

# Early diagnosed intramural ectopic pregnancy associated with adenomyosis: Report of an unusual case

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## Summary

Intramural pregnancy is the rarest type of ectopic pregnancy and almost always is diagnosed intraoperatively. It constitutes less than 1% of ectopic pregnancies and the world literature contains only 33 cases. We present an additional case of an uncomplicated intramural pregnancy in a patient with extensive adenomyosis, which is the earliest preoperatively diagnosed (sixth week of gestation) case in the literature.

**Key words:** Intramural ectopic pregnancy; Adenomyosis.

## Case Report

A 41-year-old, gravida 6, para 4, with two prior artificial abortion (2 and 15 days delayed menstruation), was admitted to a local private gynecology clinic due to pelvic-abdominal pain localized on the left side. A six-week gestational sac without fetus measuring 34 mm in diameter and an enlarged uterus measuring 13 x 8 x 5 cm in size was observed by ultrasound (Figure 1). The patient's blood pressure and pulse were normal. There was no abdominal rebound or guarding. Her serum  $\beta$ -hCG (beta-human chorionic gonadotropin) level was 1400 mIU/ml.

An explorative laparotomy demonstrated that there was no gross pathology in the bilateral adnexa and pelvic wall except slight bulging of the cornual uterus. She underwent left cornual resection of the uterus. The serum  $\beta$ -hCG level dropped to 35 mIU/ml on the fifth day and to less than 5 mIU/ml on the 15<sup>th</sup> postoperative day. There were no complications postoperatively.

The patient was discharged without complaints on the third postoperative day.

## Pathologic Findings

Cornuectomy material measured 6 x 5 x 5 cm. On section, there was a cystic space 2.5 x 2.5 x 2 cm in diameter which contained clear fluid and the inner side of the cyst wall was lined with a cloudy thin membrane which covered a spongy and papillary natured tissue (Figure 2). Microscopically, myometrium demonstrated foci of adenomyosis with decidualization of the stroma (Figure 3). There were chorionic villi involving the myometrium (Figure 4). No fetal tissue was disclosed.

## Discussion

Implantation into the muscle wall is the rarest form of ectopic pregnancy. It should be confirmed pathologically

by showing the myometrium surrounding the gestational product which is completely separated from the endometrial cavity and fallopian tubes. The presenting myometrium all around the gestational sac in this case is probably the result of early diagnosis (6<sup>th</sup> week of gestation). As in some of the reported cases the absence of myometrium in some areas might be noted [1].

The etiology of intramural pregnancy is not clear. Many theories have been proposed to account for this rare lesion. These include: serosal implantation of the conceptus following transmigration [2], implantation in a tract from uterine perforation due to prior dilatation and curettage [3], increased trophoblastic activity and defective decidualization which permit penetration into the myometrium of conceptus [1], traumatic hysteroscopy following in vitro fertilization and embryo transfer (IVF-ET) [4]. In their article on intramural pregnancy, Fait *et al.* claimed that implantation in a focus of adenomyosis may result in intramural pregnancy [5].

To date 32 cases have been documented and two cases were diagnosed preoperatively [6], and six cases were associated with adenomyosis [7]. Fetal loss occurred between ten [4] and 22 weeks' gestation except for one case with a surviving 30 week old fetus delivered by caesarian hysterectomy [5]. Our case is the earliest diagnosed (in the sixth week of gestation) intramural pregnancy and also the third case preoperatively known. Ectopic endometrium of adenomyosis can respond to estrogen and progesterone and show decidualization [8]. Blastocyst implantation may occur in this decidualized endometrium. Although one of the etiologic factors of intramural pregnancy has been suggested to be implantation in a tract caused by uterine trauma, we did not observe any sinus tract leading from the endometrial cavity to the implantation site. The differential diagnosis includes interstitial pregnancy or cornual pregnancy, uterine sacculation, pregnancy in the uterine diverticulum, pregnancy in the rudimentary horn or septate uterus.

