Review

Autoamputation of the ovary after missed diagnosis of ovarian dermoid cyst torsion: a case report and review of literature

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Abstract

Torsion is the most frequent complication of ovarian dermoid cysts. Adnexal torsion typically presents as a severe abdominal pain and is treated as an acute surgical emergency. However, if surgery is delayed or the diagnosis is not made in a timely manner, autoamputation of the ovary is a very rare, but possible, complication. Herein, we report a case of an autoamputated ovary with a dermoid cyst and review the literature. A 33-year-old patient presented with pelvic pain lasting three weeks and was scheduled for a laparoscopy due to the presence of bilateral ovarian cysts, with a dermoid cyst identified on the left ovary. During the procedure, it was discovered that both the left fallopian tube and ovary were absent. The infundibulo-pelvic ligament appeared to terminate abruptly at the pelvic brim. Moreover, an 8 cm pelvic mass was found lodged in the cul-de-sac, which was extensively adherent to the bowel and the uterus, and was covered by vascular omental tissue. Histopathological analysis revealed that this pelvic mass was a dermoid cyst. The cyst contained adipose tissue, hair, and microscopic ovarian stroma, confirming the diagnosis. This case highlights the complexity of diagnosing and managing pelvic masses. Clinicians should maintain a high index of suspicion for ovarian torsion and consider the possibility of autoamputation when an ovary is not found in its anatomical location, especially if imaging suggests the presence of a dermoid cyst. This case also underscores the importance of meticulous surgical dissection for the complete removal of such masses. [J Turk Ger Gynecol Assoc.]

Keywords: Pelvic mass, autoamputation of ovary, dermoid cyst, ovarian torsion, spontaneous oophorectomy, teratoma

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Introduction

Ovarian torsion is an infrequent yet consequential gynecologic emergency, characterized by the rotation of the ovary around its vascular pedicle, leading to compromised blood flow, ischemia, and potential ovarian compromise. Incidence rates of ovarian torsion range from 2.7% to 15% in patients undergoing surgical treatment for adnexal masses (1,2). It has been observed that ovarian torsion often occurs in cases involving mature cystic teratoma. However, their varied composition and atypical presentation make the prompt and accurate diagnosis of ovarian torsion challenging.

Ovarian autoamputation is a rare complication of ovarian torsion that may result in the formation of a parasitic ovarian teratoma (3,4). Reimplantation of the ovary after autoamputation is possible due to the phenomenon of neovascularization, which allows for the formation of new blood vessels and subsequent reperfusion. There have been relatively few documented cases in the literature available on this topic although reimplantation of the ovary following

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autoamputation has been reported to the pouch of Douglas, omentum, sacrouterine ligaments, and the broad ligament (5-8). In this case report, we present a case of autoamputation of the ovary resulting from a missed diagnosis of ovarian dermoid cyst torsion.

Case

A 33-year-old gravida 1, para 1 patient was referred to our clinic with a suspected ovarian tumor. Her medical and surgical history was unremarkable, except for hospitalization three weeks earlier for abdominal and pelvic pain ascribed to the presence of an ovarian cyst. The acute phase subsided within 24 hours, however intermittent cramping remined. At the time of presentation to our clinic, the pain had worsened, necessitating regular non-steroidal anti-inflammatory drugs for relief.

Abdominal examination revealed tenderness in the suprapubic region and left iliac fossa. Transvaginal ultrasound examination showed the presence of three ovarian cysts (Figure 1).

