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### EXTRAUTERINE ADENOMYOMA: A REVIEW OF THE LITERATURE

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#### Abstract

Adenomyosis is the presence of endometrial glands and stroma within the myometrium. The focal and localized form of adenomyosis is known as adenomyoma. It is rarely located outside the uterus which is termed as extrauterine adenomyoma. We describe three cases of extrauterine adenomyomas which were located in pararectal space, round ligament and ovary. These cases were treated by laparoscopic excision and diagnosis was confirmed by histopathological examination. A review of the literature identified 34 cases of extrauterine adenomyomas. The most common locations were pararectal space, ovary and broad ligament. Other pelvic locations

included the round ligament, paraovarian, parametrial and pelvic wall. Extrapelvic adenomyomas were located in the liver, upper abdomen, inguinal scar, appendix and small bowel mesentery. The abdominopelvic pain was the most common clinical presentation of extrauterine adenomyoma. Various imaging modalities were used to identify extrauterine masses, but a definitive diagnosis could not be made preoperatively in any of the cases. Although rare, a possible diagnosis of adenomyoma must be contemplated while dealing with extrauterine masses. Surgical excision is the mainstay of treatment. Since malignancy has been reported in extrauterine adenomyomas, this possibility must be kept in mind while offering treatment.

**Keywords**: Extrauterine adenomyoma, pararectal adenomyoma, ovarian adenomyoma, round ligament adenomyoma

### Introduction

Adenomyosis is the presence of endometrial glands and stroma within the myometrium. Three different forms of adenomyosis have been identified such as focal, diffuse and cystic. The focal and localized form of adenomyosis is known as adenomyoma. Although a definite diagnosis can be established only with histopathology, with the advent of high resolution imaging techniques a provisional diagnosis can be made.

Transabdominal sonography (TAS) or transvaginal sonography (TVS) is commonly used as the initial imaging modality. TAS does not allow reliable diagnosis of adenomyosis or consistent differentiation from leiomyoma because of its limited spatial resolution. TVS is more accurate in diagnosing adenomyosis because of its better spatial resolution. Typical appearances of adenomyosis at TVS include poorly marginated hypoechoic and heterogeneous areas within the myometrium, myometrial

cysts, and a globular or enlarged uterus. Magnetic resonance imaging (MRI) is a highly accurate noninvasive modality for diagnosis of adenomyosis. Adenomyosis appears as either diffuse or focal thickening of the junctional zone forming an ill-defined area of low signal intensity. Moreover, MRI is useful in distinguishing adenomyosis from a leiomyoma in cases of enlarged uterus<sup>1</sup>.

Adenomyomas are rarely located at extrauterine sites such as ovary, broad ligament, ovarian ligament, pararectal and extrapelvic locations such as liver and abdominal wall. We report three cases of extrauterine adenomyomas managed by laparoscopic excision with a review of the literature. The aim of this review is to provide an insight into clinical presentation, diagnosis and management of extrauterine adenomyoma as it is a rare clinical entity.

#### Case 1

A 39-year-old female presented with complaints of heavy menstrual bleeding and mid cycle pain since five years. She also had difficulty in initiating micturition since two months. She had undergone a laparoscopic right ovarian cystectomy for endometriosis two years back. She had two normal vaginal deliveries and four miscarriages. Clinical examination revealed an enlarged retroverted uterus corresponding to 14 weeks gravid uterus size. Ultrasonography had a 10.3x7.1cm hyperechoic mass posterior to the uterus, the right ovary had a 4.6x4.3cm cyst with fine internal echoes, and the left ovary had a 2x1.9cm cyst with fine internal echoes. Both ovaries were seen close to the uterus. Laparoscopy had a normal size uterus which was retroflexed due to dense adhesions with rectosigmoid. Both ovaries had 5cm endometriotic cysts and were densely adherent to the rectosigmoid. A 10cm retroperitoneal deeply embedded mass was identified in the right pararectal space which was adherent to the uterus. It was exposed partially after isolation of the right ureter. The pararectal mass was dissected from the pelvic sidewall and the rectum after delineating the rectum. The patient underwent total laparoscopic hysterectomy, excision of the pararectal mass, right oophorectomy and left ovarian cystectomy.

