

Parasitic teratoma in the pouch of Douglas

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Summary

Teratoma is the most common type of germ cell tumor, which is better known as dermoid cyst. Dermoid cyst is the most common ovarian tumor in women in the second and third decades of life. Parasitic teratoma, which is autoamputation of an ovarian teratoma and reimplantation into a different place, is rare, and it is extremely rare if it is in the pouch of Douglas. Only a few cases have been found in this location so far. Here, the authors report an interesting case of parasitic teratoma. They draw the conclusion that if intraperitoneal teratoma is suspected in an imaging examination, surgeons should consider the possibility of adnexal autoamputation, including asymptomatic torsion.

Key words: Parasitic teratoma; Ovarian teratoma; Torsion.

Introduction

Teratoma is the most common type of germ cell tumor, which is better known as dermoid cyst. Dermoid cyst is the most common ovarian tumor in women in the second and third decades of life [1]. Parasitic teratoma is extremely rare. There are three theories regarding the pathogenesis of parasitic teratoma, and the most prevalent one is autoamputation of an ovarian teratoma and reimplantation into a different place. Most patients initially have non-specific symptoms including pelvic pain, dyspareunia, or abnormal, vaginal bleeding. A diagnosis of parasitic teratoma is often an incidental event during an operation. They can sometimes cause secondary symptoms, which depend on their

location and size. Further examinations, such as transvaginal ultrasound, measurement of CA 19-9 concentration, and MRI are usually performed. The authors report an interesting case of parasitic teratoma in the pouch of Douglas. They draw the conclusion that if intraperitoneal teratoma is suspected in an imaging examination, surgeons should consider the possibility of adnexal autoamputation, including asymptomatic torsion.

Case Report

A 34-year-old woman, gravida 3, para 3, came to the present clinic in September 2016 for two years history of left ovary mass. She had a history of acute left lower abdominal pain during pregnancy one year ago but had no previous operation. Clinical ex-

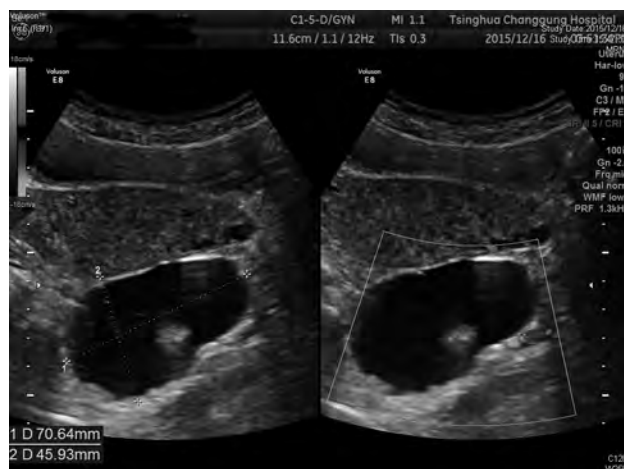


Figure 1. — Transvaginal ultrasound examination reveals a cyst measuring 70×46×53 mm with diffuse internal echoes, including a 25×17 mm hyperechogenic component without significant vascularity in the pouch of Douglas.



Figure 2. — Transvaginal ultrasound examination reveals a cyst measuring 70×46×53 mm with diffuse internal echoes, including a 25×17 mm hyperechogenic component without significant vascularity in the pouch of Douglas.

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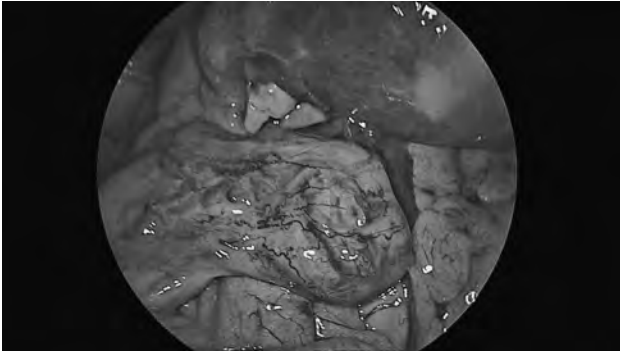


Figure 3. — On initial laparoscopic examination of the abdominal cavity, the left ovary is very small and defected.