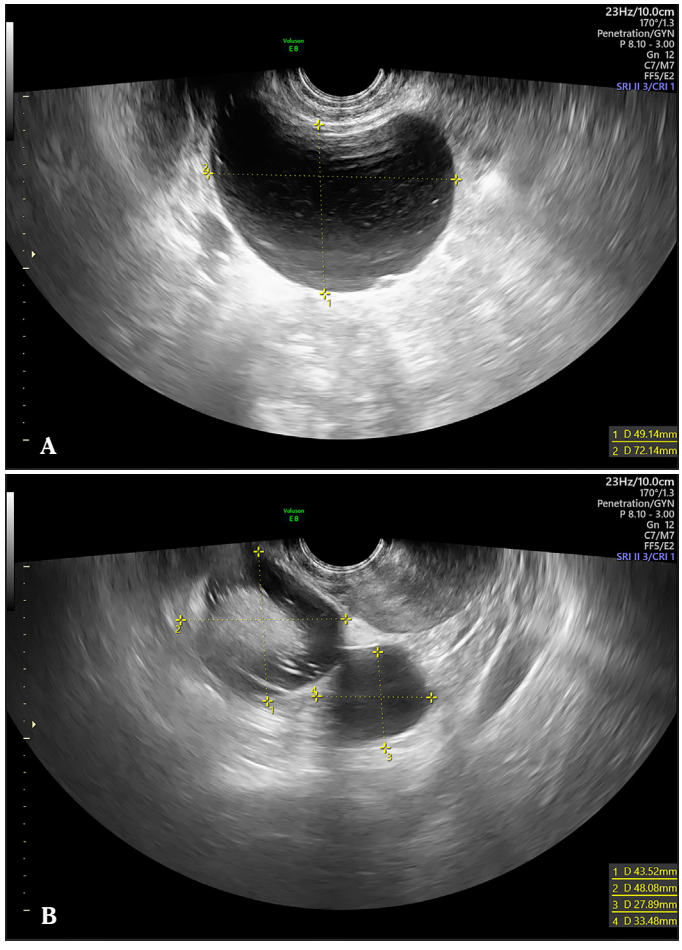

Figure 1. Transvaginal ultrasound imaging showing A) a unilocular cyst with low level echogenity in the right ovary. B) two ovarian cysts: one is a unilocular cyst and the other exhibits hyperechoic lines and dots in the left ovary

The right ovary contained an anechoic unilocular cyst measuring 7x6x5 cm, while the left ovary contained a mixed echogenic cyst measuring 5x4x4 cm and an anechoic unilocular cyst measuring 3x3x4 cm in size. The cyst in the left ovary was consistent with a dermoid cyst located at the pouch of Douglas. Ultrasound with color Doppler demonstrated the presence of blood flow. A laparoscopy was scheduled because of the persistent pain.

At laparoscopy there were extensive dense adhesions in the pelvis precluding the visualization of the internal genitalia. Following lysis of adhesions, it was noted that the left fallopian tube and ovary were absent. The infundibulo-pelvic ligament abruptly terminated at the pelvic brim, while the utero-ovarian ligament was identified as a rudimentary structure arising from the posterolateral aspect of the uterus (Figure 2). A large pelvic mass, approximately 8 cm in size, was found lodged in the cul-de-sac and densely adherent to the bowel and the posterior aspect of the uterus (Figure 3). The mass was covered by a thick and vascular layer of omentum. During careful dissection, the left ureter, adherent to the mass, was identified and dissected meticulously, followed by freeing the mass from the

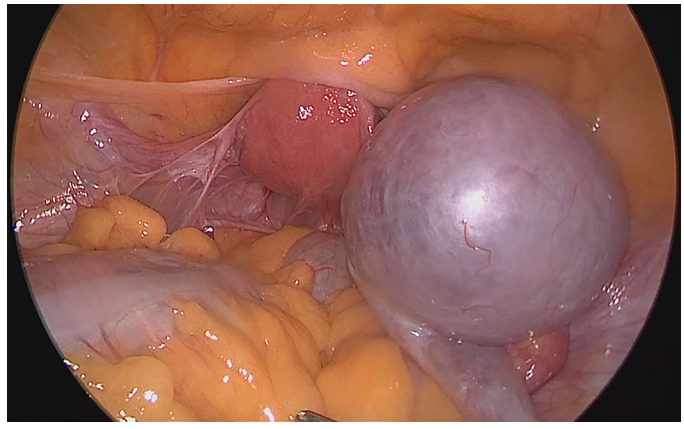

Figure 2. Illustrating the absence left ovary and fimrial portion of the fallopian tube in the fossa ovarica with abrupt end of left infundibulopelvic ligament

