Case Report

An Unusual Presentation of Ovarian Fibroma Originating from an Autoamputated Ovary

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Abstract

We describe an extremely rare case of an unusually presented ovarian fibroma adherent to the sigmoid colon originating from an autoamputated ovary. A 64-year-old woman was referred to our hospital with an abnormal shadow that was approximately 4 cm in diameter in the pelvic cavity detected on abdominal X-ray imaging. Computed tomography demonstrated an irregularly shaped tumor with calcification in the pelvic cavity. Laparoscopy revealed that the tumor was adherent to the surface of the sigmoid colon with a discontinuous shell and empty cavity. The left ovary was lacking, although the left salpinx and right adnexa were in their normal locations. The tumor was carefully resected with cutting of the serosa of the sigmoid colon. The serosal defect was repaired with sutures. Postoperative histopathological diagnosis was old fibroma with calcification. To the best of our knowledge, this is the first reported case of extragonadal ovarian tumor originating from an autoamputated ovarian fibroma.

Keywords: Autoamputation, extragonadal ovarian tumor, fibroma, teratoma

Introduction

Extragonadal ovarian tumors are rare. Most of the reported cases originate from autoamputated mature cystic teratomas (MCTs). The most common site is the greater omentum. Herein, we describe a rare case of an unusually presented ovarian tumor adherent to the sigmoid colon originating from an autoamputated ovarian fibroma and discuss the reported etiologies of extragonadal ovaries. This case report was exempt from the institutional review broad at our institute.

Case Report

A 64-year-old woman, gravida 3, para 3, was referred to our hospital with an abnormal, irregularly shaped shadow in the pelvic cavity on abdominal X-ray imaging at a nearby clinic. The lesion was approximately 4 cm in diameter and had calcifications [Figure 1a]. The patient did not complain of any symptoms such as pain, nausea, or constipation at that time. The patient did not have any history of surgery, acute or chronic lower abdominal pain, or pelvic inflammatory disease. She had hypertension and diabetes mellitus that were well controlled with medication. Magnetic resonance imaging and contrast-enhanced computed tomography demonstrated an irregular mass with calcification, measuring approximately 3 cm × 4 cm in diameter. The tumor was not cystic, and its margins were not continuous [Figure 1b]. The shape and location of the tumor in the pelvic cavity changed slightly between imaging studies. Although an ovarian tumor was strongly suspected, the origin of the tumor was not accurately diagnosed with these imaging studies; thus, laparoscopy was performed.

Article History:
Received 24 June 2018
Received in revised form 26 October 2018
Accepted 29 October 2018
Available online 23 January 2019

Access this article online

Quick Response Code:

Website: www.e-gmit.com

DOI: 10.4103/GMIT.GMIT_63_18

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How to cite this article: Yazawa H, Takiguchi K, Kato A, Imaizumi K. An unusual presentation of ovarian fibroma originating from an autoamputated ovary. Gynecol Minim Invasive Ther 2019;8:40-3.

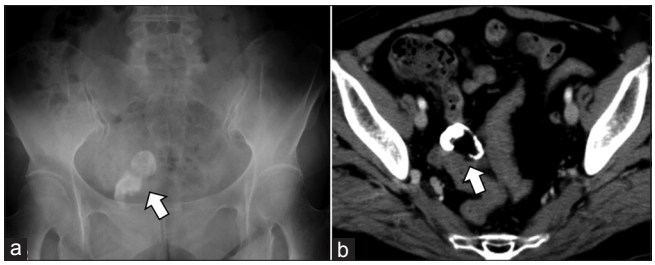

Figure 1: Findings of preoperative pelvic X-ray and computed tomography. Pelvic X-ray (a) and contrast-enhanced computed tomography (b) demonstrated an irregular mass with calcification in the pelvic cavity, measuring approximately 3 cm × 4 cm in diameter. The tumor was not cystic and its margins were not continuous

During laparoscopy, the tumor was located in the center of the pelvic cavity and adherent to the surface of the sigmoid colon with a discontinuous shell and empty cavity [Figures 2a-d and 3a]. The left ovary was lacking, but the left salpinx and right adnexa were normally shaped and positioned [Figure 2a]. There was a long narrow string connecting the left salpinx to the tumor [Figure 2b]. The tumor was carefully resected from the surface of the sigmoid colon. The serosa of the sigmoid colon was cut along the margin of the tumor [Figure 2e]. Next, the serosal defect of the sigmoid colon was repaired with sutures [Figure 2f]. Bilateral adenectomy was performed. The patient's postoperative clinical course was uneventful. Postoperative histopathological examination revealed that the tumor was old fibroma with calcification surrounded by degenerated, anuclear, spindle-shaped cells with a ghost-like appearance [Figure 3b and c].