All the specimens were sent for histopathological analysis. On cut section, the pararectal mass showed white whorled appearance with cystic spaces. Microscopic examination revealed numerous islands of endometrial glands with endometrial stroma amidst whorls and fascicles of smooth muscle cells with no significant mitosis consistent with adenomyoma. Bilateral ovarian cyst wall specimens were consistent with endometriotic cyst. The uterus had multiple leiomyomas with focal adenomyosis and both fallopian tubes showed no significant pathology. The postoperative period was uneventful, and the patient was discharged 24 hours after surgery. The patient was asymptomatic after a one month follow up.

#### Case 2

A 45 years old female presented with a history of dull aching right lower quadrant pain on and off since six months. She had two normal vaginal deliveries and had undergone a subtotal hysterectomy during her second delivery for severe post partum haemorrhage 12 years back. She had also undergone Laparoscopic left ovarian endometriotic cystectomy 20 years back and laparoscopic right adnexectomy with left salpingectomy for endometriosis four years back. Ultrasonography revealed a cervical stump of 3.5 x 2.5cm, left ovary had a multiloculated cyst of 6.5x3.8cm with fine internal echoes and right adnexa revealed a hyperechoic lesion of 3.3x2.8cm. Laparoscopy showed a multiloculated cyst containing chocolate material in the left ovary which was densely adherent to the rectosigmoid. A left oophorectomy was performed after adhesiolysis and isolation of the left ureter. The remnant right round ligament stump showed a 3cm firm mass which was excised and sent for histopathological analysis.

On cut section, round ligament specimen showed white firm areas with foci of haemorrhage. Microscopic examination showed dilated endometrial glands with stroma amidst hypertrophied whorls and fascicles of smooth muscle cells. Histopathological analysis of left ovary was consistent with endometriosis. The postoperative period was uneventful, and the patient was discharged 24 hours after surgery. The patient was asymptomatic after a one month follow up.

#### Case 3

A 37 years old nulligravida with a history of subfertility came with complaints of intermenstrual spotting and dysmenorrhea of one-year duration. She also complained of constipation since one year. She had undergone laparoscopic myomectomy five years back. On clinical examination, the uterus was enlarged corresponding to 12 weeks gravid uterus size with restricted mobility. Ultrasonography revealed a fundal hyperechoic mass of size 3.7x 3.5 cm and another hyperechoic mass in the left posterolateral wall of 5.9x 4.2 cm size. The posterior uterine wall was thickened suggestive of adenomyosis. The left ovary contained a cyst of 2.6x 1.9 cm with fine internal echoes, the right ovary was normal, and both ovaries were located close to the uterus. Laparoscopy revealed an enlarged uterus to 10 cm size possibly due to adenomyotic changes. The uterus was retroflexed due to dense rectosigmoid adhesions to the posterior uterine surface and both adnexae. The pouch of Douglas was completely obliterated. A 6cm mass was seen deeply embedded in the left pararectal space. The mass was dissected from the pelvic side wall and rectum with difficulty after isolating the ureter and delineating the rectum with a probe (Figure 1A). A 3cm well-defined, solid and encapsulated mass was seen in the right ovary (Figure 1B). As the consent for oophorectomy was not taken, excision of the mass was done. The left ovarian chocolate cyst wall was enucleated.

All the specimens were sent for histopathological analysis. Cut section of the pararectal and ovarian mass revealed whorled white firm areas. Multiple representative microscopic sections were analysed, these showed islands of endometriotic glands with stroma amidst whorls of benign smooth muscles with uniform cigar-shaped nuclei and no significant mitosis suggesting a diagnosis of adenomyoma (Figure 1C, 1D). The ovarian cyst wall specimen was consistent with an endometriotic cyst. The postoperative period was uneventful, and the patient was discharged 48 hours after surgery. The patient was asymptomatic after a one month follow up. She conceived in the second cycle of Intrauterine Insemination (IUI) after surgery.