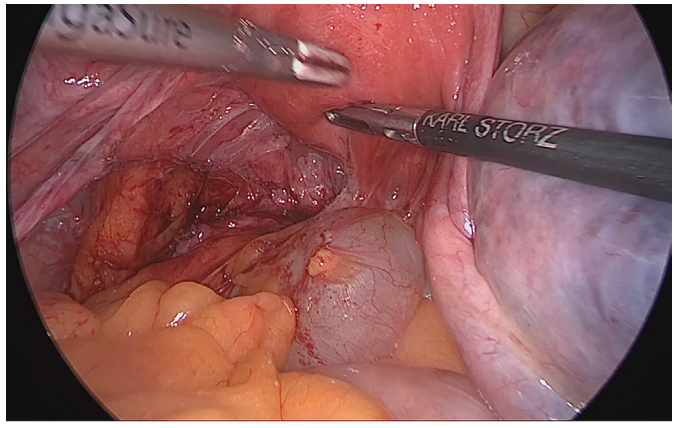

Figure 3. Left ovarian mass and obliteration of Douglas

underlying bowel using the medial pararectal space. Upon full mobilization, two dermoid cysts were removed together with the pelvic mass that had no connection with the right adnexa. The right ovary, adherent to the right pelvic side wall, contained a large cyst that was successfully excised. All specimens were removed inside an endobag through a culdotomy incision. The culdotomy incision was closed using a 3-0 V-Lock suture, and the removed specimens were sent to histopathology. The procedure was terminated without complications, and the patient was discharged the next day.

Histopathological evaluation of the excised specimens showed that the mass removed from the cul-de-sac was a dermoid cyst and the one removed from the right ovary a mucinous cystadenoma. The parasitic mass that was identified as a teratoma contained adipose tissue, hair and also microscopic ovarian stroma (Figure 4).

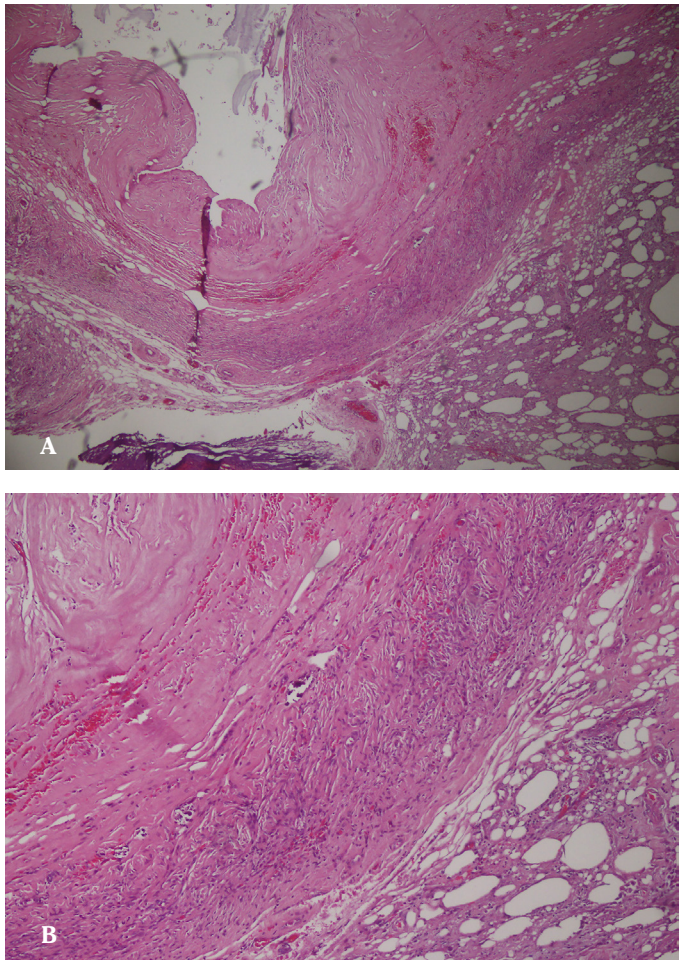

Figure 4. Microscopic findings of the tumour: A) ovarian cyst wall with with hair shafts in the lumen B) ovarian cyst wall and ovarian stroma (hematoxyline & eosin staining, original magnification x40, x200)

