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Broad Ligament Pregnancy

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Abstract

An ectopic pregnancy implanted between the leaves of the broad ligament is a challenge to diagnose and manage. It can be found in almost any week of pregnancy, and it can present with a great spectrum of symptoms. This makes it necessary for the clinician to keep a high level of alertness and suspicion for this uncommon diagnosis and to be prepared when treating any ectopic pregnancy with poor response to medical treatment. The broad ligament surrounds a number of anatomical structures, structures necessary to be preserved when removing the ectopic pregnancy. This can be a challenge for the surgeon, especially when the patient presents with severe symptoms and the pregnancy is advanced. In these cases, even hysterectomies have been reported, a severe operation especially when the patient is young and without having completed her family planning. Finally, in the last three decades with the increasing widespread of laparoscopy and the added experience, there is a growing effort to manage these ectopic pregnancies with minimally invasive procedures, offering the patient a faster recovery, minimum blood loss, and lower surgical morbidity.

Keywords: broad ligament, ectopic pregnancy, abdominal pain, laparotomy, laparoscopy, hysterectomy

1. Introduction

One of the rarest places of an ectopic pregnancy to be implanted is between the leaves of the broad ligament. Loschge in 1816 made the first known report of a broad ligament pregnancy [1], while Champion et al. in 1938 published a large series of 62 cases [2]. Kennedy in 1925, after recording one broad ligament pregnancy among 613 ectopic cases, calculated the incidence of intraligamentary pregnancy as 1 for every 183,900 pregnancies [3]. Even today when the medical literature is so extended and continuing, the exact incidence of this rare type of ectopic pregnancy is difficult to be exactly calculated. The extensive use of assisted reproductive technology has made even more difficult tracking every case. The rarity of ectopic pregnancies in the broad ligament makes it challenging for the clinician to diagnose, and it needs a high index of suspicion. In most of cases the diagnosis is done during an operation for treatment of a presumed ruptured tubal pregnancy. Despite the fact that most cases are diagnosed in the first trimester of pregnancy, there are a few cases reported that the diagnosis was not made until the third trimester and the final outcome was a

live birth. The rarity of these cases also makes it impossible to find extensive series of patients so as to determine the best treatment, making the clinician rely on previous case reports and the methods used in other treated cases.

2. Anatomy

The broad ligament of the uterus is a double-layer fold of peritoneum (anterior and posterior leaves) extending from each side of the uterus to the lateral pelvic walls and the pelvic floor. It covers the lateral uterine corpus and the upper cervix as well. The structures within the broad ligament (uterine tubes, ovarian artery, uterine artery, ovarian ligament, round ligament of the uterus, suspensory ligament of the ovary, ovary) are considered retroperitoneal. The broad ligament itself is composed of visceral and parietal peritonea that contain smooth muscle and connective tissue [4, 5].

3. Pathophysiology

The way the trophoblast can be implanted retroperitoneally is still not clear. Broad ligament pregnancies are classified as abdominal ectopic pregnancies and can be further classified as primary or secondary. Many of them are considered to have started as a tubal or ovarian pregnancy, which later ruptured intraperitoneally and was implanted again in another location [6]. Studdiford in 1942 [7] suggested three criteria, later modified by Friedrich and Rankin [8], for the diagnosis of a primary peritoneal pregnancy, (i) normal tube and ovaries with no evidence of recent or remote surgery, (ii) absence of any uteroperitoneal fistula, and (iii) the presence of a pregnancy related exclusively to the peritoneal surface and young enough to eliminate the possibility of secondary implantation following a primary nidation in the tube.

Sotus in 1977, in a case report of a retroperitoneal ectopic pregnancy, hypothesized that an ectopic trophoblast could invade and penetrate the peritoneum, which soon thereafter covered the gestational tissues [9].

Another possible way for retroperitoneal implantation is during the embryo transfer in IVF, with uterine perforation and direct retroperitoneal placement. The arguments against this proposal are based on the softness and the flexibility of the catheters used for embryo transfer [7]. Another argument against this mechanism that has been reported is a broad ligament pregnancy after an ultrasound-guided embryo transfer [11].

The above proposals cannot explain how a broad ligament pregnancy can occur in a patient with bilateral salpingectomy. The possibility of a microscopic fistulous tract through which the transferred embryos entered the abdominal cavity has also been proposed by Fisch et al. in 1996 [10]. Apantaku et al., also in 1996, proposed the possibility of recanalization of a fallopian tube stump in a patient with bilateral salpingectomy [11].

Fisch et al. made another proposal trying to explain a reported broad ligament pregnancy. They suggested that the spermatozoa could enter the abdominal cavity through a cornual fistulous tract and fertilize an oocyte already migrated there. This proposal was made when trying to explain a broad ligament pregnancy to a patient with bilateral salpingectomy who had intercourse the day after follicular aspiration [10].

