

CASE REPORT

An unusual occurrence of ovarian fibroma originating from an autoamputated ovary: a case report

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Abstract

Autoamputation of an ovary is a rare condition that is difficult to diagnose. The primary cause of this condition seems to be the chronic torsion of the adnexa. It is challenging to diagnose and is usually discovered during surgery. A 65-year-old woman came to the hospital complaining of abdominal fullness and multiple solid masses. A pelvic computed tomography (CT) scan showed a well-defined, solid mass measuring 11 cm × 8 cm in the cul-de-sac (CDS), which appeared to be a subserosal uterine leiomyoma. Multiple solid masses in the uterus were also observed, which were believed to be uterine leiomyomas. The patient underwent laparotomy, and it was discovered that the cause of the masses was an ovarian fibroma that had originated from an autoamputated ovary. The tumor was successfully removed with no complications, and the patient recovered well. Autoamputation of an ovary is challenging to diagnose, especially in postmenopausal women, due to its rarity and nonspecific symptoms. However, clinicians should consider this condition when a patient presents with early symptoms of abdominal discomfort or pain. This case highlights the importance of considering rare conditions in the differential diagnosis when a patient presents with unusual symptoms.

Keywords

Autoamputation; Chronic torsion; Fibroma; Leiomyoma; Ovary

1. Introduction

Autoamputation of the ovary is an infrequent occurrence with an uncertain cause, and as a result, the actual prevalence of this condition is not apparent due to its asymptomatic nature. According to a report, the incidence of ovarian autoamputation was found to be very low, less than 0.01% [1]. Ovarian amputation can be either congenital or acquired. While the exact mechanism remains unknown, chronic ovarian torsion is considered the most likely cause. On the other hand, even though it is uncommon, ovarian fibroma is the most common benign solid tumor of the ovary. It is typically misdiagnosed as uterine leiomyoma preoperatively due to its similar imaging features [2, 3]. Ovarian fibroma is usually found in perimenopausal and postmenopausal women, often coexisting with uterine leiomyoma, and is asymptomatic despite its significant size [4]. Due to its rarity, vague symptoms, and nonspecific imaging findings, accurate preoperative diagnosis of autoamputation of the ovary is challenging. This report presents a rare ovarian fibroma in the CDS originating from an autoamputated ovary.

2. Case presentation

A 65-year-old woman, gravida 3, para 2, presented with pelvic discomfort. She was referred to our gynecology unit after

a CT scan detected multiple solid masses in her pelvis that were suspected to be uterine leiomyomas. The patient reported mild abdominal discomfort and fullness but did not experience any acute abdominal pain, fever, nausea, vomiting, urinary symptoms, or constipation. She had not undergone any previous surgery but had Type II diabetes mellitus, which was being managed through medication. Laboratory tests, including fasting blood sugar, Hemoglobin A1c (HbA1c), and thyroid function tests, showed average results. Tumor marker analysis was not conducted. Multiple palpable masses in the pelvis and fullness in the cul-de-sac were detected during the pelvic examination. Transvaginal ultrasonography (US) and contrast-enhanced CT scans revealed numerous leiomyoma-like masses with calcifications. Additionally, an 11 cm × 8 cm well-defined mass was found in the CDS, which showed minor contrast enhancement and no internal calcification and abutted the left lateral aspect of the uterus (Fig. 1). It was observed that the left ovary was not visible due to postmenopausal status. A pelvic mass was suspected to be a subserosal uterine leiomyoma, considering multiple uterine leiomyomas were observed. Imaging showed no signs of torsion. Laparotomy was performed as the mass size was large, and mobility was decreased. A provisional diagnosis of multiple uterine leiomyomas was made. The left ovary was not visible during the operation, but the left salpinx and right adnexa were in their usual shape



FIGURE 1. Radiological findings. A contrast-enhanced CT image shows a well-defined hypodense mass (arrow) measuring 11 cm × 8 cm abutting the left lateral aspect of the uterus in the CDS. Several intramural leiomyomas with calcifications (arrowheads) were also noted.

and position. A distinct, non-infarcted, solid, whitish, and pedunculated mass was observed, along with a long, narrow, cord-like vascular structure connecting it to the left salpinx (Fig. 2). Compensatory hypertrophy of blood vessels was visible in the left mesosalpinx, but compensatory hypertrophy of contralateral adnexa was not observed. The tumor was removed easily, and no peri-tumor adhesion was noted. Later, total hysterectomy and bilateral adnexectomy were performed. The patient's postoperative clinical course was uneventful. The postoperative histopathological examination showed that the CDS tumor was a fibroma with bland spindle to ovoid cell proliferation (Fig. 3). These findings were interpreted as ovarian fibroma arising from an autoamputated left ovary.