#### **Materials and Methods**

We conducted an electronic-based search using databases PubMed and Google Scholar. The following keywords and their combinations were used: "extrauterine adenomyoma", "pelvic adenomyoma", "extrapelvic adenomyoma" and "uterus-like mass". 'Uterus-like mass' represents a type of adenomyoma with an organized arrangement of tissues characterized by a single central cavity lined by endometrium and surrounded by a thick wall of smooth muscle, resembling a normal uterus. Thus 'uterus-like mass' was also included in this review. The reference list of identified studies was searched manually for additional articles. The search was limited to studies in humans and those published in the English language. No limit or filter was used for the time period. A record of the following information was made manually such as patient's age, clinical presentation, size and location of extrauterine adenomyoma, diagnosis, management and follow up. Non English articles, articles with incomplete information and unavailable full text were excluded from the review.

### **Results**

Our search yielded 176 articles. Initially articles and abstracts of these articles were screened and reviewed. After reviewing the abstracts, 118 articles were excluded owing to duplication, non-English articles and citations unrelated to extrauterine adenomyomas and 58 articles (66 cases) of extrauterine adenomyoma were found. After further exclusion of articles with unavailable full text and articles with incomplete information about history, diagnosis and management, 30 publications (34 cases) of extrauterine adenomyoma were eligible for the review (Table no.1).

#### **Comment**

The first case of extrauterine adenomyoma was reported by Cozzutto et al. in 1981[2]. Extrauterine adenomyoma of both pelvic and extrapelvic locations have been described in the literature. Out of these 34 cases, 19 were pelvic, 8 were extrapelvic and 7 cases involved multiple sites (Table no. 2). The most

common locations of extrauterine adenomyoma were pararectal space [3-7], ovary [8-13] and broad ligament [1], 14-18]. Other pelvic locations include the round ligament [19], paraovarian [20], parametrial [20] and pelvic wall [21]. Our first case had a single extrauterine adenomyoma located in the right pararectal space. Six other cases with pararectal extrauterine adenomyoma have been described similar to our case [3-7]. Our second case presented with an extrauterine adenomyoma of right round ligament. Only one similar case with round ligament adenomyoma has been reported so far [19].

In our third case, two extrauterine adenomyomas were found. One was located in the right ovary and the other in the left pararectal space. Seven other cases involved multiple sites [1] [4] [6] [12] [22] like sigmoid colon, ovary, omentum and upper abdomen. Ki Yong et al [22] reported multiple adenomyomas in the caecum and descending colon which were diagnosed after colonoscopy. Carinelli et al [4] was the first to report 2 cases of multiple extrauterine adenomyoma. He described a case with pararectal and ovarian adenomyoma similar to our case.

Extrapelvic adenomyomas have been described in 8 cases which were located in the liver [23] [24] [25], upper abdomen [26] [27], inguinal scar [28], appendix [29] and small bowel mesentery [30].

Various theories have been proposed for explaining the extrauterine location of adenomyoma. Rosai et al [31] suggested the theory of defective mullerian duct fusion to explain the extrauterine location of adenomyoma. Partial or complete fusion defects of bilateral mullerian ducts may result in uterine duplication or atresia. A unicornuate uterus with a rudimentary horn may develop, and this rudimentary horn may detach and implant elsewhere, resulting in extrauterine adenomyoma. This theory explains cases of extrauterine adenomyoma accompanying congenital urogenital abnormalities like renal agenesis, double excretory system and anomalies of the genital tract[21] [31-33]. Batt et al [34] proposed the theory of mullerianosis which states that a heterotropic organoid structure of embryonic origin composed of mullerian cell rests may get incorporated into normal organs at the time of organogenesis and result in

this condition. They proposed three criteria such as absence of pelvic endometriosis, no communication of adenomyoma with endocervix, endometrium and endo-salpinx and absence of surgeries on reproductive organs. This theory provides an explanation for unusual extrapelvic locations of adenomyomas. The theory of mullerianosis cannot be applied to our cases due to the presence of endometriosis and previous surgeries. Cozzuto et al [2] proposed the theory of smooth muscle cell metaplasia. This theory suggested that an already existing focus of endometriosis undergoes metaplasia into smooth muscle, explaining the pathogenesis of adenomyoma [2]. Histopathological finding of predominant smooth muscle with scattered endometrial glands in our cases is not consistent with the theory of smooth muscle metaplasia.