Finally it has to be mentioned that Deshpande et al. in 1999 reported a twin pregnancy in the broad ligament to a patient who underwent in vitro fertilization [12], while in 2001, Phupong et al. reported a twin pregnancy in the broad ligament after spontaneous conception [13].

4. Symptoms

There is a wide range of symptoms which can lead a patient to seek help and find out that she is pregnant with an ectopic pregnancy. Most of the symptoms are the same as for every other ectopic pregnancy, with abdominal pain and vaginal bleeding to be the most prominent. The abdominal pain can be severe and of sudden onset [14] or intermittent and associated with other symptoms [15]. The pain can also be characterized as “mild” or “moderate” [16, 17] or just a “discomfort” [18], or finally it can be gradually increased in intensity [19]. The location of the pain has also to be reported to be related with the side of the ectopic pregnancy [19–21]. The duration of the pain can also vary between different cases, with even an extreme case of 4 months of intermittent pain to have been reported [22]. Finally it is not unusual for a broad ligament pregnancy to be completely asymptomatic and to be found during an early routine visit [11, 12].

The vaginal bleeding, if present, can also be of different severities. Cases with just vaginal spotting [21, 23, 24] have been reported, while in other reported cases, vaginal bleeding was present and more severe [22, 25–31], leading the patient to seek medical assistance.

Other symptoms that have been reported in an early broad ligament pregnancy include nausea [20, 24, 32], chills and fever [20], vomiting [20, 28], and dysuria [15]. These symptoms can be present alone or accompanying others, with various severities and to be evaluated as important or of no importance by the patient.

Despite the modern techniques and progress, there are places (more often in the developing world) where a pregnant woman cannot have access to proper antenatal care, making possible for an intraligamentary pregnancy to advance. In such a case, it is possible for the pregnant woman to seek help because of inability to perceive fetal movement [33, 34] or to be referred to a more specialized hospital due to severe oligamnion [35, 36] or intrauterine growth retardation [35]. Abdominal pain and vaginal bleeding are other possible symptoms for an advanced intraligamentary pregnancy like any other ectopic pregnancy.

It is also possible for a broad ligament pregnancy to have no symptom at all and to be found during a routine visit. Suchánková et al. reported a case where a patient with bilateral salpingectomy was referred to hospital from an assisted reproduction center to solve suspicion of molar intrauterine pregnancy, while the ultrasound and the following laparoscopy revealed a right broad ligament pregnancy [37]. Apantaku et al. in 2006 presented a case where a broad ligament pregnancy was discovered during a routine visit of a patient who had an IVF 6 weeks earlier [11], while Deshpande et al. in 1999 presented the first case of a twin broad ligament pregnancy 7 weeks after IVF [12]. Siow et al. in 2004 reported a case of a 10-week asymptomatic woman who presented for routine antenatal care and was found with a right adnexal mass and empty uterus. This was also the first published broad ligament pregnancy which was managed laparoscopically [38]. Laparoscopy was also used to treat another case presented by Cosentino et al. in 2017 of an asymptomatic woman who presented to be submitted to a noninvasive prenatal diagnosis procedure and was found to have a broad ligament pregnancy [39].

Finally there have been reported cases where the pregnancy went uneventfully until term. Seckin et al. in 2011 reported a case with a primigravid patient who was admitted due to breech presentation and oligamnion. Because of the breech presentation, they decided to proceed with primary cesarean section, where an unruptured gestational sac was found between the layers of the broad ligament with both adnexa being normal [36]. In another case report by Schramm in 1982, a case of a patient who presented with pains every 4–5 minutes for 1 hour was described. Another three mild contractions the next 90 minutes were recorded, and after that

it was difficult to identify the fetal lie and a definite uterine outline. Emergency laparotomy was carried out, and the placenta was found to be attached to the anterior layer of the broad ligament [40]. The above cases were with favorable result; both mother and child survived through gestation and birth. Rakotomahenina et al. in 2014 reported a post-term pregnancy in the right broad ligament. The patient was not followed up during gestation and presented to the hospital at the 44th week. The fetus was dead in transversal position and was extracted from the right broad ligament by laparotomy [41]. This case showed that a broad ligament pregnancy can reach term without any serious symptoms.

From the above it can be concluded that a broad ligament pregnancy can present with a variety of symptoms or no symptom at all. When preparing for an operative solution for an ectopic pregnancy, the clinician must be prepared for this rare presentation, and it needs a high level of suspicion. When conservative management with methotrexate is chosen for an ectopic pregnancy and does not have the expected results, broad ligament pregnancy should also be part of the differential diagnosis.

