

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Intramural pregnancy at 6 and 8 weeks gestation with no predisposing factors: As two uncommon case reports [☆]

Ayoub Aouragh^{a,b}, Marouane Boukroute^{a,b,*}, Youssef El Aloua^{a,b}, Abdelmajide Reagraui^{a,b}, Ibtissam Bellajdel^{a,b}, Hafsa Taheri^{a,b}, Hanane Saadi^{a,b}, Ahmed Mimouni^{a,b}

^a Department of Obstetrics and Gynecology, Mohammed VI University Hospital Center, Oujda, Morocco.

^b Faculty of Medicine and Pharmacy, Mohammed First University, Oujda, Morocco

ARTICLE INFO

Article history:

Received 12 February 2024

Revised 23 February 2024

Accepted 26 February 2024

Keywords:

Ectopic pregnancy

Intramural pregnancy

Pelvic ultrasound

Pelvic MRI

Laparotomy

Metrorrhagia

ABSTRACT

Ectopic pregnancies are extremely rare causing severe consequences such as uterine rupture and may lead to death. The clinical symptomatology is very atypical, essentially pelvic pain and metrorrhagia, for this reason, a misdiagnosis can be fatal. Here we report two cases of an intramural pregnancy in a 33-year-old primigravida at 6 weeks gestation after a medically assisted reproduction and a 26-year-old primigravida at 8 weeks gestation with no predisposing factors, making the diagnosis more challenging.

© 2024 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Introduction

Ectopic pregnancy corresponds to the implantation and development of the egg outside the uterine cavity. They are extremely rare and represent less than 1% of all ectopic pregnancies. Pregnancies have been reported to implant in the cervix, ovary, interstitial tubal segment, and at various intra-

abdominal sites [1]. Only 21 published cases of intramural ectopic pregnancies were published between 2000 and 2012 [2].

The diagnosis of intramural pregnancies through ultrasound may be challenging, leading to potentially severe consequences such as uterine wall rupture and catastrophic hemorrhage [3]. In the absence of early intervention, hemorrhagic shock, further can lead to death. Maternal mortality rate is about 2.5% when uterine rupture develops [4].

[☆] Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

* Corresponding author.

E-mail address: marouaneboukroute4@gmail.com (M. Boukroute).

<https://doi.org/10.1016/j.radcr.2024.02.092>

1930-0433/© 2024 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Case presentation

Case 1

A 26-years-old woman was referred to the Early Pregnancy Assessment Unit during her first pregnancy. The patient presented with lower abdominal pain at 8 weeks gestation based on her last menstrual period, with no reported vaginal bleeding and a high level of serum beta-human chorionic gonadotropin (bhCG) measured at 4800 IU/L. A repeat bhCG level 48 hours later showed an elevation to 10,519 IU/L. Therefore, she was admitted for observation, and a suprapubic ultrasound was performed. In our case, supra-pubic ultrasound could not visualize the exact location of the gestational sac, which prompted us to complete the scan with an endo-vaginal ultrasound. This showed a mass measuring 31 x 28 x 29 mm seen on the right side closely located to the cornua surrounded by myometrium without any communication with the endometrium. It appeared to contain a 25 mm gestational sac containing an embryo with positive heart activity, with no effusion in the Douglas cul de sac (Fig. 1). On that account, the intramural ectopic pregnancy diagnosis was confirmed. Given the uncommon site of the pregnancy, a laparotomy was conducted.

During laparotomy, the visceral peritoneum lining the uterine wall surrounding the intramural pregnancy appeared violated. An incision was made, and a 3 x 3 x 26 mm gestational sac appeared without any visualized breaches, the sac was removed followed by aspiration of the preoperative residues of the sac (Fig. 2). Both Fallopian tubes appeared to be normal along their full span. Thus, the ectopic pregnancy was extracted and postoperative follow-up was straightforward.

Postoperative follow-up consisted in performing quantitative bhCG kinetics within 2 weeks, at 2-day intervals. BhCG values decreased from 32 UI/L To 4 UI/L, and ultrasound images showed an empty endometrial cavity surrounded by the myometrium, with no visible gestational sac.