Redman et al [3] proposed the theory of sub-coelomic mesenchymal metaplasia. Subcoelomic mesenchymal layer lies below the mesothelial surface of the peritoneum and envelops the uterus, ovaries and tubes. This layer contains multipotent cells which have the capacity to differentiate into the endometrial stroma, decidua and smooth muscles under hormonal stimulation. The histopathological findings in their case had predominant smooth muscle with foci of endometrium. Similar histopathological findings were observed in our case series, supporting the sub-coelomic mesenchymal metaplasia theory.

Abdominopelvic pain is the most common clinical presentation of extrauterine adenomyoma. Few patients had additional symptoms like abnormal bleeding [18] [19] and infertility [10] [21]. Six cases had a history of pelvic endometriosis [4] [21] [22] [24] [26] [28]. Seventeen cases had a history of gynaecological surgery [3-6] [12] [14] [16] [20-26] [28] [29] such as hysterectomy, myomectomy or ovarian cystectomy. Our first case presented with heavy menstrual bleeding, mid-cycle pain and a history of right ovarian endometriotic cystectomy. Our second case presented with right lower quadrant pain and history of hysterectomy, left ovarian cystectomy, right adnexectomy and left salpingectomy. The third case presented with subfertility, dysmenorrhea, intermenstrual spotting and a history of myomectomy.

Although no definitive predisposing factors have been listed, Redman et al [3] reported a case of extrauterine adenomyoma after hysterectomy and prolonged estrogen therapy supporting the estrogen responsiveness of these lesions. Bayar et al [10] reported a case of extrauterine adenomyoma after gonadotropin treatment for infertility. None of our patients had a history of any prolonged hormonal therapy.

The age of the patients in these cases ranged from 17-70 years, and the diameter of the lesions ranged from 0.8 -20 cm. Our first patient was 39 years old with a 10cm lesion whereas the second case presented at 45 years of age with a 3cm lesion and the third case presented at 37 years of age with lesions of 6cm and 3cm.

Diagnosis of extrauterine adenomyoma is a clinical challenge as none of the cases was diagnosed preoperatively. Imaging modalities used in these 34 cases were ultrasonography (USG), computerized tomography (CT) and magnetic resonance imaging (MRI). Intravenous pyelography (IVP) was done in few cases to exclude co-existing renal abnormalities. Ultrasonography was the most common imaging modality used. CT was used in 17 cases [3] [4] [6-8] [11] [12] [14] [18] [19] [22-29], MRI was used in 8 cases [1] [4] [5] [11] [12] [16] [28] [35] and IVP was used in 6 cases [3] [8] [9] [11] [13] [21]. Ultrasonography alone was used as an imaging modality in all three of our cases. There is a paucity of literature on the radiological appearance of extrauterine adenomyoma. A definitive diagnosis of extrauterine adenomyoma in all 34 cases was established only postoperatively after confirmation on histopathology similar to our cases.

On histopathology, adenomyoma shows endometrial glands, endometrial stroma with smooth muscle cells. Uterus-like masses are defined as extrauterine organoid masses that are characterized by a single central cavity lined by endometrium and surrounded by a thick wall of smooth muscle, resembling a normal uterus and most likely representing a particular form of extrauterine adenomyoma [23].