Discussion

When torsion occurs, the ovary usually rotates around both the infundibulo-pelvic and the utero-ovarian ligaments. The rotation of the infundibulo-pelvic ligament leads to compression of the ovarian vessels, compromising partial or complete obstruction of lymphatic and venous outflow as well as arterial inflow. Persistent arterial perfusion, coupled with blocked outflow, causes ovarian edema, resulting in significant ovarian enlargement and increased vascular compression. Subsequently, ovarian ischemia develops, potentially leading to ovarian necrosis and local hemorrhage. The treatment of ovarian torsion is via prompt detorsion usually via laparoscopy. However, if the diagnosis is missed and the patient placed on analgesics, the pain usually subsides within a few days and the affected tissues becomes necrotic and eventually undergoes atrophia. In some rare cases, adnexal torsion may lead to autoamputation and subsequent reimplantation of the amputated adnexal mass to the neighboring structures. This may lead to the formation of a parasitic pelvic or abdominal mass, supported by new collateral blood flow (3,7,9-16). Based on earlier case reports, the autoamputated ovary, most commonly harboring a teratoma, was found to be situated at the omentum, pouch of Douglas, attached to the uterosacral ligaments and even within an inguinal hernia. The majority were covered by the omentum, which is unsurprising as omentum is a very mobile organ that controls inflammation and promotes revascularization due to its rich vascular supply. Doppler flow in a torsional ovary may be present, decreased, or absent. Normal Doppler flow does not rule out torsion, as preserved flow can result from incomplete occlusion, intermittent torsion, or collateral blood supply, as observed in our case, where the omentum provided the blood supply.

There are two possible hypotheses for parasitic teratomas; one originating from an autoamputated dermoid cyst, as in the presented case, and the other originating from ectopic ovarian tissue (17-19). The absence of the ovary and the presence of a separate cyst in the pelvic or abdominal cavity covered by omentum supports the first cause. Histopathological observation of ovarian tissue in the excised tumor adds further evidence to support this suggestion.

The second hypothesis for supernumerary ovaries proposes that if the migration of certain primitive germ cells is halted during their journey to the gonadal ridges, their influence on the surrounding epithelium can result in the formation of ectopic ovarian tissue (20,21). Supernumerary ovaries are ovarian tissue that is completely separate from the normally positioned ovary. There is no ligamentous or direct connection with the ovaries, broad ligament, utero-ovarian ligament, or infundibulo-pelvic ligament, as it originates from a distinct

primordium. A supernumerary ovary is typically located in the pelvic region, near structures such as the uterus, bladder, pelvic wall, retroperitoneum, omentum, mesentery, and inguinal region (22-25). In rare cases, supernumerary ovaries have been reported to be located in unusual areas, such as the hepatorenal space, near the right psoas major muscle, and at the intrarenal pole (26,27).

A search was conducted in the PUBMED database to investigate the incidence of reported cases concerning autoamputated ovarian dermoid cysts between 2000 and 2024. Table 1 presents the clinical data for the 10 cases identified. The patients' age ranged from 14 to 77 years. Of these patients, three reported no abdominal pain, four had chronic abdominal pain and the remainder experienced acute abdominal pain. Notably, among those with chronic pelvic pain, the duration of symptoms ranged from 1 to 5 years. Size of the tumor varied between 4 and 10 cm. Autoamputation due to torsion of the ovarian dermoid cyst did not show a preference for laterality. The laterality of origin of the dermoid cyst was found to be similar (six from the left and four from the right ovary). In the majority of cases, preoperative diagnostic imaging was conducted using ultrasound (n=9), followed by computed tomography (n=2) and magnetic resonance imaging (n=2). Preoperative diagnosis was inaccurate in 30% and the presence of a dermoid cyst was confirmed in only half of the cases. Dermoid cyst was bilateral in 30% of cases.

