial embolization (UAE), and chemotherapy. The authors completed a literature review and mentioned that cesarean scar molar pregnancy was associated with intraoperative bleeding, and that performing UAE before surgery could be beneficial to prevent continuous bleeding.

As the authors cited, cesarean scar pregnancy (CSP) is an unusual form of ectopic pregnancy, which is defined as an embryo implanted in the myometrium of a previous cesarean scar [2,3]. Its rate is estimated to be between 1:1800 and 1:2216 pregnancies [4]. However, the prevalence of CSP has been rising with the increasing frequency of cesarean section [5]. Gestational trophoblastic disease (GTD) include complete and partial hydatidiform molar pregnancies, malignant disorders including invasive molar pregnancies, choriocarcinoma, and placental-site trophoblastic tumor. According to the literature, benign hydatidiform mole, whether it is complete or partial, is the most common form of GTD [6]. Therefore, hydatidiform mole is predominantly found in cesarean scar sites. This can result in serious complications, such as uterine rupture, hemorrhage, and uncontrolled hemorrhage [7]. Timely detection, diagnosis and properly coordinated treatment are particularly important to life-saving patient care.

Commonly, CSP is considered to be a rare complication of cesarean section. The exact cause and pathology are still unknown. Nonetheless, up to 72% of cesarean scar pregnancies occur in women who have had 2 or more cesarean deliveries [8,9]. Cesarean scar pregnancies can appear at any time, from implantation to termination, but it has been reported to occur more often in the first trimester [5]. The clinical presentation of CSP is non-specific, with the most common symptom being vaginal bleeding [1,10-12]. However, it also can be an incidental finding in asymptomatic women. As Kaluarachchi et al. [13] reported, the patient had no abdominal pain, vaginal bleeding or any other obvious complaints. In the majority of cases, it can be found that the serum β-hCG levels are markedly increased to varying degrees.

There is no agreement on the best method and criteria to diagnose this rare disease. However, ultrasound examination is usually the first-line diagnostic protocol. Additionally, some researchers have found that the use of three-dimensional power Doppler imaging is a useful tool which might provide important instruction in distinguishing the neovascularization features related to cervical pregnancy [2,5,14-16]. To date, many cases of CSP have been diagnosed by using transvaginal scan (TVS) in the early weeks of pregnancy. Michael and colleagues [17] reported the sensitivity of ultrasound for detecting CSP as 84.6%. Vial et al. [18] proposed the following sonographic criteria for the diagnosis of this condition, which were accepted subsequently by many other authors [8,19]: 1) Empty uterus with a clearly visualized endometrium; 2) Empty cervical canal; 3) Gestational sac within the anterior portion of the lower uterine segment at the presumed site of the cesarean scar; 4) thinned or absent myometrium between the gestational sac and bladder (<5 mm in two-thirds of cases).

Previously, few authors had suggested magnetic resonance imaging (MRI) as an assistant to ultrasonography to increase diagnosis and surgery accuracy [20]. Nevertheless, in order to minimize invasive surgery as much as possible, a detailed and accurate evaluation before surgery is extremely important. MRI is excellent at assessing the pelvic structures owing to its advanced differentiation of soft tissue, spatial resolution and the possibility of a multiplanar imaging [21]. Ash et al [5] indicated that sagittal and transverse T1- weighted images (T1WI) and T2WI could properly display the gestational sac which was implanted in the anterior lower uterus [5,22]. Recently, some authors began advocating the use of MRI if ultrasound failed to identify the typical findings of a CSP [23-25]. In 2016, Yamada et al. [26] reported the first case of ectopic molar pregnancy preoperatively diagnosed using MRI. The mass was located on the right cornu, and its margins were clear. Subsequently, Ling et al. [12] described another case of partial molar pregnancy in which the cesarean scar was preoperatively assessed utilizing pelvic MRI. The lesion demonstrated hypo-intensity on T1WI and mixed with hyper-intensity on T2WI. Discontinuous myometrium of the anterior wall of the uterine isthmus incision was also found, while the uterine serosa was still continuous. The MRI examination of present case was performed after surgery, if the examination had been performed before surgery, whether the patient will be treated differently, we have no evidence. It is undeniable that MRI examination did play an essential role in this case. In addition, Kumar et al. [27] argue that cesarean scar thickness and T2WI intensity measured on MRI can be applied to prognosticate scar dehiscence and uterine rupture. They also recommend that T2WI could be used as a more standardized and objective criterion, therefore justifying the use of this complex inspection in the assessment of scar behavior.