3. Discussion

Autoamputation of the adnexa, fallopian tube, or ovary is a rare condition, and its exact incidence is not known, as several asymptomatic cases have been reported. Most cases of ovarian torsion are associated with symptoms such as acute abdomen, nausea, and vomiting, which may require emergency surgery. However, in some isolated cases, torsion may occur gradually and remain asymptomatic, leading to a delayed diagnosis until chronic torsion causes infarction and subsequent amputation [5]. Our patient only experienced mild pelvic discomfort and fullness, which are not typical symptoms of acute torsion.

While ovarian autoamputation is more commonly seen in

the perinatal period, it is rarely reported and often under-diagnosed in adults because most patients do not experience any symptoms. It is usually diagnosed incidentally during surgery for other reasons or during laparoscopy when a definite radiological diagnosis of a pelvic mass is absent [6]. Correctly diagnosing autoamputation before surgery is exceptionally challenging.

The patient in this study was initially thought to have a subserosal uterine leiomyoma based on US and CT scans (Fig. 1). However, during surgery, it was discovered that the patient had an ovarian fibroma originating from an autoamputated ovary (Fig. 2). Although US and CT are commonly used to diagnose this condition, they often fail to predict the origin of tumors accurately [2, 3]. For example, ovarian fibroma is frequently misdiagnosed as uterine leiomyoma in 34% of cases due to similar symptoms and ultrasound features [3]. Larger ovarian fibromas are even more challenging to diagnose as they closely resemble uterine leiomyoma [2].

Thus, clinicians should consider the possibility of an ovarian fibroma when multiple leiomyomas are detected radiologically and CT densities differ. In this case, magnetic resonance imaging (MRI) was not used in the preoperative work-up. However, MRI is recommended to characterize complex pelvic masses and may provide a diagnosis of adnexal autoamputation when used in combination with ultrasonography. Nevertheless, CT is often used due to its low cost and availability, and adnexal autoamputation may be incidentally found. In a case study,