Case 2

At the time of the consultations of medically assisted procreation a patient of 33 years, with a regular cycle made of 5 days / 28, presented for a primary infertility of 2 years. Clinical examination showed no signs of hyper-androgenism or genital malformation. Biological work-up revealed normal AMH (Anti mullerian hormone) at 2.1 ng/mL, FSH (Follicle stimulating hormone) at 3.4 mUI/ml, the rest of the fertility work-up was normal.

Pelvic ultrasound showed a retroflexed retroverted uterus, with a 39 mm endometriotic left ovarian cyst. Hysterosalpingography was performed showing a bicornuate uterus with conserved tubal permeability. MRI revealed a uterus with an arched fundus, with a 40 mm endometriotic ovarian cyst. The patient underwent ovarian stimulation with 2 cycles of clomiphene citrate and 2 cycles of recombinant FSH, with a successful second attempt of intrauterine insemination.

During pregnancy follow-up, a pelvic ultrasound was performed at 6 weeks, revealing a 17-mm intramural cornual gestational sac containing an embryo with positive heart activity and no visible communication with the endometrium, followed by pelvic MRI, which confirmed the intramural cornual pregnancy.

An exploratory laparoscopy was suggested, yet the patient declined. As a result, the patient was placed under observation, and an obstetric ultrasound was conducted at 9 weeks, revealing a 31mm intramural cornual gestational sac containing an embryo with a positive heart activity complicated by placental abruption and an empty endometrium (Fig. 3).

A subsequent MRI was carried out showing an intramural mass measuring 29 x 27 x 27 mm, located to the cornua surrounded by myometrium without any relation with the endometrium 5mm from the anterior wall of the uterus (Fig. 4).

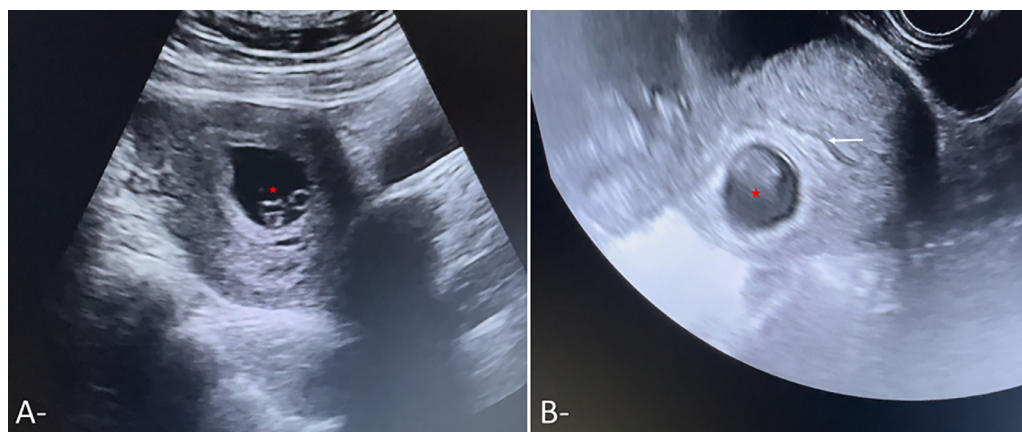


Fig. 1 – (A) suprapubic ultrasound showing the gestational sac with a positive heart activity (Star), (B) endo-vaginal ultrasound showing a right-sided gestational sac (Star) located in the myometrium with no visible relation with the endometrium (arrow).