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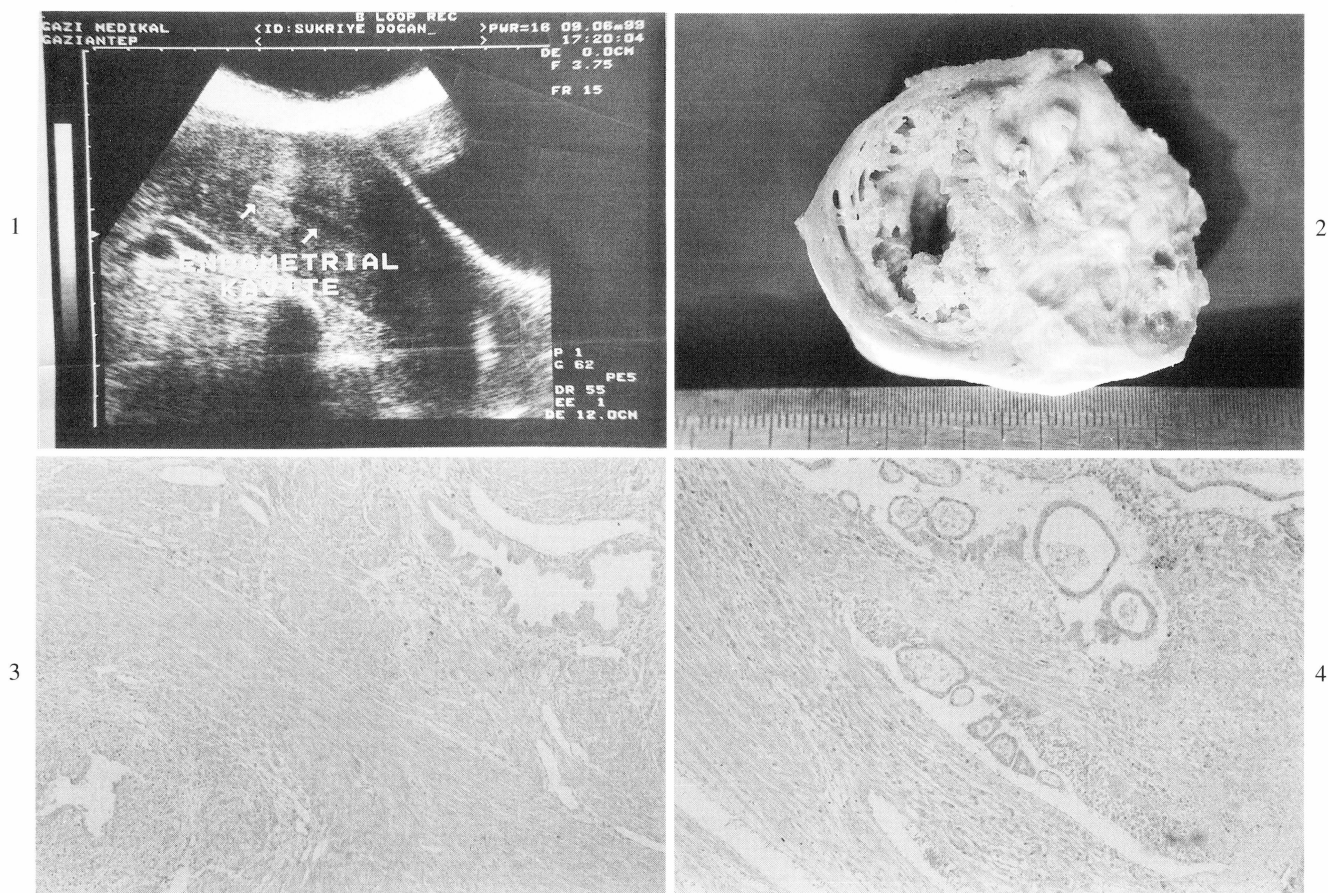


Figure 1. — Ultrasound image showing a six-week gestational sac located in the left cornual region without a fetus (arrows). Endometrial cavity.

Figure 2. — Gross view of cornuectomy specimen revealing the cavity formed by intramural implantation. The cavity is filled with spongy, papillary tissue.

Figure 3. — Microscopic view of myometrium demonstrating foci of adenomyosis with decidualization of stroma (upper right and lower left) HEx100.

Figure 4. — Microscopic finding showing chorionic villi involving the myometrium. (HEx100).

In some cases transvaginal ultrasonography or color doppler ultrasound may contribute to the diagnosis but the pathologist examining the specimen should make the definite diagnosis [5].

In conclusion, the clinician should consider an intramural ectopic pregnancy in a woman with delayed menstruation, an elevated serum  $\beta$ -hCG level, an intramural cyst, and no other possible sites of ectopic pregnancy exposed prior to uterine trauma. We believe that early diagnosis of ectopic intramural pregnancy will lead to adequate timing to prevent complications such as acute abdomen thus preserving the patient's reproductive capability.

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