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# Uterus Like Mass of Broad Ligament Presenting as Palpable Mass – A Rare Case Report

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Abstract: We report a case of uterus like mass of left broad ligament presenting as palpable mass in a 45-year-old female. A mass in the region of left broad ligament demonstrated a cavity lined by endometrium and surrounded by bundles of smooth muscles resembling myometrium. Review of literature disclosed only about 30 cases of uterus like mass at different sites. Clinically uterus like mass of broad ligament presents as dysmenorrhoea with or without per vaginal bleeding. Presentation as palpable mass is very rare. Histogenesis of this entity is still unknown, however several theories have been proposed. Theory of transformation of subcoelomic mesenchyme is most likely in present case.

Keywords: uterus like mass, endomyometriosis, adenomyoma, broad ligament

### 1. Introduction

The entity 'uterus like mass' is proposed by Cozzutto in 1981 [1]. It is characterised by a cavity lined by pseudostratified columnar epithelium resembling endometrium and surrounded by bundles of smooth muscles resembling myometrium. Very few cases involving ovary [2], small bowel mesentery [3], obturator lymph node [4], colon [5] have been reported. To the best of our knowledge only five cases of uterus like mass of broad ligament have been published [6]-[10]. Although extremely rare, it is a well-recognized clinicopathological entity.

## 2. Case Report

A 45-year-old female presented with pain in left lower abdomen for two months. The intensity of pain increased during her last menstrual cycle. Her past history was otherwise unremarkable. Patient's general condition was good. On per abdominal examination, an ill-defined lump was palpated in left iliac fossa. Her per vaginal examination revealed fullness in left fornix. Ultrasonography revealed well defined hypoechoic lesion measuring  $7.3 \times 7.1 \times 6.7$  cm in the region of left adnexa. The mass was seen attached to the ovary. Magnetic resonance imaging study showed a well-defined lobulated mass of size  $7.8 \times 7.3 \times 7.1$  cm in left adnexa. Differential diagnosis of left ovarian neoplasm or broad ligament fibroid was proposed.

We received total abdominal hysterectomy and bilateral salpingo-oophorectomy specimen with a mass attached on left side of uterus in the region of broad ligament. The well circumscribed, encapsulated mass measured 7 x 7 x 6.5 cm<sup>3</sup> with smooth external surface (figure 1A). Cut surface of the mass revealed a longitudinal, slit like cavity surrounded by firm trabeculated areas (figure 1B). Dark red viscous fluid exuded on sectioning the mass. Unremarkable left ovary and fallopian tube could be identified attached to the mass. Microscopic examination of the mass demonstrated cavity lined by endometrium like epithelium and surrounded by bundles of smooth muscles resembling myometrium, confirming the diagnosis of uterus like mass (figure 2). Gross and microscopic examination of uterus, cervix and bilateral adnexa were unremarkable.

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Figure 1 A: Gross photograph showing uterus like mass



Figure 1 B: Cut surface showing longitudinal slit like cavity (arrow) attached to left side of uterus



Figure 2 A: Microphotograph (H and E- 40X) showing slit like cavity lined by epithelium resembling endometrium and surrounded by thick bundles of smooth muscles resembling myometrium.



**Figure 2 B:** Microphotograph of the same, higher power (H and E - 100X).

## 3. Discussion

Uterus like mass is extremely rare entity and is defined as an extra-uterine mass composed of smooth muscle and a central cavity lined by endometrium, similar in structure to a normal uterus [11]. The debate is going on whether the finding is termed endomyometriosis, adenomyoma or ovarian leiomyoma. [3] Moghadamfalahi et al [12] have differentiated these entities in their case report of multiple extrauterine adenomyomas. They have excluded endomyometriosis due to absence of adhesion or obvious continuity of the masses with surrounding normal stromal tissue. Uterus like masses should be differentiated from adenomyomas, which lack a uterus-like organization. The lesions described as extrauterine adenomyomas are composed of what appears to be a leiomyoma with scattered foci of endometriosis without making a distinct central cavity.