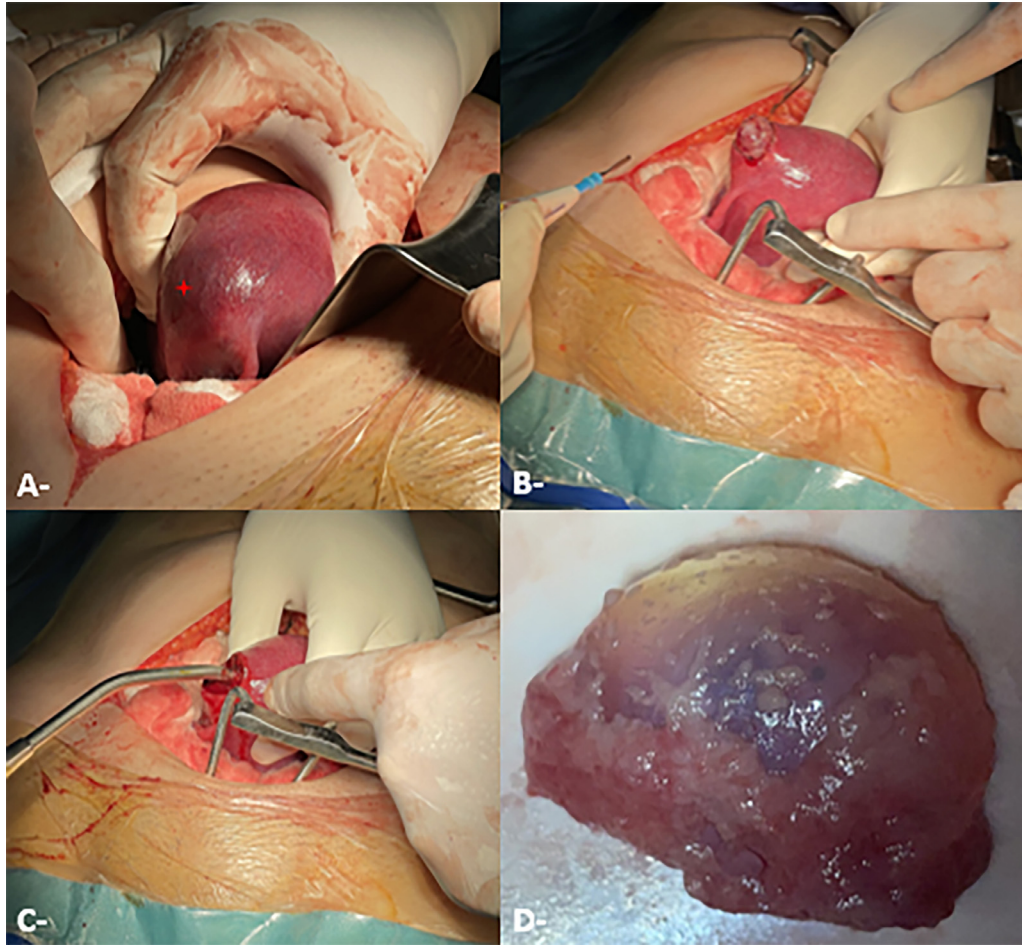


Fig. 2 – Laparotomy per-operative images showing (A) a violated peritoneum surrounding the gestational sac (Star) (B) incision of the uterine wall and excision of the gestational sac (C) aspiration of peroperative residues (D) the gestational sac with no visible breaches.

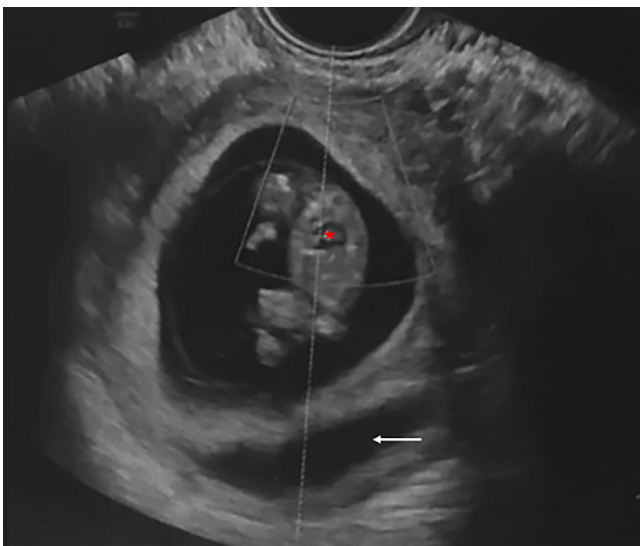


Fig. 3 – Ultrasound image showing the gestational sac (Star) containing an embolus with positive cardiac activity complicated by a placental abruption (arrow).