Microscopic appearance of extrauterine adenomyomas should be carefully analysed to confirm the diagnosis as they can be confused with endometriosis with smooth muscle component and leiomyomas with endometriosis. Smooth cell metaplasia is focal, unlike adenomyoma which shows dominant smooth muscle component and is more circumscribed both on gross and microscopic appearance. In leiomyoma with endometriosis, the endometriotic cyst is usually peripheral and separate from the smooth muscle component<sup>23</sup>. The lesions in our cases had endometrial gland dispersed within smooth muscle component consistent with adenomyoma.

Surgical management was done in all 34 cases. None of the studies reported any major intraoperative or postoperative complications except for one study by Carinelli et al [4] where in one case, partial colectomy was done along with excision of adenomyomas due to adhesions with sigmoid colon and in the second case sigmoid perforation was detected seven days later, and partial colectomy with a colostomy was done. Our cases were managed by laparoscopic excision without any complications.

Few cases provided insight into follow up of these patients ranging from two months to 10 years and recurrence has also been reported. All our cases were recently operated and are asymptomatic after a one month follow up. Carinelli et al [4] reported the recurrence after surgical excision of a sigmoid nodule with partial resection of right paraovarian nodule and omentectomy for multiple site adenomyomas. One year later laparoscopy, revealed multiple nodules over the peritoneum, the intestine and the left fallopian tube. Excision of the nodules with left salpingectomy followed by GnRH agonist therapy was offered. After a ten year follow up, there was no relapse. Carvalho et al [12] reported recurrence after 15 months. The patient received goserelin followed by anastrozole for recurrence. Medical treatment was not given to any of our patients.

Although most of these cases were benign, a few cases were associated with malignancy. Torres et al [18] reported clear cell adenocarcinoma in a case of broad ligament adenomyoma. Ulm et al [19] reported focal endometrioid adenocarcinoma in extrauterine adenomyoma (round ligament) with concurrent stage 1 uterine endometrioid adenocarcinoma. Rahilly et al [13] reported a concurrent occurrence of ovarian adenomyoma with ovarian endometrioid carcinoma and uterine endometrial cancer. So, clinician should be aware of the malignancy potential while dealing with extrauterine masses. An endometrial biopsy may be performed to detect any coexisting uterine malignancy especially in cases with abnormal uterine bleeding.

### **Strengths and limitations**

The strengths of this review include its comprehensive search strategy and methodological design. No other review summarizes information about common locations, clinical presentation, diagnosis, management and malignant transformation of extrauterine adenomyomas. The limitation of this review is that only 11 out of the 34 cases provide information on follow up. 12 cases have not provided details of surgical procedure performed. There is paucity of data on radiological appearance of this rare pathology.

### Conclusion

Although rare, a possible diagnosis of adenomyoma must be contemplated while dealing with extrauterine masses. A preoperative diagnosis is difficult to establish, as confirmation can only be made after histopathological analysis. Surgical excision is the mainstay of treatment. Since malignancy has been reported in extrauterine adenomyomas, this possibility must be kept in mind while offering treatment.

#### Author's role

PPG was the surgeon. PPG conceived, designed and wrote the review. PPG and all the other authors contributed together in manuscript drafting and critical discussion.

### Role of the funding source

The funding needs of this review were met by the institutional funds for research granted by Paul's

hospital, Kochi, Kerala, India. No external funding was received.

#### **Ethics Committee Approval**

Written informed consent was obtained from the patients for publication of the case report and accompanying images. The retrospective observational nature of the study did not necessitate the local institutional ethics committee approval.