Only five cases of uterus like mass of broad ligament have been described. Most common clinical presentation is dysmenorrhoea. It is due to hormone responsive endometrial tissue within a confined cavity of uterus like mass. Severe progressive dysmenorrhoea results due to absence of channel for outflow. Second most common clinical presentation is lower abdominal pain with or without bleeding per vaginum. It is seldom presented as a palpable mass.

The Histogenesis of uterus like mass is uncertain, however four theories have been proposed; Mullerian duct fusion defect theory, subcoelomic mesenchyme transformation theory, theory of heterotopia and theory of mullerianosis.

The basis of mullerian duct fusion defect theory is developmental abnormality occurring during formation of female genital tract. Among the two pairs of genital ducts; mesonephric (Wolffian) and paramesonephric (Mullerian); lack of fusion of latter in localised area or throughout the length of the duct may explain various duplications and atresias of the uterus [13]. Pueblitz-Peredo [2] et al. and

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Rosai [14] postulated that these uterine-like masses result from a müllerian duct fusion defect. Moreover, this theory is based on the association of the condition with congenital anomalies, including renal agenesis, pelvic kidney, double excretory system, anomalies of the lower genital and intestinal tracts, sacral agenesis, and spinal dysraphism [2]. Present case did not have any of the congenital anomalies, thus the mass is less likely to have originated from mullerian duct fusion defect.

The subcoelomic mesenchyme is layer of tissue that lies underneath the mesothelial surface of peritoneum. In embryonic life it gives rise to mesenchyme of urogenital ridge that surrounds the early Mullerian and Wolffian ducts. In adults it is represented by layer of flattened cells that blend into the subserosal stroma of the uterus, ovaries, tubes, and uterine ligaments. Epithelial lesions of this system may show endometrioid, serous, mucinous or transitional differentiation. While proliferation of the mesenchyme may give rise to mesenchymal lesions composed of endometrial stromal-type cells, decidua, or smooth muscle. This is known as subcoelomic mesenchyme transformation theory [15], [16].

The cells of subcoelomic mesenchyme also known as secondary mullerian system are thought to be pluripotent and may proliferate in response to hormonal stimulation. These cells may be converted into uterine tissue by differentiating into endometrial stromal cells, decidua, or smooth muscle cells under hormonal influences. In a case of uterus like mass with features of an extrauterine adenomyoma presenting 22 years after total abdominal hysterectomy with bilateral salpingo-oophorectomy of a 50year-old lady, Redman et al [16] has supported metaplasia of subcoelomic mesenchyme theory, as the patient had been receiving regular estrogen therapy because of premature menopause. Scully [17] has described uterus like mass in the scrotum of men receiving estrogen therapy for prostate carcinoma. This also supports the hormonal responsiveness of secondary mullerian system.

Peterson et al [18] reported a 12 years old girl with uterus like mass in the ileum with multiple lower intestinal & urogenital tract abnormalities, sacral agenesis and sacrocoocygeal teratoma. They concluded that neither the congenital anomaly nor the metaplasia theory provided an entirely satisfactory explanation and that heterotopia or choristoma was the cause for the uterus like mass.

Liang et al [8] have reported a case of 'uterus like mass of broad ligament' immediately adjacent to uterus but without definite connection to the uterus, in a 17-year-old girl. They have suggested theory of metaplasia as both glandular and stromal smooth muscle cells proliferate like a true neoplasm. Batt [19] have suggested theory of mullerianosisdevelopmentally misplaced mullerian tissue for this parauterine uterus like mass. Suspected mullerian choristomas with only one mullerian tissue like this mass can be diagnosed only when three criteria are met; no evidence of pelvic endometriosis, no direct communication with endocervix, endometrium or endosalpinx and no history of surgery on reproductive organs. The mass in present case was directly attached to uterus and ipsilateral adnexa, thus reducing chance of mullerianosis as a cause.

## 4. Conclusion

In conclusion, the presented case of uterus like mass of broad ligament attached directly to normal uterus, without any associated congenital malformations, suggests subcoelomic mesenchyme transformation theory as most likely cause. Although rare, uterus like mass should be considered in differential diagnosis of dysmenorrhoea of uncertain etiology and clinically palpable pelvic mass.

## 5. Conflicts of interest

The authors have no conflicts of interest to disclose.