Autoamputation of the ovary is a rare phenomenon with various potential locations for implantation. The most common location was the pouch of Douglas (n=5), followed by the left adnexal region (n=1), peritoneal cavity (n=1), vesicouterine space (n=1), right subhepatic region (n=1), and pelvic side wall (n=1). The unexpected intraoperative finding of the absence of an ovary and the corresponding fallopian tube being blind-ended without fimbriae and infundibulum usually leads to the diagnosis of ovarian autoamputation. In cases when bilateral dermoid cysts are present and one ovary has been autoamputated and migrated, misdiagnosis is quite common, resulting in incomplete removal of all cystic components. In a 14 year-old girl, the initial intraoperative diagnosis was right ovarian dermoid cyst and congenital absence of left ovary. Following right ovarian cystectomy, the 4 cm left dermoid cyst was successfully removed after a thorough pelvic exploration and meticulous dissection from the surrounding omental adhesions. In another case, a 42-year-old patient was preoperatively diagnosed with an 8 cm right ovarian cyst, suggestive of cystadenoma, and a 2 cm echogenic left ovarian cyst. Initially, the plan was to perform a bilateral ovarian cystectomy.

However, due to technical difficulties encountered during surgery, an adnexectomy for the right ovarian cystadenoma was performed. Unexpectedly, the left ovary was not found in its usual location in the ovarian fossa; instead, it was discovered completely detached and located in the pouch of Douglas. Consequently, the patient underwent a bilateral adnexectomy, which inadvertently triggered the onset of menopause. Meticulous localization of anatomical structures can significantly influence surgical outcomes and patient well-being. It is important for the surgeon to have a high level of suspicion, thorough preoperative evaluation, and intraoperative assessment to identify any migrated cysts and ensure complete removal, especially in cases when ovary is not identified in its normal anatomical location and there are pelvic adhesions enveloped by omentum.

The removal of adnexal specimens through an abdominal port site or posterior culdotomy incision is a feasible and safe option for pelvic specimen extraction. This approach can be tailored based on the preferences of the patient and surgeon, as well as individual patient factors. Colpotomy negates the need to enlarge abdominal incisions or performing intracorporeal specimen size reduction. We particularly prefer this method in obese patients to minimize the risk of port-site hernias and skin infections. For closing the cul-de-sac incision, we favor the use

Table 1. Articles about autoamputation of the ovary

Author, year	Age	Symptom	Size	Preop diagnosis	Location of AO
Daccache et al. (9)	42	Aysmptomatic	80 mm	Cystadenoma	Douglas pouch
Gorginzadeh et al. (10)	14	Chronic pelvic pain	101x60 mm (RO) and 40x25 mm (LO)	Dermoid cyst	Douglas pouch
John (11)	32	Asymptomatic	60 mm	Complex cyst	Douglas pouch
Kim et al. (12)	34	Chronic pelvic pain	50x27 mm	Complex cyst	Left adnexal region
Lee et al. (3)	77	Acute abdominal pain	143x140 mm, 90 mm	Dermoid cyst	Right subhepatic space
Kusaka and Mikuni (7)	24	Chronic pelvic pain	50x35 mm	Dermoid cyst	Douglas pouch
Ollapallil et al (13)	46	Acute abdominal pain	80x60x40 mm	Complex cyst	Peritoneal cavity
Peitsidou et al. (14)	33	Asymptomatic	Incidental finding	NA	Douglas pouch
Shah et al. (15)	26	Chronic pelvic pain	40x40 mm	Dermoid cyst	Vesicouterine space
Üreyen et al. (16)	27	Acute abdominal pain	68x40 mm	Complex cyst	Pelvic side wall

of a barbed suture material, which provides consistent tension along the suture line without the need for knots. This feature is particularly advantageous in laparoscopic settings, where space is limited and visibility can be challenging. This suture type not only enhances the speed of closure but also improves the overall security of the incision, thereby reducing the risk of cuff dehiscence.

Conclusion

It is important for the surgeon to have a high level of suspicion, thorough preoperative evaluation, and intraoperative assessment to identify any migrated cysts and ensure complete removal, especially in cases when ovary is not identified in its normal anatamical location and there are pelvic adhesions enveloped by omentum.

Footnotes

Author Contributions: *Surgical and Medical Practices: B.U., Concept: A.S., İ.U.K., Design: A.S., İ.U.K., Data Collection or Processing: A.S., İ.U.K., Analysis or Interpretation: A.S., İ.U.K., Literature Search: A.S., İ.U.K., Writing: A.S., İ.U.K., B.U.*

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