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Concurrent Descending Mesocolon, Ovary And Anterior Abdominal Wall; A Rare Presentation Of Extrauterine Adenomyoma

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ORIGINAL

Abstract

Extrauterine adenomyoma is an extremely rare condition with no known incidence rate. It is a benign tumour that develops from smooth muscle and endometriotic tissue. We present a case of a 42-year-old woman who had a history of myomectomy for uterine fibroid and presented with a growing left-sided abdominal tumour for 3 years. A CT scan of the abdomen revealed several heterogeneous masses affecting the descending mesocolon, ovary, and anterior abdominal wall. An ultrasound-guided tru-cut biopsy of a left iliac fossa intraperitoneal tumour confirmed the diagnosis of extrauterine adenomyoma. She had a segmental descending colon resection with side-to-side anastomosis, as well as additional lesions removed. Despite the rarity of the occurrence, clinical, imaging, and histological examinations remain the primary diagnostic modalities.

Introduction

Adenomyoma is a benign tumor made up of smooth muscle cells and endometriotic tissue that usually originates in the uterus. Extra uterine adenomyoma is extremely rare. Because of its rarity, extrauterine adenomyoma has no exact incidence rate. The most common sites being reported were the pararectal space, the ovary, and the broad ligament. There are other sites include round ligament, paraovarian, parametrial, pelvic wall, liver, upper abdomen, inguinal scar, appendix, small bowel as well as large bowel mesentery. To the best of our knowledge, only 10 cases of concurrent multiple sites extrauterine adenomyoma have been reported up until 2018. Therefore, we like to present another case of concurrent multiple extra uterine adenomyoma involving descending mesocolon, ovary and anterior abdominal wall.

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Case Report

A 42-year-old lady who had a history of myomectomy for uterine fibroid presented with left side abdominal mass for 3 years duration. It was increasing in size and associated with occasional prinking pain. Otherwise, there was no intestinal obstruction symptom, per rectal bleeding, loss of weight, or loss of appetite. Clinically there was 8cm x 8cm anterior abdominal wall mass at the left iliac fossa region which was firm and mobile. The mass becomes more prominent upon the rising of the head. The overlying skin was normal. On the other hand, there was vague fullness intraabdominal at the left iliac extending to the suprapubic region. Computerized tomography (CT) scan of the abdomen and pelvis showed a large left-sided heterogenous intraperitoneal enhancing mass measuring 9.1 x 12.3 x 10.2 cm (AP x W x CC) with an area of necrosis within (Figure1). There is a fat component or calcification within. The mass also has no clear fat plane

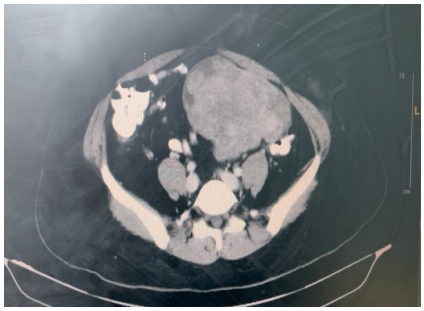
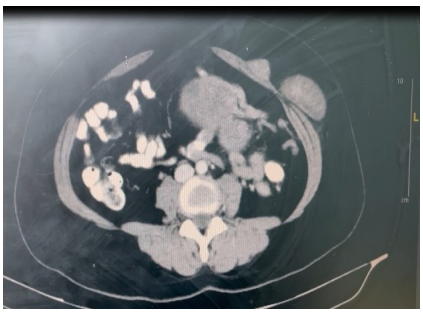


Figure 1



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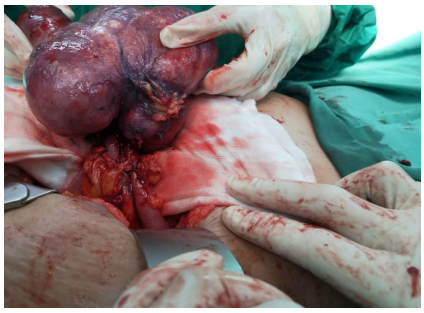

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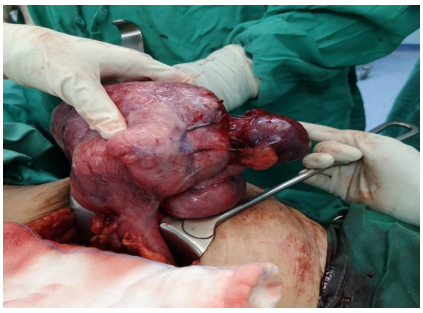

Figure 4

to the adjacent small bowel loop as well as the sigmoid colon. There was another heterogenous enhancing lesion near the rectosigmoid colon measuring 1.9 x 2.0 x 1.9cm. There were also two subcutaneous heterogenous enhancing masses at the left anterior abdominal wall measuring 3.0 x 7.1 x 6.4 cm and 1.6 x 2.5 x 6.4 cm respectively (Figure2).