The patient provided informed consent for this case report and associated images.

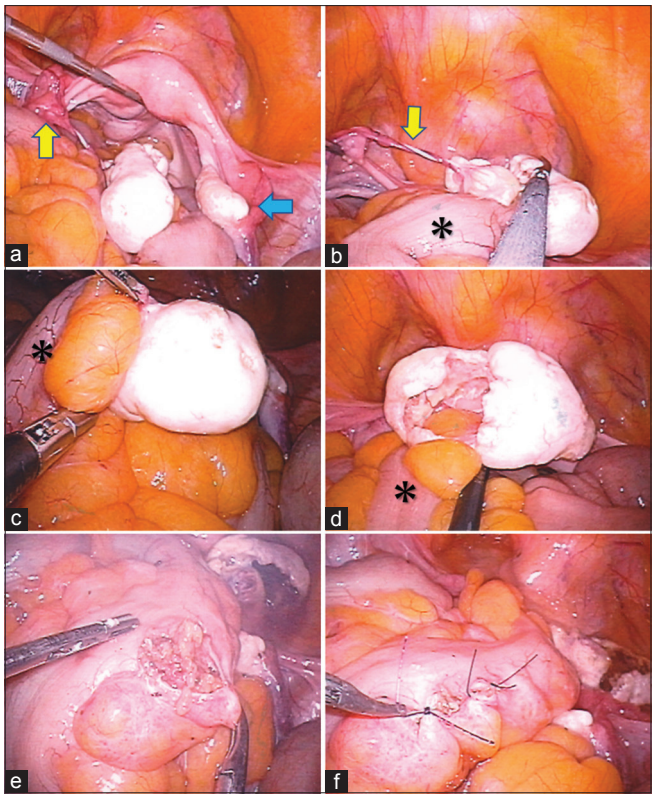

Figure 2: Findings of laparoscopic surgery. Laparoscopy revealed that the tumor was located in the center of the pelvic cavity (a). The left ovary was lacking, although the left salpinx was normal in shape and location (yellow arrow in a). The right adnexa was normal in position and shape (blue arrow in a). There was a long narrow string connecting the tumor and the left salpinx (yellow arrow in b). The sigmoid colon is indicated by * in (b and c). The tumor was adherent to the surface of the sigmoid colon (c), with a discontinuous capsular shell, calcification and an empty cavity (d). The tumor on the sigmoid colon was resected and the serosal defect in the sigmoid colon was repaired with sutures (e and f)

Discussion

Extragonadal ovaries have been rarely reported. Several terms have been used to describe this condition, such as ectopic ovary, accessory ovary, supernumerary ovary, parasitic ovarian tumor, and ovarian implant syndrome.[1] The etiology of an extragonadal ovary has been debated. Two types of mechanisms have been suggested: acquired and embryologic origin.[1] Most reported cases have suggested the former mechanism. Most reported cases consisted of autoamputated MCT implanted on the greater omentum or mesentery, although other implantation sites such as cul-de-sac or right subhepatic space have also been reported.[1-6] When Ushakov *et al*. and Lee *et al*. reviewed omental teratoma and autoamputation of an ovarian MCT, they concluded that autoamputation and reimplantation of an ovarian MCT on the greater omentum were considered the most common etiologies of extragonadal ovarian tumor.[2,6] Torsion of the pedicle is known to be the most frequent complication of ovarian tumors, which usually involve severe intrapelvic pain and require surgical intervention. If severe torsion was less symptomatic or asymptomatic, autoamputation might occur without surgical intervention. When torsion occurs acutely, the blood supply is impaired immediately, leading to necrosis and atrophy of the tumor due to ischemia.[3] When chronic torsion occurs, the tumor can become adherent to adjacent structures before necrosis and new collateral blood supplies may form, leading to a viable parasitic ovarian tumor, if it has completely detached from the original pedicle.[3] Although other sites such as the pouch of Douglas have been rarely reported as the location of a parasitic ovarian tumor, the greater omentum is reported to be the most plausible site for parasitic teratoma,[1,3,6-9] because of its special role in defense against intra-abdominal inflammation and adhesion formation. Due to easy mobility toward sites of inflammation and the rich surface capillary supply, the greater omentum may be the most favorable site for parasitic ovarian tumors.[6]

Ladenhauf *et al*. recently reported two cases of autoamputated ovary in newborns.[10] Pathological evaluation of these two