Despite advanced imaging technology, CSP is commonly missed or misdiagnosed as a normal intrauterine pregnancy, abnormal intrauterine pregnancy such as an abortion or trophoblastic disease, or cervical ectopic pregnancy [28-30]. Wu et al. [31] revealed a case of partial molar pregnancy in a cesarean scar originally misdiagnosed as a threatened abortion. Qian et al. [32] reported a case of cesarean scar choriocarcinoma, which was also misdiagnosed as a normal CSP. Usually, the myometrium of the uterus is thin and merges with the thin and fibrous scar from cesarean section, and the placental attachment in the lower segment might lack the decidua basalis and myometrium, only composing of some connective tissue [5,33], so this condition may lead to various complications, such as uterine rupture, massive bleeding, hysterectomy, and severe maternal morbidity. Timor-Tritsch et al. [34] indicated that approximately 107 out of 751 cases (13.6%) were missed or misdiagnosed resulting in numerous complications (heavy bleeding, shock, hemoperitoneum or hysterectomy) which can be as serious as death or loss of fertility.

Because of the rarity and lack of clinical experience, there are no universal treatment guidelines for CSP. In general, early pregnancy termination in the first trimester is highly recommended, because of the high risk of subsequent life-threatening complications. The conventional treatment methods reported in the literature are mainly related to expectant management, medical therapy such as local or systemic injection of methotrexate (MTX), and surgical options, which include dilatation and curettage (D&C) or vacuum extraction under the guidance of ultrasound, UAE, hysteroscopy, laparotomy or laparoscopic excision. Surgical treatment and medical treatment can be performed alone or in combination.

UAE is effective as an adjunct treatment and in emergent cases secondary to persistent bleeding [14]. It can minimize hemorrhage particularly in cases when the trophoblasts are deeply implanted in the myometrium. In present case , comprehensive treatment was applied. However, intraoperative hemorrhage took place, which lead to a subsequent emergency UAE, fortunately the result was satisfactory. Vimercati and colleagues [35] chose local and systemic MTX for patient treatment. However, some side-effects of MTX injection has been reported, which include oral ulceration, nausea, vomiting, bone marrow depression, and bleeding [36]. These seriously limit its application.

Larsen et al. [37] reported the first case of CSP in 1978, they managed the patient with laparotomy, resection through a hysterotomy and reconstruction of the uterine scar dehiscence. It is generally agreed that D&C is not a favorable treatment for CSP as it can cause serious bleeding and uterine rupture [3]. But Liu et al.'s research [38] demonstrated that ultrasound-guided D&C successfully removed 97.67% of CSPs in patients with cesarean scars thicker than three millimeters compared with 50% of those in patients whose scars thinner than three millimeters. Meanwhile, no patients had complications and the level of serum β-HCG decreased to normal within three weeks. On the other hand, Wu et al. [31] treated a partial mole with repeat suction curettage without any medication apart from uterotonic agents. Giampaolino et al. [39] indicated that MTX and D&C represented an effective treatment in a case series of early CSP in a retrospective review, and their study demonstrated an overall complication rate of 22.2%. Vacuum extraction alone was also found to be effective in a series of 12 cases of CSP without any adjuvant intervention [40]. This study had selective requirements for patients: 1) pregnancy <8 weeks gestation, 2) the serum β-hCG levels <10,000 mIU/mL, 3) hemodynamically stable patients, 4) and without evidences of uterine rupture. Surgical excision of CSP by laparoscopy is applicable when the gestational sac is growing towards the bladder and abdominal cavity, the patient should be hemodynamically stabilized during surgery [41]. Additionally, hysterectomy could be an appropriate treatment approach in the surgically unstable patient or the family who desires no further fertility [10].

Jurkovic et al. [8] proposed post-therapeutic follow-up as a weekly outpatient clinical assessment of β-hCG until it is undetectable (<2.5 IU/L) and monthly TVS examinations to assess the size of retained products of conception. To the best of our knowledge, some patients can still retain their fertility after treatment and few information related to the relapse after CSP treatment has been reported. Although Maymon et al. [42] and Wang et al. [43] indicated that the recurrence rate was approximately 4%, and 15.6%, respectively. However, the patient numbers were relatively small in their studies. Further study with larger sample is needed. Thus, the prognosis of CSP is considered to be relatively favorable. There is no consensus on how long to wait until attempting conception again or the risk of future pregnancy [4,7] and little information is accessible on the subsequent fertility rates following treatment. Undoubtedly, patients should be advised to delay novel pregnancy after treatment. Nagi et al. [44] recommend that the interval between an abnormal pregnancy located in the site of previous cesarean scar and a future normal pregnancy should be 12 to 24 months.

In conclusion, Jiang et al. have done a thoroughly reviewed evidence and cases regarding cesarean scar molar pregnancy. Although continued studies and a consensus statement regarding treatment of this disease is needed, this particular case report emphasizes that there is a vital role for uterine arterial embolization prior to surgery, and the authors are cautious about advocating its application. Furthermore, patients should not be treated based on imaging studies, alone. We would like to stress the importance of acceptable treatment selection, as well as detailed consideration of individual patient characteristics and each patient's relevant history. Proper treatment selection can potentially reduce complications, readmissions, number of postoperative visits, and a more thorough understanding of the disease.

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