Figure 6. — The mass is carefully dissected from the surrounding omentum using a harmonic scalpel.

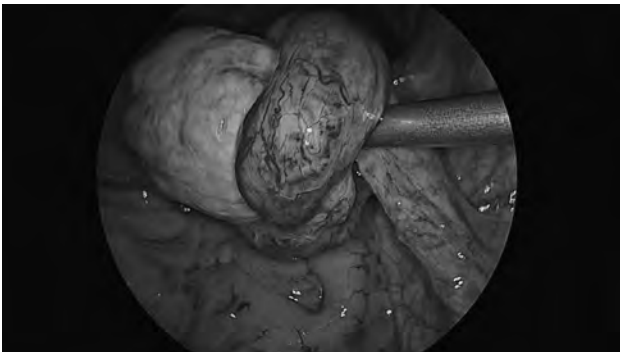


Figure 4. — The uterus, both of the oviduct and the right ovaries, are observed as normal.

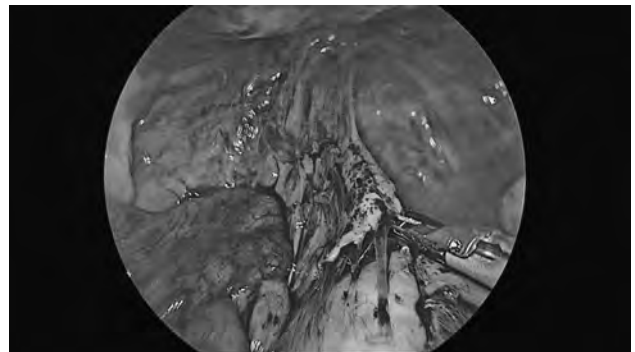


Figure 7. — The mass is attached to the left sacral ligament.

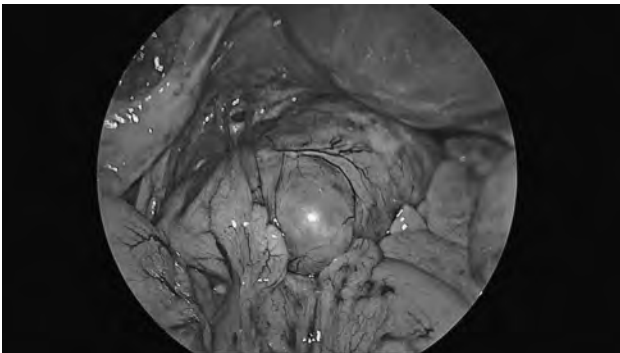


Figure 5. — The abdominal mass is surrounded by thin filmy adhesions to the omentum.

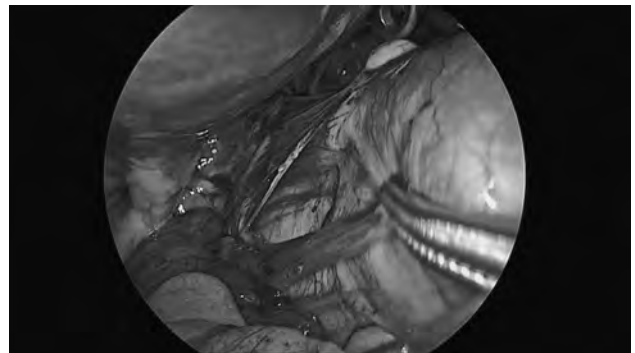


Figure 8. — The pedicle division appears to be on the left ovary.

amination revealed palpable a mass in the left lower quadrant without tenderness and rebound tenderness. Transvaginal ultrasound examination revealed a cyst measuring 70×46×53 mm with diffuse internal echoes (Figure 1), including a 25×17 mm hyper echo-genic component without significant vascularity in the pouch of Douglas (Figure 2). The left ovary was not seen and the uterus and right ovary were normal. The suggested preoperative diagnosis was benign teratoma of the left ovary. With regards to preoperative examination, the laboratory tests, complete blood counts, urinalysis, biochemical tests, and blood coagulation profile were all normal. CA 19-9 was elevated at 52.87 (normal range