5. Diagnosis

The diagnosis of an ectopic pregnancy located in the broad ligament is difficult to be established preoperatively. In most cases the broad ligament diagnosis is done during the operation for a misdiagnosed tubal pregnancy, which can be either by laparotomy or laparoscopy. In this case the pregnancy can be found laterally to the uterus, medial to the pelvic side walls, superior to the pelvic floor, and inferior to the fallopian tube [42].

Ultrasound, transvaginal or abdominal, is the method of choice to identify the location of an ectopic pregnancy. If finding the site of an ectopic pregnancy is difficult, the easiest way to rule out an ectopic pregnancy is to identify an intrauterine one [43], something almost 100% possible in a gestation greater than 5,5 weeks [44, 45]. An empty uterus with positive pregnancy test and the presence of a gestational sac or mass outside the uterus gives high suspicion for ectopic pregnancy [46]. A broad ligament pregnancy can grow significantly before giving any symptoms [47], in which the size can lead to distorted anatomy and increased difficulty in making an accurate diagnosis [48]. Phupong et al. in 2003 based on an empty uterus, a mass with a single viable fetus in a gestational sac located just beside the right side of the lower part of the uterus, and the clinical findings and their experience in a previous managed case, they reported a correct preoperative diagnosis by transvaginal ultrasound [31]. These two findings (empty uterus and an ectopic pregnancy mass just beside the lower part of the uterus when using transvaginal ultrasound) are two suggested ultrasonographic clues which can make the clinician suspect a broad ligament pregnancy [31]. Also in 2001 Sharma et al. reported that they managed to diagnose preoperatively a broad ligament pregnancy based on the anterior place with peritoneal reflection of the placenta combined with free fluid in the abdominal cavity and a bulky with thick endometrium uterus [28]. Allibone et al. in 1981 [49] suggested six criteria in order to assist the identification of an abdominal pregnancy: (a) demonstration of a fetus in a gestational sac outside the uterus, or the depiction of an abdominal or pelvic mass, identifiable as the uterus, separate from the fetus [49, 50], (b) failure to see a uterine wall between the fetus or products of conception and the urinary bladder, (c) recognition of a close approximation of fetal parts to the maternal abdominal wall, or gestational products like the placenta, both features alerting the sonographer to the absence of enveloping uterine walls, (d) demonstration of eccentric position and/or abnormal attitude of the fetus, (e)

localization of the placenta outside the confines of the uterine cavity, (f) visualization of the placenta immediately adjacent to the fetal chest and head with no intervening amniotic fluid [51]

Magnetic resonance imaging (MRI) can offer great help to the diagnosis of an ectopic pregnancy, especially when then location is unknown. MRI can also offer valuable information for the preoperative planning so as to avoid cutting in the placenta and reduce the bleeding during surgery [52, 53]. MRI can also highlight the involvement of pelvic structures and organs in an ectopic pregnancy, providing more helpful data to the preoperative planning [46]. Finally, it is also important to remember the contraindication of some contrast agents in pregnancy, which has to be considered when the life of the mother is at risk. In any case an informed consent should be obtained by the patient [54].

Angiogram is another tool which can prove its usefulness preoperatively, in order to reveal the location of the placental vessels, while embolization and pre- or postoperative can be used to control hemorrhage. Embolization could also be used for difficult-to-reach vessels intraoperatively [49, 55], although the location of a broad ligament pregnancy and the fact that such a pregnancy as often as not presents with acute abdomen limit the possible use of the above techniques.

6. Management

The rarity of this type of ectopic pregnancy makes it impossible to have prospective trials so as to conclude the best possible treatment. The reported cases that already can be found in the international medical literature can propose different management options [56], taking into consideration the different means every managing clinician can use.

Broad ligament pregnancies are difficult to be diagnosed before the time of surgical management. Therefore, methotrexate cannot be considered the first-line treatment for these conditions but may have been initiated if diagnosed incorrectly as tubal pregnancies [22, 57]. Despite that, Direkvand-Moghadam et al. in 2015 reported a case where they treated a left broad ligament pregnancy with a single dose of methotrexate in a patient with left abdominal pain, 33 days since last menstrual period, β -hcg = 212 IU.L, and no abnormal sonographic findings. The maximum level of β -hcg was 659 IU.L, the forty sixth day after the last menstrual period, and returned to normal 3 weeks after the injection [21].

The way a patient with broad ligament pregnancy presents (abdominal pain in the first trimester) makes it very difficult to follow conservative treatment in order to achieve a favorable result such a live birth. Cachón López et al. in 1989 reported such a case of an 18-year-old woman diagnosed with a 30.4 week abdominal pregnancy. The diagnosis was established by clinical and imaging tests. They decided to keep the patient under close surveillance for 2 weeks and to induce fetal lung maturation. At 32.4 weeks of gestation, a laparotomy was performed giving birth to a 1.100gr baby. The placenta was found on the surface of the anterior leaf of the broad ligament and was removed together with the right salpinx [58].

