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True ectopia of left ovary with left renal agenesis: a case report Sol renal agenezi ile birlikte sol overin gerçek ektopisi: olgu sunumu

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Abstract

True ectopia of ovarian tissue is a clinical entity which is rarely encountered by the gynecologists. The present report describes a case of true ectopic ovary accompanied with ipsilateral renal agenesis with a review of related literature. A 23-year-old woman was admitted to the department of gynecology due to two-year-long primary infertility. Although transvaginal ultrasonography revealed a normal sized and shaped uterus and right ovary, the sonographer was unable to observe the left ovary. Laparoscopy confirmed the existence of a normal uterus and right ovary. However left ovary was found to be situated within the pelvis where it was left in situ. The magnetic resonance imaging performed during the postoperative period demonstrated ipsilateral renal agenesis and slight enlargement of the right kidney. To the best of our knowledge, this is the first case report describing the co-occurrence of true ectopia of left ovary and ipsilateral renal agenesis.

Key Words: Agenesis, ectopic tissue, kidney, ovary

Özet

Ovaryan dokuların gerçek ektopisi jinekologlar tarafından nadir karşılaşılan bir durumdur. Bu vaka gerçek ovaryan ektopiyle birlikte ipsilateral renal agenezi saptanan bir olguyu ve literature derlemesini içermektedir. 23 yaşında bayan hasta kliniğe 2 yıllık primer infertilite nedeniyle başvurmuştur. Transvajinal ultrasonda normal büyüklükte ve şekilde uterus ve sağ over izlenmesine rağmen sol over izlenmemiştir. Laparoskopide de sağ over ve uterus gözlemlenmiş fakat sol over pelviste olması gereken yerin solunda tespit edilmiştir. Postoperatif dönemde yapılan magnetic resonons görüntülemede sol böbreğin olmadığı sağ böbreğin de hafif genişlemiş olduğu izlenmiştir. Bildiğimiz kadarıyla vakamız aynı taraflı renal agenezi ile sunulan ilk gerçek ovaryan ektopi vakasıdır.

Anahtar kelimeler: Agenez, ektopik doku, böbrek, over

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Introduction

True ectopia of ovarian tissue is a clinical entity which is infrequently encountered by the gynecologists (1). After it was described by Wharton in 1959 for the first time, approximately 40 cases have been reported in the literature (1,2). Due to its rarity, there is much discussion about the nomenclature and classification of the ovarian tissues which are found to be located elsewhere than the ovarian fossa. The contradictions and obscurities related with the description of ectopic ovarian tissue have led to the documentation of different values for the incidence of this rare pathology (1,2,3). That is, it has been reported to vary between 1 in 29000 to 1 in 700000 gynecologic admissions (3).

The present report describes a case of true ectopic ovary accompanied with ipsilateral renal agenesis with a review of related literature.

Case

A 23-year-old woman was admitted to the department of gynecology at Siirt Women Health Hospital due to two-year-long primary infertility. It was learnt that she had regular menses since her menarche at 12 years of age. There was nothing particular in her medical history and the patient claimed that she did not undergo any surgical intervention. Physical examination was unremarkable.

The basal hormonal profile of the patient (at day 3 of menstrual cycle) was as follows; follicle stimulating hormone: 4,5 mIU/L (range:0-12 mIU/L), luteinizing hormone: 3,3 mIU/L (range:0-12 mIU/L), estradiol: 66,5 pg/ml (range:20-80 pg/ml), prolactin: 14,1 ng/ml (range:<35 ng/ml), thyroid stimulating hormone: 1.4 mIU/L (range:0,5-5,0 mIU/L), free T3: 0,3 ng/dl (range: 0.2-0.5 ng/dl), free T4: 1,1 ng/dl (range:0.8-1.8 ng/dl).

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Transvaginal ultrasonography revealed a uterus of 45x40x38 mm with a 4 mm thick endometrium. Although right ovary was detected to be normal, the

sonographer was unable to observe the left ovary. On the contrary, hysterosalpingography demonstrated a unicornuate uterus. However tubal patency was bilaterally absent (figure 1).

Figure 1. Hysterosalpingography demonstrated a unicornuate uterus and bilateral absence of tubal patency.