#### **Conflict of interest**

The authors declare that they have no conflict of interest

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#### References

- 1. Bulut AS, Sipahi TU. Abscessed uterine and extrauterine adenomyomas with uterus-like features in a 56-year-old woman. Case Rep Obstet Gyneco. 2013; 2013: 238156.
- 2. Cozzutto C. Uterus-like mass replacing ovary: a report of a new entity. Arch Pathol Lab Med. 1981; 105:508 -511.
- 3. 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. Arch Pathol Lab Med. 2005; 129:1041–1043.
- 4. S. Carinelli, F. Motta, G. Frontino, E. Restelli, and L. Fedele, "Multiple extrauterine adenomyomas and uterus-like masses: case reports and review of the literature" Fertil Steril. 2009;91: 1956; e1959-1911.
- 5. Moon SK, Park SJ, Lim JW, et al. Interval changes of an extrauterine adenomyoma on magnetic resonance imaging. Am J Obstet Gynecol. 2012; 206(6): e3-e5.
- 6. Moghadamfalahi M, Metzinger DS. Multiple extrauterine adenomyomas presenting in upper abdomen and pelvis: a case report and brief review of the literature. Case Rep Obstet Gyneco 2012; 2012:565901.
- 7. Kim MJ, Seok SH. Uterus-Like Mass with Features of an Extrauterine Adenomyoma: A Case Report and Literature Review. Korean J Pathol. 2007 Oct; 41(5): 347-351.
- 8. Choudhrie L, Mahajan NN, Thomas A, Kale J, Mahajan K. Ovarian ligament adenomyoma: a case report. Acta Chir Belg. 2007; 107:84–5.
- 9. Sisodia SM, Khan WA, Goel A: Ovarian ligament adenomyoma: report of a rare entity with review of the literature. J Obstet Gynaecol Res. 2012;38: 724–728.
- 10. Bayar U, Demirtas E, Usubutun A, Basaran M, Esinler I, Yarali H: Ovarian adenomyoma following gonadotrophin treatment for infertility. Reprod Biomed Online. 2006;13: 676–679.
- 11. Takeda A, Imoto S, Mori M, Yamada J, Nakamura H: Uterus-like mass of ovarian ligament: image diagnosis and management by laparoendoscopic single-site surgery. J Obstet Gynaecol Res. 2011;37: 1895–1899.
- 12. Carvalho FM et al. Leiomyomatosis peritonealis disseminata associated with endometriosis and multiple uterus-like mass: report of two cases. Clin Med Insights Case Rep. 2012; 5:63–68.
- 13. Rahilly MA, Al-Nafussi A. Uterus-like mass of the ovary associated with endometrioid carcinoma. Histopathology. 1991; 18: 549-551.
- 14. Liang YJ, Hao Q, Wu YZ, Wu B: 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. Fertil Steril. 2010, 93(1347):e1313–e1346.
- 15. He J, Xu J, Zhou H-Y. Uterus-like mass: A very rare and elusive entity a case report. Medicine. 2016; 95(39):e4961.

- 16. Menn KA, Qin L, Fedoriw GD, et al. Uterus-like mass: MRI appearance of a very rare entity. J Magn Reson Imaging. 2007; 26:162–4.
- 17. Goswami M. A rare case of giant uterus-like mass arising from broad ligament: a case report with review of histogenesis. Indian J Case Reports. 2017; 3(1): 30-33.
- 18. Torres D, Parker L, Moghadamfalahi M, Sanders MA, Metzinger DS. Clear cell adenocarcinoma arising in an adenomyoma of the broad ligament. Int J SurgPathol. 2015; 23(2):140-3.
- 19. Ulm MA, Robins DB, Thorpe Jr EM, Reed ME. Endometrioid adenocarcinoma in an extrauterine adenomyoma. Obstet Gynecol. 2014; 124(2 Pt 2 Suppl 1):445-8.
- 20. Stewart CJ, Leung YC, Mathew R, Mccartney AL. Extrauterine adenomyoma with atypical (symplastic) smooth muscle cells: a report of 2 cases. Int J Gynecol Pathol. 2009; 28:23-8.
- 21. Kaufman Y, Lam A.The pelvic uterus-like mass— a primary or secondary mullerian system anomaly. J Minim Invasive Gynecol. 2008; 15:494–7.
- 22. Na KY, Kim GY, Won KY, et al. Extrapelvic uterus-like masses presenting as colonic submucosal tumor: a case study and review of literature. Korean J Pathol. 2013;47: 177–81.
- 23. Huanwen et al.: Extrauterine adenomyoma of the liver with a focally cellular smooth muscle component occurring in a patient with a history of myomectomy: case report and review of the literature. Diagn Pathol. 2013;8:131.
- 24. Tandon, N., Showalter, J., Sultana, S., Zhao, B., & Zhang, S. Extrauterine adenomyoma of the liver in a 50 year old female with pelvic endometriosis. Ann Clin Lab Sci. 2017;47(2):208-212.
- 25. Sopha SC, Rosado FG, Smith JJ, et al. Hepatic uterus-like mass misdiagnosed as hepatic abscess. Int J Surg Pathol. 2015; 23:134–9.
- 26. Khurana A, Mehta A, Sardana M. Extrauterine adenomyoma with uterus like features: A rare entity presenting 17 years post hysterectomy.Indian J Pathol Microbiol.2011; 54:572-3.
- 27. Sampaio R, Garcia JP, Macedo CS, Vizcaíno JR. A 22nd case report of extrauterine adenomyoma of the abdominal wall. Case Rep Clin Patho. 2017; 4(2):11.
- 28. Seki A, Maeshima A, Nakaqawa H, Shiraishi J, Murata Y, Arai H, et al. A subserosal uterus-like mass presenting after a sliding hernia of the ovary and endometriosis: a rare entity with a discussion of histogenesis. Fertil Steril. 2011; 95:1788.e15–19.