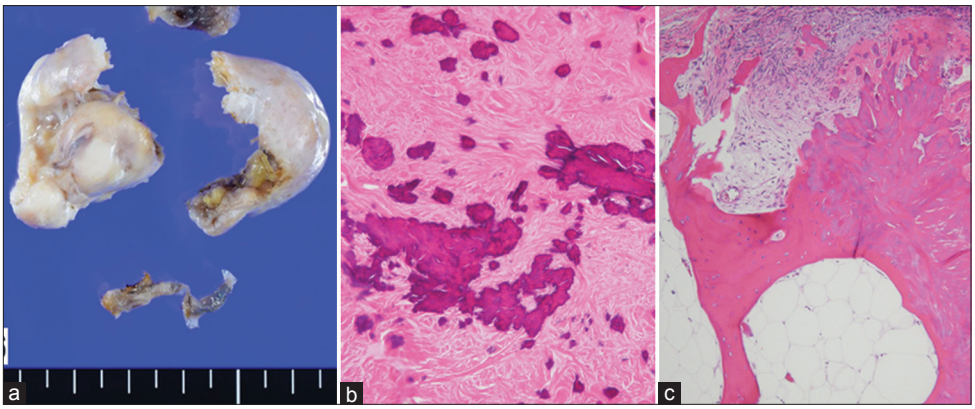

Figure 3: Histopathological findings. Resected tumor with a discontinuous shell and empty cavity (a). Postoperative histopathological examination revealed that the tumor was old fibroma with calcification surrounded by degenerated, anuclear, spindle-shaped cells with a ghost-like appearance (b and c)

cases revealed no signs of teratoma or malignancy. The most widely accepted etiology of this rare condition is chronic adnexal torsion. Possible risk factors for ovarian torsion in newborns include anatomic malformation, such as elongation of the tubo-ovarian ligament and increased weight of the ovary. Ovarian hyperstimulation during pregnancy may lead to increased size and weight of the ovary. Immature negative feedback mechanisms in the hypothalamic–pituitary–gonadal axis due to withdrawal of placenta steroids increase the levels of follicle-stimulating hormone and luteinizing hormone, which may lead to increased size and weight of the ovary and may predispose to ovarian torsion.[11]

On the other hand, evidence supporting embryonic origin as the mechanism includes a report of supernumerary ovary of the omentum diagnosed in neonates, suggesting aberrant migration of germ cells in the omentum during embryogenesis.[12] During early fetal development, migration of germ cells from the yolk sac along the hindgut (mesenteric route) toward the genital ridge (primitive gonad) takes place. These totipotent cells (primordial germ cells of early embryonic cells) may give rise to a variety of tissues originating from the three primitive embryonic layers. One theory on the cause of this type of extragonadal teratoma suggests that they arise from displaced primordial germ cells.[13] Primordial germ cells may stop differentiating during migration in embryonic life, thus causing a germ cell tumor occurring anywhere along the midline of the body.[14] In cases of congenital absence of the ovary or extragonadal ovary, congenital anomalies of the genitourinary tract are often observed.[6]

In our case, the findings of laparoscopic surgery suggest that the tumor was autoamputated from the left ovary, maybe due to asymptomatic torsion, and then became adherent to the serosa of the sigmoid colon. The tumor was pathologically diagnosed as an old ovarian fibroma. Normal ovarian structure was lost and replaced with sclerosis with severe calcification surrounded by degenerated, anuclear, spindle-shaped cells with a ghost-like appearance. Although ovarian fibromas are the most common benign solid tumors of the ovary, the incidence (1%–4%) is much fewer than that of MCT, the most common ovarian tumor (20%–25%).[3,15,16] As mentioned above, the most common pathological diagnosis for extragonadal ovarian tumor in previously reported cases was MCT, and we could not find any reports of extragonadal ovarian fibromas in a literature search using PubMed and Medline. To the best of our knowledge, this is the first reported case of extragonadal ovarian fibroma adherent to the colon that resulted from an autoamputated ovarian tumor.

Conclusion

We presented a very rare case of extragonadal ovarian fibroma thought to be due to autoamputation of a left ovarian tumor. Preoperative diagnosis of asymptomatic adnexal torsion, especially ovarian autoamputation, is very difficult and it was not possible in our case. Although it is rare, extragonadal ovarian tumor should be suspected when a curious tumor is found in the extragonadal portion of the abdominal cavity, even if the patient has no symptoms and no history of acute or chronic abdominal pain.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/he/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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