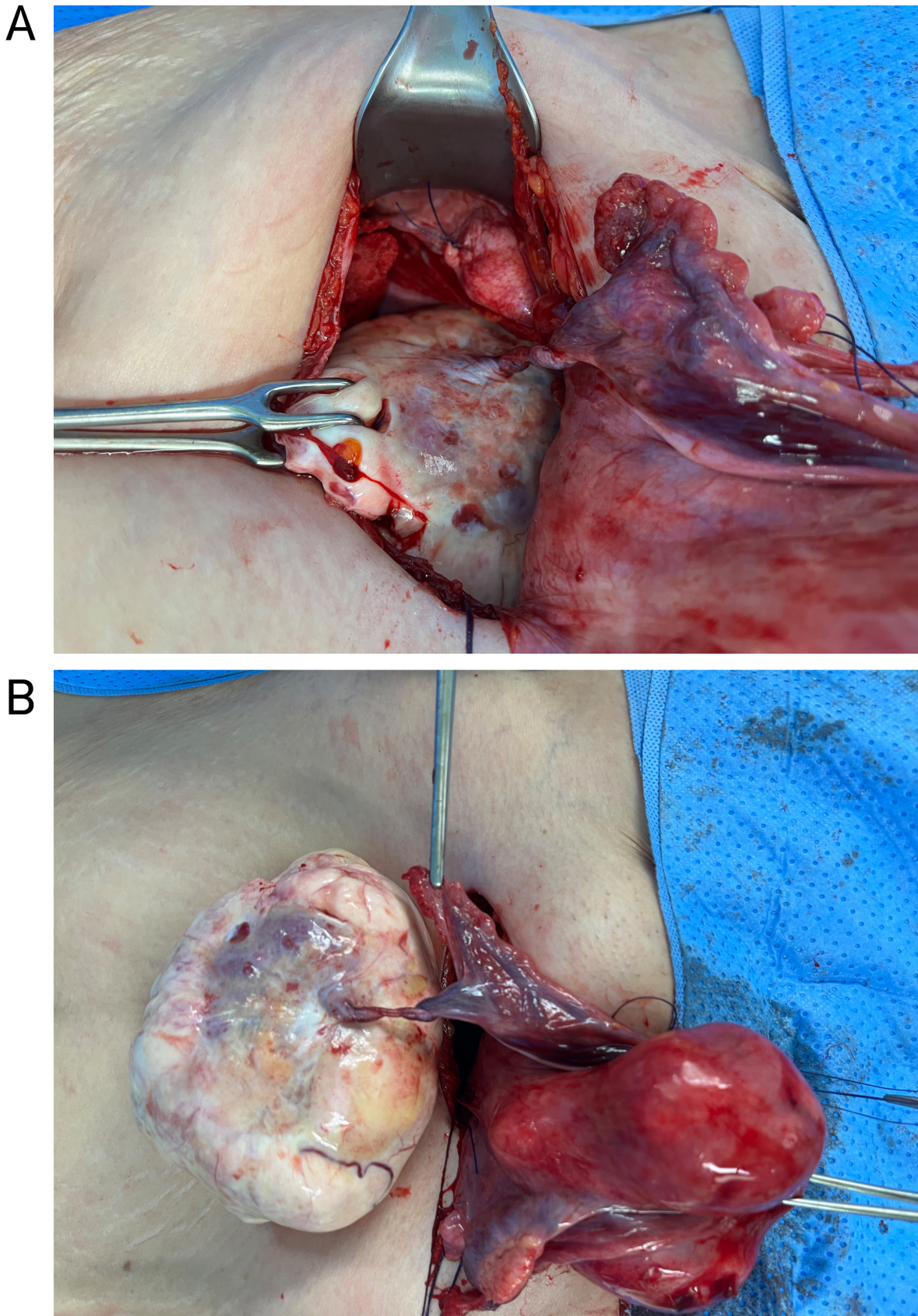


FIGURE 2. Intraoperative appearance of ovarian fibroma from the autoamputated ovary. Photograph showing the absence of the left ovary and a whitish, non-infarcted, solid tumor in the CDS (A) and outside the pelvic cavity (B). A long, narrow, cord-like vascular structure was observed connecting the left salpinx to the mass. Torsion of the pedicle in a spiral manner was grossly observed. Compensatory hypertrophy of blood vessels was observed in the left mesosalpinx.

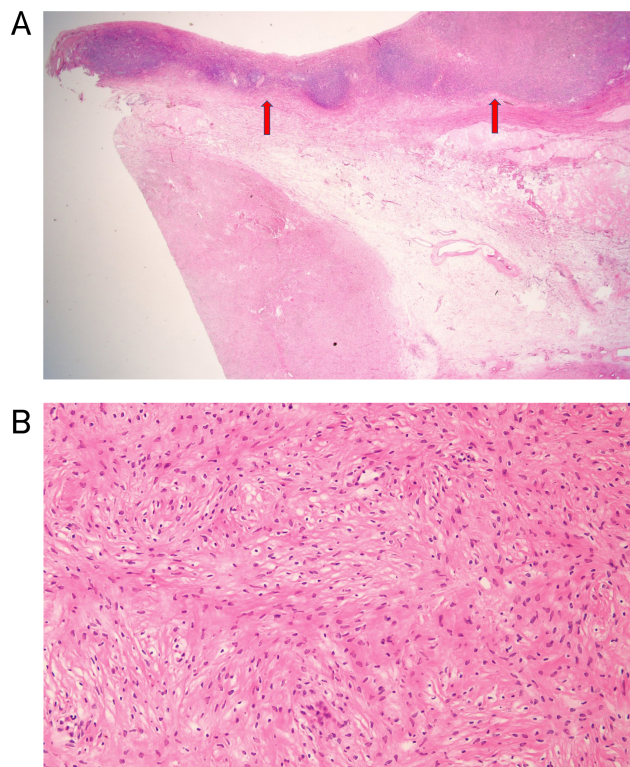


FIGURE 3. Microscopic findings. (A) Low power field image showing ovarian tissue (arrowed) and tumor tissue (H&E, $\times 10$). (B) High power field image showing bland ovoid to spindle tumor cells with collagenous stroma (H&E, $\times 200$).

Borukh *et al.* [2] reported that a preoperative MRI revealed an 11 cm mass resembling a torsioned pedunculated leiomyoma in a postmenopausal woman with pelvic and abdominal pain radiation to her lower back and abnormal vaginal bleeding. However, surgical intervention revealed a torsioned right ovarian fibroma.

It can be difficult to differentiate between ovarian fibroma and uterine leiomyoma, as some symptoms are similar. However, some clues can help. A vascular pattern can indicate ovarian fibroma on color flow Doppler, CT and MR images. Meanwhile, uterine leiomyoma is usually connected to the uterus and exhibits bridging vascular signs on CT. However, the final diagnosis is determined during surgery and confirmed through histologic examination. Although MRI can aid the differentiation, it can still be a challenge to distinguish between the two conditions, especially in cases where fibromas are cellular or atypical and resemble subserosal or pedunculated

leiomyomas [7].