0-37 U/mL), other concentrations of carcinoembryonic antigen (CA-125, AFP, CEA) were normal. The patient was adequately informed of the possible risks and benefits of laparoscopic surgery and signed a written consent agreeing to undergo laparoscopic exploration. The case was approved by the Institutional Review Board of the Beijing Tsinghua Changgung Hospital. Laparoscopic exploration was performed. On initial laparoscopic examination of the abdominal cavity, the left ovary was very small and defected (Figure 3). The uterus, both of the oviduct and the right ovaries were observed as normal (Figure 4). The tumor identified preoperatively located at the back of the uterus, with no con-

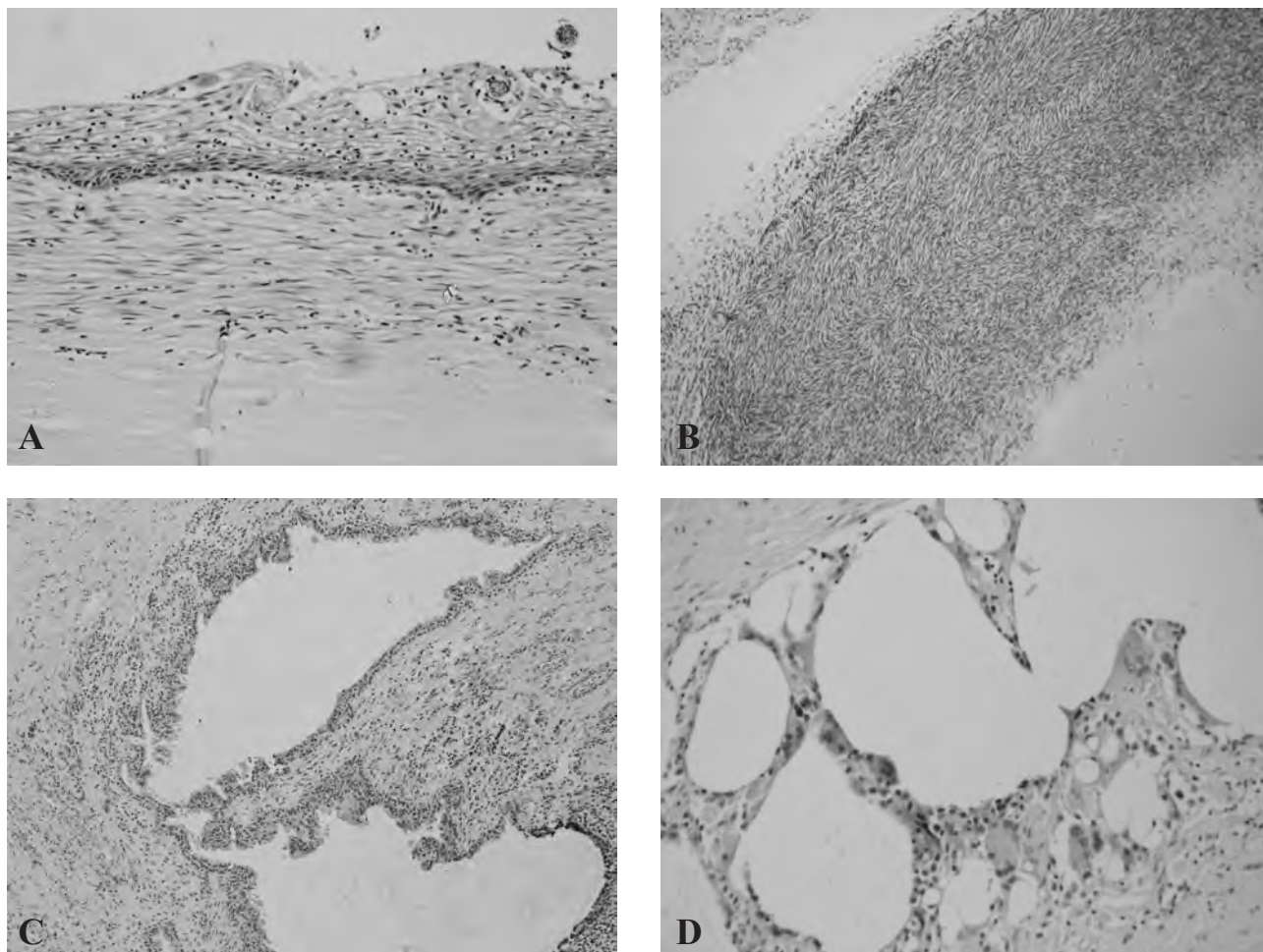


Figure 9. — Histologic examination revealed a typical mature teratoma involving squamous and respiratory epithelium, lipogranulomatous reaction. Ovarian interstitial tissue was observed in the cyst wall, and the diagnosis of parasitic dermoid cyst can be made.