A colonoscopy was done, and no evidence of intraluminal colonic origin was found. An ultrasound-guided tru-cut biopsy of a left iliac fossa intraperitoneal mass was performed, which was initially diagnosed as a smooth muscle tumor favoring leiomyoma. Following that, a laparotomy was performed, which revealed a large lobulated mass measuring 20 x 15cm arising from mesocolon of the descending colon with no clear serosa layer of the colon (Figure3) (Figure4) and a 3 x 3 cm mass attached to the right ovary (Figure5). There was a large subcutaneous tumor measuring 8 x 8 cm in the left iliac fossa region (Figure6), as well as a smaller tumor attached to the left external oblique aponeurosis and filled with necrotic material (Figure7).

A segmental descending colon resection with side-to-side anastomosis was performed. All other tumors were completely excised. On the fifth day after surgery, she was discharged without complications. Histopathology examination showed a large glandular mass arising from the mesocolon. The mass has a well-circumscribed outline composed of intersecting fascicles of band-looking spincle cells with large foci of myxoid degeneration. The cells display uniform, cigar shape nuclei with small nucleoli and eosinophilic cytoplasm with indistinct cellular borders. No overt cytological atypia or mitosis is found. The cystic cavity shows a dilated endometrial gland lined by columnar endometrial-type epithelium with the presence of endometrial stroma. Otherwise, the colono shows unremarkable mucosa, muscular wall, and intermyenteric plexus. Furthermore, microscopic examination of the mass attached to the right ovary and large subcutaneous lesion showed the presence of myxoid stroma with the proliferation of bland spindle-shaped cells in interlacing fascicles. There was no atypia seen as well. On the other hand, mass arising from left external oblique aponeuresis has an area of a cavity lined by a single layer of benight flat columnar epithelium with the presence of endometrial stroma. Therefore the histophatohy diagnosis of extrauterine adenomyoma was made.

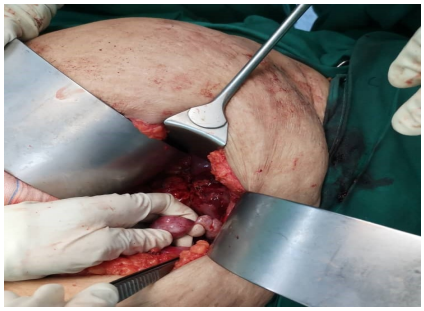

Figure 5

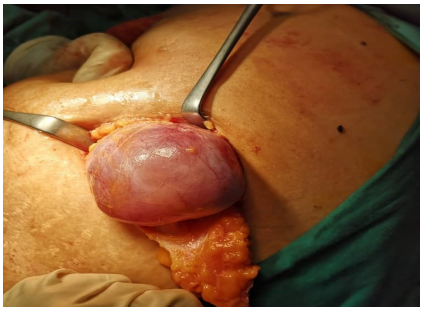

Figure 6

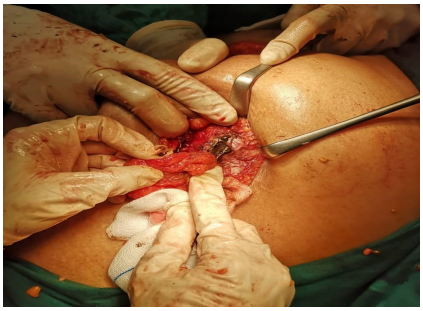

Figure 7

Discussion

Adenomyomas are benign tumours characterized by benign endometrial glands and endometrial stroma surrounded by leiomyoma-like smooth muscle and typically found in the uterus (1). Because of its rarity, there is no recorded incidence rate for the disease. The pararectal space, the ovary, the broad ligament, round ligament, para ovarian, parametrial, pelvic wall, liver, upper abdomen, inguinal area, appendix, small bowel, and large bowel mesentery were among the sites documented (2). Only 10 cases of concurrent multiple locations extrauterine adenomyoma have been recorded to our knowledge(Table 1). As in our case, the mass is located at descending mesocolon, right ovary, and anterior abdominal wall.