## References

- [1] Cozzutto C. Uterus like mass replacing ovary. Report of a new entity. Arch Pathol Lab Med 1981; 105:508-511
- [2] Pueblitz-Peredo S, Luevano-Flores E, RinconTaracena R, Ochoa-Carrillo FJ. Uterus-like mass of the ovary: endomyometriosis or congenital malformation? A case with a discussion of histogenesis. Arch Pathol Lab Med 1985;109:361–4.
- [3] Horie Y, & Kato M. (2000). Uterus-like mass of the small bowel mesentery. Pathology International Pathol Int, 50(1), 76-80.
- [4] Rohlfing MB, Kao KJ, Woodard BH. Endomyometriosis: possible association with leiomyomatosis disseminata and endometriosis. Arch Pathol Lab Med. 1981;105:556–557.
- [5] Na KY, Kim GY, Won KY, Kim, HS, Kim SW, Lee CH, & Cha, J. M. (2013). Extrapelvic Uterus-like Masses Presenting as Colonic Submucosal Tumor: A Case Study and Review of Literature. Korean Journal of Pathology, 47(2), 177–181.
- [6] Ahmed AA, Swan RW, Owen A, Kraus FT, Patric F. Uterus like mass arising in the broad ligament: a metaplasia or Mullerian duct anomaly? Int J Gynecol Pathol 1977; 16:279-281.
- [7] Takeda A, Imoto S, Mori M, Yamada J, Nakamura H. (2011). Uterus-like mass of ovarian ligament: Image diagnosis and management by laparoendoscopic single-site surgery. Journal of Obstetrics and Gynaecology Research, 37(12), 1895-1899.
- [8] Liang Y, Hao Q, Wu Y, Wu B. (2010). Uterus-like mass in the left broad ligament misdiagnosed as a malformation of the uterus: A case report of a rare condition and review of the literature. Fertility and Sterility, 93(4).
- [9] Matsuzaki S, Murakami T, Sato S, Moriya, T, Sasano H, & Yajima A. (2000). Endomyometriosis arising in the uterosacral ligament: A case report including a literature review and immunohistochemical analysis. Pathology International Pathol Int, 50(6), 493-496.
- [10] Nechi S, Znaidi N, Rammah S, M'Farej MK, Zermani R. (2013). Uterus-like mass of the broad ligament. International Journal of Gynecology & Obstetrics, 123(3), 249-250.

- [11] Eble JN, Tavassoli FA,Devilee P. Pathologyand genetics of tumours of the breast and female genital organs. International Agency for Research on Cancer, World Health Organization. Lyon, France: IARC, 2006. 228.
- [12] Moghadamfalahi M, Metzinger DS (2012). Multiple Extrauterine Adenomyomas Presenting in Upper Abdomen and Pelvis: A Case Report and Brief Review of the Literature. Case Reports in Obstetrics and Gynecology, 2012, 1-3.
- [13] Sadler TW. Langman's Medical Embryology. 6th ed. Baltimore, Md : Williams and Wilkins, 1990 : 270-83.
- [14] Rosai J.Uterus-like mass replacing ovary [letter]. Arch Pathol Lab Med, 1982, 106 : 364.
- [15] Lauchlan SC. The secondary Mullerian system. Obstet Gynecol Surv 1972;27:133–46.
- [16] Redman R, Wilkinson EJ, Massoll NA. Uterine-like mass with features of an extrauterine adenomyoma presenting 22 years after total abdominal hysterectomy-bilateral salpingo-oophorectomy: a case report and review of the literature. Arch Pathol Lab Med 2005;129:1041–3.
- [17] Scully RE. Smooth muscle differentiation in genital tract disorders. Arch Pathol Lab Med 1981; 105:505-507
- [18] Peterson CJ, Strickler JG, Gonzalez R, Dehner LP. Uterus-like mass of the small intestine. Heterotopia or monodermal teratoma? Am J Surg Pathol 1990;14: 390–4.
- [19] Batt RE, Pathogenesis of a parauterine uterus-like mass: developmentally misplaced müllerian tissue müllerianosis [letter]. Fertil Steril. 2010 Jul;94(2).

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