tinuity to any of the ovaries, and countless tumor feeding vessels from the peritoneum were observed. The abdominal mass was surrounded by thin filmy adhesions to the omentum (Figure 5). The mass was carefully dissected from the surrounding omentum using a harmonic scalpel (Figure 6). The authors found the mass attached to the left sacral ligament (Figure 7) and the pedicle division seemed to be on the left ovary (Figure 8). The authors supposed that the left ovarian teratoma underwent torsion, autoamputation, and then reimplanted in the omentum and Douglas pouch. They completed the resection of the mass and the teratoma was removed intact through the right incision without leakage of content using a plastic bag. Macroscopically, the size of mass was 7×4×3 cm. It was a cystic mass filled with sebaceous material. Histologic examination revealed a typical mature teratoma involving squamous and respiratory epithelium, lipogranulomatous reaction. Ovarian interstitial tissue was observed in the cyst wall, and the diagnosis of parasitic dermoid cyst was made (Figure 9). These findings could also be interpreted as an autoamputation of the adnexa due to torsion of a previous ovarian cyst arising from the left ovary. The patient recovered quickly and was discharged three days after the surgery. The patient had an unremarkable postoperative period and has been asymptomatic to date.

Discussion

Teratoma is the most common type of germ cell tumor. Most teratomas are benign. The teratoma refers to a neoplasm that differentiates toward somatic-type cell populations (typically including cell populations that would normally derive from ectoderm, endoderm, and mesoderm) which can be typical of either adult or embryonic development. The ingredient tissues in a teratoma range from immature to well-differentiated and are foreign to the anatomic site in which they are found. Most teratomas are cystic and composed of mature differentiated elements (mature); they are better known as dermoid cysts. The mature cystic teratoma accounts for more than 95 percent of all ovarian teratomas and is almost invariably benign [1]. Dermoid cysts are the most common ovarian tumor in women in the second and third decades of life. Although teratomas most commonly occur in the ovaries, they have been reported in a number of other locations (0.4 % of all teratomas) [2]. Extragonadal teratomas occur anywhere along

the midline of the body, such as the mediastinum, because of the migration of germ cells in embryonic life. However, parasitic or extragonadal teratomas in the abdominal cavity are extremely rare and the most common extragonadal site is the omentum.

The present authors report a case of extragonadal teratoma in the pouch of Douglas that appeared to be a parasitic dermoid cyst. From the authors' experience and the literature review, they discussed the potential mechanism leading to the development of extragonadal teratomas. The mechanism underlying the development of extragonadal teratomas remains unknown. Three proposed theories for extragonadal sites of teratoma exist: [1] autoamputation of an ovarian teratoma and reimplantation into a different place, [2] development in a supernumerary ovary, and [3] origination from displaced germ cells [3]. The first theory is that autoamputated extragonadal teratomas from an ovarian site may reimplant in another site in the abdominal cavity. This theory is widely accepted as the etiology of extragonadal teratomas in the abdominal cavity. Torsion of the pedicle has been reported to be the most frequent complication of ovarian teratomas. Autoamputation was first reported, along with possible mechanisms, by Thornton in 1881[4].

In acquired cases of autoamputation, it mainly results from chronic torsion or a delay in the diagnosis of acute adnexal torsion. Torsion of the pedicle has been reported to be the most frequent complication of ovarian teratoma, occurring in 16.1% of cases [2]. Ovarian autoamputation is a pathologic complication of ovarian torsion that may result in the formation of a parasitic ovarian teratoma. The blood supply of the involved organ is impaired when torsion occurs, which may result in venous congestion and aseptic inflammation of the tumor wall. In an acute event, the tumor undergoes necrosis and subsequent atrophy due to ischemia. In a subacute or chronic event, the tumor may become adherent to adjacent structures with the formation of new collateral blood flow [5]. In a rare event, the tumor will become completely detached from the pedicle and become a parasitic teratoma. Although the mechanisms of autoamputation are still unclear, adnexal torsion is believed to lead to infarction, necrosis, and autoamputation. Theoretically, a parasitic ovarian teratoma results from torsion, autoamputation or detachment, and subsequent reimplantation or persists as a peritoneal loose body. The detachment of an ovarian teratoma may also occur without adnexal torsion. It has been suggested that adhesion formation with neovascularization between the tumor and the omentum may occur.