After the informed consent was obtained from the patient, laparoscopy was performed to assess the tubal patency and visualize the right ovary. Observation by laparoscopy confirmed the existence of a uterus, a right fallopian tube and a right ovary. However neither the left ovary nor the left fallopian tube was present and there were no attachments to the uterus on the left side. The uterus, right fallopian tube and right ovary had normal location, size and appearance. A through

exploration ended up with the detection of the left fallopian tube and left ovary within the pelvis where the abnormally located ovarian tissue was left in situ. Considering the close association of genital and urinary abnormalities, magnetic resonance imaging (MRI) was performed during the postoperative period. Thus MRI scans demonstrated a slightly enlarged right kidney and the absence of left kidney (figure 2).

Figure 2. Sagittal T2 scans of magnetic resonance imaging showed the ectopic localization of left ovary and ipsilateral renal agenesis





Discussion

The embryological development of the urinary and genital systems is associated closely. The intermediate mesoderm which extends along the dorsal wall of the embryo gives rise to the urogenital ridge. The primordium of urogenital ridge is made up by the nephrogenic ridge and gonadal ridge from which the urinary system and the genital system originate respectively.

The primitive genital system in two sexes is similar. The gonads are derived from three sources: mesothelium, mesenchyme and primordial germ cells. The paramesonephric ducts form the uterine tube and uterus in the female (4).

By 18 weeks, many primordial ovarian follicles have emerged within the ovaries which are being formed on the dorsal wall of the embryo. Two weeks later, the ovaries begin their descent under the guidance of both the gubernaculum and processus vaginalis. As the ovaries descend to a point inferior to the pelvic brim, the gubernaculum gives rise to the ovarian ligaments and round ligaments of the uterus (4,5).

Previously published case reports usually claim about the co-occurrence of ovarian ectopia and congenital abnormalities related with paramesonephric ducts including uterine agenesis, unicornuate uterus or the absence of fallopian tubes (6,7,8). Also associated with the ovarian ectopia is the existence of pelvic kidney (6). According to the classification system described by Wharton in 1959, ectopic ovaries can be defined as either accessory or supernumerary, as distinguished by their relationship to a normal ovary. By definition, the accessory or supernumerary ovaries require the presence of two normal ovaries in order to be called so. That is, accessory ovaries were defined as excess ovarian tissue adjacent and connected to a normal ovary while supernumerary ovaries were described as those ovaries situated away from normal ovaries (2). Apart from the accessory ovaries which are found in close neighborhood of normal ovaries, supernumerary ovaries may be located within the retroperitoneum or omentum (8,9).

However not all abnormally placed ovaries fit this classification as is the case with the ovarian tissues that are found to be abnormally located after the fulfillment of pelvic surgery. A quite well-known example of this condition is the ovarian implant/remnant syndrome (10).

A review by Lachman and Berman documented that almost 50% of the reported cases with ovarian ectopia since 1959 were in patients who underwent previous pelvic surgery. Therefore Lachman and Berman proposed a new classification of abnormally located ovarian tissues. They suggested that ectopic ovary should be the proper term and that this can be further sub-grouped as: (i) Post-surgical implant secondary to pelvic surgery (ii) Post-inflammatory implant occurring to infection or inflammation (iii) True/Ectopic ovarian tissue (11).

Much similar to the normally located ovaries, ectopic ovaries may present with menstrual irregularities, infertility or abdominal pain (4). Another similarity is that cysts and tumors may be detected in ectopic ovaries as well (3,4,12,13). However, there is a general consensus that an ectopic ovary should be treated no differently than a normally situated ovary unless there is an obvious pathology in the ectopic ovary (1,4).

To the best of our knowledge, this is the first case which reports about the co-existence of ovarian ectopia and renal agenesis. The laparoscopic and radiological findings suggest that an interruption has occurred during the fetal development of the genitourinary system on the left side of the present case. Subsequently the left nephrogenic ridge failed to develop and abolished giving rise to unilateral renal agenesis. Meanwhile the left gonadal ridge developed but its descent was improper resulting in ipsilateral ovarian ectopia.

In conclusion, congenital genitourinary abnormalities are associated closely and it would be prudent to expect a urinary abnormality in the presence of a genital anomaly. It would be more convenient to use the more general term "ectopic ovary" in order to define an abnormally located ovarian tissue and the necessary diagnostic work up should be carried out to diagnose any co-occurring urinary abnormalities.

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