Table 1

No.	Paper (Year)	Site
1	Carinelli et al, 2009(3) • Case 1 • Case 2	Sigmoid, pelvis, ileal, paravesicle Sigmoid, right ovary
2	Moghadamfalahi et al, 2012(4)	Para- rectal and upper abdomen
3	Carvalho et al, 2012(5) • Case 1 • Case 2	Pelvis, peritoneum, omentum, left ovary Pelvis, peritoneum, omentum
4	Ki Yong Na et al, 2013(6)	Caecum, descending colon, mesocolon
5	Bulut et al, 2013(7)	Bilateral broad ligament
6	Paul et al, 2018(2)	Pararectal and right ovary
7	Gruttadauria et al, 2019(1)	Right ovary,anterior to rectum, bilateral uterosacral area, and sigmoid mesentery
8	Belmarez et al, 2019(8)	Multiple mass adherent to rectosigmoid colon, vaginal cuff and descending colon

Several theories have been proposed to explain the etiology of extrauterine adenomyoma. Rosai (1982) proposed two theories: the Müllerian duct fusion defect and the subtelomeric mesenchyme transformation theory (9). A lack of fusion of the Mullerian duct system may explain various

uterine duplications or atresias. When this defect results in a unicornuate uterus with a primitive horn, the horn can detach and implant elsewhere, resulting in a uterus-like mass. Subcoelomic mesenchyme gives rise to the urogenital ridge mesenchyme that surrounds the early Müllerian and Wolffian ducts in a fetus. In adults, the subcoelomic mesenchyme appears as a thin layer of flattened cells just beneath the subserosal stroma of the uterus, ovaries, tubes, and uterine ligaments. The cells in this layer, also known as the secondary Müllerian system, are thought to be multipotent and capable of proliferating in response to hormonal stimulation.

Another theory proposed by Batt (2010) is that heterotopic Müllerian-like organoid tissue of embryonic origin could develop within other normal organs during organogenesis, explaining Müllerian tissue with no obvious source of dispersion (10; 11). Following that, (Belmarez et al., 2019) discussed another possibility of disease etiology as pelvic seeding during surgeries to the reproductive tract. It has been proposed that cells can be seeded within the abdomen and pelvis during hysterectomy or myomectomy, giving rise to extrauterine leiomyomas (8).

The most common symptom of extrauterine adenomyoma is abdominal and pelvic pain (12; 13). History of gynecological surgery, such as hysterectomy, ovarian cystectomy, or myomectomy, may be useful in determining the presence of extrauterine adenomyoma (14). In our situation, the patient had previously had a myomectomy and presented with a palpable mass on both the intraperitoneal and anterior abdominal walls. Other nonspecific symptom includes heavy menstrual bleeding, mid-cycle pain, and infertility (2). Because of its rarity and non-specific clinical and radiological findings, extrauterine adenomyoma is difficult to diagnose before surgery.

Histopathologically, the presence of endometrial gland scattered throughout smooth muscle component concomitant with endometrial stroma is a common histopathological hallmark of adenomyoma, which is congruent with our case (15). In the cases that presented with uterine-like mass, histological examination revealed extrauterine organoid masses, which are characterized by a single central cavity lined by endometrium and surrounded by a thick wall of smooth muscle, mimicking a normal uterus (16).

Surgical resection is by far the most common method of treating extrauterine adenomyoma. The surgical approach, whether laparoscopic or open, is determined by the available expertise as well as the location of the disease. For long periods of observation, a GnRH agonist was used to inhibit the estrogen-stimulated growth of the neoplastic tissue and prevent recurrence (17). This has a connection to the subtelomeric mesenchyme transformation idea, according to which the secondary Müllerian system can proliferate with hormonal stimulation.

Even though extrauterine adenomyoma is a benign disease, it has an association with the malignant condition. Torres et al reported clear cell adenocarcinoma in a case of broad ligament adenomyoma (18). Another case of concurrent malignant condition is reported by Rahilly et all in which presence of ovarian endometrioid carcinoma and uterine endometrial carcinoma together with ovarian carcinoma (19). Long-term follow-up is suggested for potential recurrence of the disease as reported before (20).

Conclusion

In numerous sites, extrauterine adenomyoma is a rare occurrence and a diagnostic challenge. The definitive diagnosis is dependent on histopathology examination. Surgical resection remains the mainstay of treatment and long-term follow-up is suggested because of the risk of recurrence.

Conflict Of Interest

All authors declare no conflict of interest of any kind.

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