Discussion

Ectopic pregnancy involves the fertilization of an ovum and its development outside the uterine cavity. In some cases, growth can occur intramurally, resulting in a pregnancy with a high risk of complications. Early diagnosis is therefore crucial. Predisposing factors for intramural pregnancy include a prior uterine trauma (Cesarean section, myomectomy, dilatation, and curettage), adenomyosis, and in vitro fertilization [5]. In contrast to the first report, we could not identify any predisposing factor related to the obstetrical history of our patient.

The diagnosis of intramural pregnancy is very challenging since the clinical presentation can range from being asymptomatic, amenorrhea, pelvic pain and, very rarely, vaginal spotting, to uterine perforation and massive hemorrhage. Therefore, diagnosis can be supported by high BHCG levels. Nonetheless, an ectopic pregnancy may manifest despite the absence of detectable maternal serum Beta-hCG [6].

Therefore, the polymorphic clinical symptomatology necessitates the use of imaging modalities, including ultrasound and magnetic resonance imaging (MRI) [7,8]. Ultrasound diagnosis relies on the observation of a gestational sac entirely en-

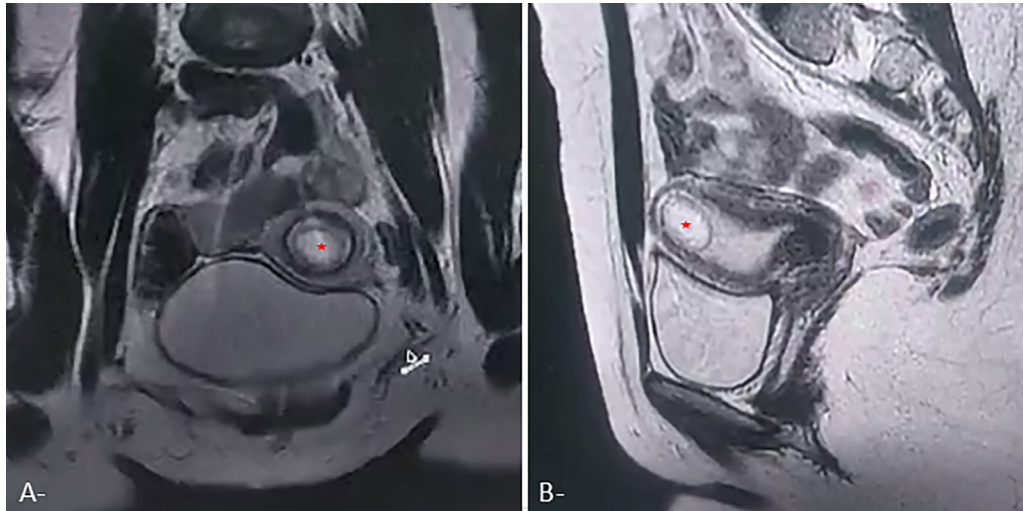


Fig. 4 – T2 MRI images in frontal (A) and sagittal (B) sections showing a progressive ectopic intramural cornual pregnancy (29mm x 27 mm x 27mm) (star) at 09 weeks completely surrounded by the myometrium, undamaged myometrium junction between the gestational sac and the uterin cavity.

veloped by myometrium, devoid of any connection with the endometrium [9]. However, visualizing the embryo remains the gold standard for diagnosis. In our case, endo-vaginal ultrasound has been shown to be more effective than suprapubic ultrasound in visualizing intra-mural pregnancy.

According to Luo and al, there are three ultrasound forms of intramural pregnancy: cystic, nodular, and ruptured [10]. As reported by the imaging, ultra-sonography allowed us to specify the exact location of the gestational sac and to determine it as an embryonic cystic form.

Diagnosis of intramural pregnancy can be complex, and may not be identified early by conventional imaging studies, such as ultrasound. In such cases, the implementation of an additional MRI may be imperative. This not only aids in diagnosis but also allows for the precise localization of the embryo, evaluation of perimetric vascularity in the implantation zone, acquisition of information regarding the blood supply linked to intramural pregnancy, and detection of potential complications such as uterine rupture or other pathological manifestations.