Most patients have non-specific initial symptoms including pelvic pain, dyspareunia or abnormal vaginal bleeding. If present, symptoms depend upon the size of the mass. They can sometimes cause secondary symptoms, which depends on their location [6]. Torsion is known to be the most frequent complication of ovarian teratomas. Torsion of the adnexa usually manifests with severe ab-

dominal pain and is treated as an acute surgical emergency. However, it may be asymptomatic. Autoamputation of an ovary, due to previous torsion is extremely rare. A history of acute left lower abdominal pain may support the diagnosis in the present case. These tumors have a characteristic ultrasound appearance. The reported specificity is 98-100% [5, 7]. In the present case, transvaginal ultrasound examination revealed a cyst measuring 70×46×53 mm with diffuse internal echoes, including a 25×17 mm hyperechoic component without significant vascularity in the pouch of Douglas. The left ovary was not seen and the uterus and right ovary were normal. Serum CA19-9 in patients with parasitic teratoma is normal or slightly increased. The present patient's serum CA19-9 was 52.87 (normal range 0-37) U/mL.

Definitive diagnosis is made at the time of laparoscopic exploration. Teratoma may be removed via either laparoscopy or laparotomy. During the laparoscopic examination of the abdominal cavity, the left ovary was very small and defected. The tumor located at the back of the uterus, with no continuity to any of the ovaries, and countless tumor feeding vessels from the peritoneum and the left sacral ligament, and the pedicle division seemed to be on the left ovary. The present authors suppose the left ovarian teratoma that underwent torsion, autoamputation, and reimplantation was located in the omentum and Douglas pouch of a patient. They placed the patient in reverse Trendelenburg position after complete excision and repeatedly irrigated the abdomen and pelvis with normal saline may be helpful to wash out small pieces in pelvis.

The characteristic macroscopic appearance of teratomas is a multicystic mass that contains hair, teeth, and/or skin that is mixed into sebaceous, thick, sticky, and often foul-smelling material. A solid prominence (Rokitansky's protuberance) is located at the junction between the teratoma and normal ovarian tissue. In the present case, the lesion was a 7×3×4-cm cystic mass filled with sebaceous material.

Mature cystic teratomas contain mature tissue of ectodermal (eg, skin, hair follicles, sebaceous glands), mesodermal (eg, muscle, urinary), and endodermal origin (eg, lung, gastrointestinal) [8]. The mechanism by which these cysts develop is possibly by failure of meiosis II or from a premeiotic cell in which meiosis I have failed [9]. In the present case, histologic examination revealed a typical mature teratoma involving squamous and respiratory epithelium, lipogranulomatous reaction, and ovarian interstitial tissue was seen in the wall, which led to the diagnosis of parasitic dermoid cyst. These findings could also be interpreted as an autoamputation of the adnexa due to torsion of a previous ovarian cyst arising from the left ovary.

Conclusion

In conclusion, because the parasitic teratoma specimen included ovarian tissue and the left ovary was small and at-

rophied in the present case, the authors concluded that the teratoma resulted from the autoamputation of a left ovarian teratoma. The present patient had a history of acute pain that was suspicious for previous ovary torsion. Although ultrasonography was performed preoperatively in the present patient, the correct diagnosis of autoamputation and exact localization of the teratoma were extremely difficult.

If intraperitoneal teratoma is suspected in an imaging examination, surgeons should consider the possibility of adnexal autoamputation, including asymptomatic torsion. It is important that all physicians treating female patients in their reproductive age, especially with a pelvic mass and lower abdominal pain, they must be aware of the possibility of adnexal torsion with regards to fertility and ovary preservation. If symptoms persist, early diagnostic laparoscopy for exact diagnosis and therapy is recommended.

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