Although the exact cause of ovarian autoamputation is not yet clear, it is believed to be a result of chronic ovarian torsion. Adnexal torsion, with or without an underlying ovarian lesion, can disrupt blood flow, leading to infarction, necrosis, and, ultimately, adnexal amputation. Additionally, the remaining autoamputated ovary may become resorbed entirely. After devascularization and autoamputation due to chronic adnexal torsion, dystrophic calcification occurs, and the autoamputated ovary floats freely in the abdominal cavity [8]. Most histopathological specimens reveal calcified necrotic tissues [9], although necrosis is observed more frequently. However, as Matsushita *et al.* [10] cautioned, physicians should be aware that an autoamputated ovarian cyst may be present even when the preoperative radiograph shows no calcification. In the present case, the histopathologic findings indicate the presence of viable ovarian tissue and no necrotic tissue or foci of calcification (Fig. 3).

Many cases of autoamputation are caused by dermoid cysts. These cysts make up around 25% of all ovarian neoplasms, and the twisting of their pedicle is the most common complication, occurring in 16.1% of cases. While ovarian fibromas are the most common benign solid ovarian tumors, they are less common than dermoid cysts, with an incidence of only 1–4%. Yazawa *et al.* [11] reported autoamputation in ovarian fibroma in a 64-year-old woman with no specific symptoms; our case is the second (Table 1). Other rare cases include the discovery of a free-floating ovary during gynecologic surgery in a postmenopausal woman [4] and the autoamputation of the salpinx after acute ovarian endometrioma torsion [12]. When an autoamputated ovary forms a mass, it may be attached or surrounded by the omentum, peritoneum, CDS or freely floating in the pelvic cavity [6].

This present study has some unique features. Firstly, while dermoid cysts have been the most common pathological diagnosis for tumors in an autoamputated ovary in previously reported cases, our case originated from an ovarian fibroma. This makes it the second case of ovarian fibroma in the CDS from an autoamputated ovary. Additionally, infarction was evident in most cases due to a compromised blood supply. However, no infarction was observed in our case because a cord-like vascular structure connected the left salpinx to the mass, an intact salpinx accompanied by isolated ovarian autoamputation. Secondly, the previously reported cases have shown that the right adnexa was more frequently involved. The proximity of the left ovary to the sigmoid colon may act as a barrier to torsion. Yazawa *et al.* [11] reported a

TABLE 1. Cases of autoamputated ovary with fibroma.

Author (yr)	Age (yr)	Size (cm)	Location	Side of ovary	Preoperative diagnosis	Necrosis	Calcification	Vascular connection	Normal ovarian tissue
Yazawa <i>et al.</i> [11]	64	4	sigmoid colon	left ovary	ovarian tumor	(–)	(+)	(+)	(–)
Our case	65	11	cul-de-sac	left ovary	subserosal myoma	(–)	(–)	(+)	(+)

case where the tumor originated from the left ovary, which is also the case for our patient. Furthermore, the tumor was initially believed to be a subserosal uterine leiomyoma based on US and CT findings. However, it was later confirmed to be intraoperatively an ovarian fibroma from an autoamputated ovary.

4. Conclusion

In summary, ovarian fibroma in postmenopausal women can cause chronic torsion, which may lead to adnexal autoamputation. Patients may show no significant symptoms, as in our case, and diagnosis before surgery is often impossible. Clinicians must consider this rare condition and take early abdominal discomfort/pain symptoms seriously. Although ovarian amputation is rare, it should be considered in the differential diagnosis when postmenopausal women show early symptoms of abdominal discomfort/pain.

ABBREVIATIONS

CT, computed tomography; CDS, cul-de-sac; MRI, magnetic resonance imaging; US, ultrasonography.

AVAILABILITY OF DATA AND MATERIALS

The data generated in this study are available upon request from the corresponding author. All authors had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis.

AUTHOR CONTRIBUTIONS

HJY—conception, manuscript writing and editing. HGK—manuscript writing and editing. NKL and KUC—analyzed the data. DSS—designed the research study. KHK—manuscript writing and editing. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The Institutional Review Board (IRB) of Pusan National University Hospital approved the report of this case (IRB #2401-019-134). Written consent was obtained for the publication of this case report. All accompanying images were anonymized.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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