Intramural pregnancy under conservative management can involve either medical or surgical approaches. Methotrexate is recognized as the most frequently employed drug, administered either locally or systemically, especially when the ectopic pregnancy is early detected. According to literature, a successful treatment of a posterior wall intramural pregnancy using a single dose of intramuscular methotrexate (50 mg/m²) was described [11].

Conforming to literature, the major surgical option remains local excision of the intramural pregnancy via laparoscopy [7,12].

After successful treatment, medical follow-up is often recommended to ensure complete elimination of the ectopic pregnancy and to assess the patient's future reproductive health. According to Steier et al. [13], following surgical intervention, ectopic pregnancies exhibited the briefest elimina-

tion duration. Elevated initial concentrations correlated with an accelerated decrease in human chorionic gonadotropin levels during subsequent monitoring, surpassed only by the rate observed in cases of spontaneous abortions.

Conclusion

Early diagnosis is crucial to avoid serious complications, often requiring medical or surgical intervention.

The combination of endo-vaginal ultrasonography and serum quantitative BHCG assay can lead to early diagnosis and a significant reduction in the mortality rate due to uterine rupture and hemorrhagic shock in extra uterine pregnancies and preserving their fertility.

Patient consent

The patients gave their informed consent for this case report to be published.

REFERENCES

- [1] Molinaro T, Barnhart K. Ectopic Pregnancies in Unusual Locations. *Semin Reprod Med* 2007;25(2):123–30. doi:10.1055/s-2007-970051.
- [2] Kirk E, McDonald K, Rees J, Govind A. Intramural ectopic pregnancy: a case and review of the literature. *Eur J Obstet Gynecol Reprod Biol* 2013;168(2):129–33. doi:10.1016/j.ejogrb.2012.12.036.
- [3] B. Sanaa, G. Abdel-Naser, A. Lamyia M., R. Hassanein, Ectopic pregnancy in uncommon implantation sites.

- [4] Neiger R, Weldon K, Means N. Intramural pregnancy in a cesarean section scar. A case report. *J Reprod Med* 1998;43(11):999–1001.
- [5] Ong C, Su L-L, Chia D, Choolani M, Biswas A. Sonographic diagnosis and successful medical management of an intramural ectopic pregnancy. *J Clin Ultrasound* 2010;38(6):320–4. doi:10.1002/jcu.20703.
- [6] Hsieh YY, Chang CC, Tsai HD, Yeh LS, Hsu TY, Yang TC. Intramural pregnancy with negative maternal serum beta-hCG. A case report. *J Reprod Med* 1998;43(5):468–70.
- [7] Lee GSR, Hur SY, Kown I, Shin JC, Kim SP, Kim SJ. Diagnosis of early intramural ectopic pregnancy. *J Clin Ultrasound* 2005;33(4):190–2. doi:10.1002/jcu.20107.
- [8] Ko H-S, et al. Sonographic and MR findings in 2 cases of intramural pregnancy treated conservatively. *J Clin Ultrasound* 2006;34(7):356–60. doi:10.1002/jcu.20245.
- [9] Eray ÇALIFKAN, Yiçit ÇAKIRO/LU, Ayd>n ÇORAKÇI, Expectant Management of an Intramural Ectopic Pregnancy in a Primigravid Woman.
- [10] Luo Z, Zhou P, Gao F, He W, Luo S. [Diagnosis of intramural pregnancy by endoluminal color Doppler ultrasonography and review of the literature]. *Nan Fang Yi Ke Da Xue Xue Bao* 2010;30(10):2343–6.
- [11] Bouzari Z, Keshani M, Yazdani S, Barat S, Zinalzadeh M. Intramural pregnancy. *J. Obstet. Gynaecol. J. Inst. Obstet. Gynaecol.* 2010;30(2):195–6. doi:10.3109/01443610903443921.
- [12] Jin H, Zhou J, Yu Y, Dong M. Intramural pregnancy: a report of 2 cases. *J Reprod Med* 2004;49(7):569–72.
- [13] Steier JA, Bergsjo P, Myking OL. Human chorionic gonadotropin in maternal plasma after induced abortion, spontaneous abortion, and removed ectopic pregnancy. *Obstet Gynecol* 1984;64:391–4.