Laparoscopic management of a broad ligament pregnancy is not impossible, and it has many advantages as shown by the laparoscopic management of abdominal pregnancies in other unusual sites. These advantages include the faster recovery of the patient, the lower surgical morbidity, and the better control of the blood loss [38]. It can be considered when the size is small [23, 59] and the patient stable. The presence of proficient laparoscopists and the option to convert to laparotomy at any time are also crucial [38]. Olsen in 1997 reported the first case of a successful laparoscopic management of a broad ligament pregnancy [23], while in 2004 Siow

et al. reported a successful laparoscopic management of a pregnancy presenting with a 6.5 cm mass. Siow et al. injected vasopressin into the broad ligament before starting the excision, the pregnancy was removed, and the surgery was completed uncomplicated [38]. Since then there has been an increase in the reports of broad ligament pregnancies which were managed with laparoscopy [11, 17, 24, 25, 30, 60–62], while Mo et al. in 2018, in the case they reported, managed laparoscopically a broad ligament pregnancy with a hemoperitoneum of about 2800 ml. In most cases coagulation was used to control the bleeding. Cheung et al. in 2014 reported that they needed to use three 1-0 Biosyn stitches in order to close the implantation site inside the broad ligament, so as to control the bleeding [17], while Kar in 2012 used superficial infiltration of Pitressin (vasopressin) so as to control the hemorrhage from the base [62]. Finally in a case report by Yang et al. in 2017, they used absorbable hemostatic cellulose which was inserted into the retroperitoneal space in order to prevent bleeding [63].

Despite the extensive use of laparoscopy and the willingness to treat an ectopic pregnancy with more conservative ways, explorative laparotomy is still the way most cases are treated. The presentation to the hospital—most common with abdominal pain—of a missed broad ligament pregnancy plays an important role to this. Laparotomies have been reported in order to treat a wide spectrum of broad ligament pregnancies, starting with 5 weeks of amenorrhea [60], till broad ligament pregnancies at term [15, 36] which were admitted for an emergency cesarian section. Finally a case of a post-term broad ligament pregnancy has been reported [41].

In the published medical literature, different outcomes for women who underwent laparotomy or laparoscopy for a broad ligament pregnancy can also be found. There have been reported cases where the size of the pregnancy made it possible to be removed without any damage to the surrounding organs [10–12, 14, 15, 17, 19–26, 33, 36–38, 41, 60–62, 64–66], where in other cases salpingectomy [18, 27, 31, 32, 40, 56, 58, 67], oophorectomy [30], or salpingo-oophorectomy [28, 34, 35, 39] was necessary to be performed due to the damage these organs presented. There have also been reported cases where the need for hysterectomy presented [20, 29, 40, 68], and finally in a report by Wolfe and Neigus in 1953, among three cases reported, the two patients died [20]. The above show that a broad ligament pregnancy can be a life-threatening situation or a cause of a serious disability, especially when excision of ovaries or the uterus is a necessity in order to save the patient. It also makes it necessary to inform the patient before the operation of the possible outcomes, especially when the suspicion of a broad ligament pregnancy is present.

Finally, the fact that a missed broad ligament pregnancy can present from an early week of amenorrhea or it can reach term and post-term, with a great range of symptoms (from none to very acute) makes the broad ligament pregnancy part of the differential diagnosis of nearly every pregnant woman presenting to the emergency room, no matter in which week of pregnancy she is. In the case that a broad ligament pregnancy is considered to be a possibility, it is important for the managing surgeon to gather the best available team. Because of the complexity of these cases, this team cannot be limited to gynecologists but should also include—if possible—general surgeons, urologists, and interventional radiologists. Each one of them can play an important role to the management of a broad ligament pregnancy depending on the size of the pregnancy and the damage to the surrounding organs.

7. Conclusions

An ectopic pregnancy located between the leaves of the broad ligament is a rare but possibly life-threatening presentation of an ectopic pregnancy. The fact that it

can be easily missed is it can cause worrying symptoms in any week of pregnancy, or it can go uneventful till term makes it a possibility in differential diagnosis of almost every pregnant woman presenting to the emergencies. The diagnosis preoperatively is also difficult to be established. MRI can play an important role if there is ultrasound suspicion for a broad ligament pregnancy. It must also be mentioned that, although there is growing need to manage ectopic pregnancies with minimal invasive techniques, methotrexate cannot be considered a first-line treatment for this type of ectopic pregnancy. On the other hand, there are numerous reports, and becoming more often every year, of broad ligament pregnancies managed laparoscopically. Finally, every gynecologist or surgeon who must operate a woman with a suspicion of a broad ligament pregnancy must provide the patient the best information for possible excision of organs crucial to reproduction.

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