- 29. Kim HC, Yang DM, Kim SW, et al. Uterus-like mass involving the appendix: US and CT findings. J Clin Ultrasound 2012; 40:518–21.
- 30. Horie Y, Kato M. Uterus-like mass of the small bowel mesentery. Pathol Int.2000; 50:76-80.
- 31. Rosai J. Uterus-like mass replacing ovary. Arch Pathol Lab Med.1982; 106:364-5.
- 32. Pueblitz-Peredo S, Luévano-Flores E, Rincón-Taracena R, Ochoa- Carrillo FJ. Uterus-like mass of the ovary: endomyometiosis or congenital malformation? A case with a discussion of histogenesis. Arch Pathol Lab Med. 1985; 109: 361–364.
- 33. Shutter J. Uterus-like ovarian mass presenting near menarche. Int J Gynecol Pathol. 2005; 24: 382-384.
- 34. Batt RE. Pathogenesis of a parauterine uterus-like mass: developmentally misplaced müllerian tissue—müllerianosis. Fertil Steril. 2010, 94:e45.
- 35. Ko J.K.Y., Cheung V.Y.T. Uterus-like mass: Issues on pathogenesis. J Minim Invasive Gynecol. 2015; 22(7):1133-1134.

### Figure/Table Caption List

Figure 1A- Laparoscopic view of pararectal adenomyoma- Third case

Figure 1B- Laparoscopic view of right ovarian adenomyoma- Third case

Histopathological view of ovarian adenomyoma- Figure 1C- Low power, Figure 1D - High Power - Third case

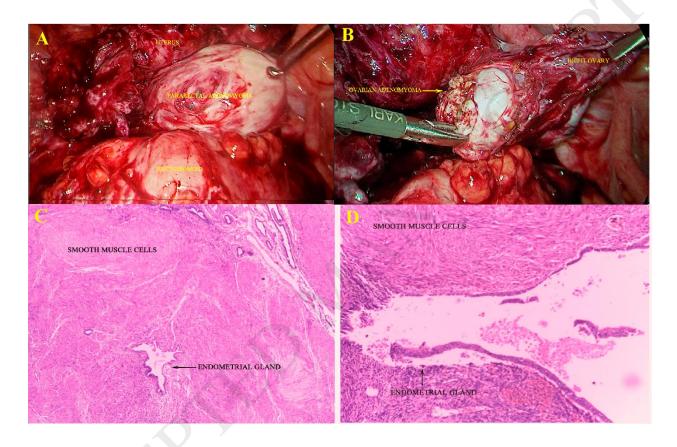


Table No.1- Flowchart of studies included in the review

### Table no.1

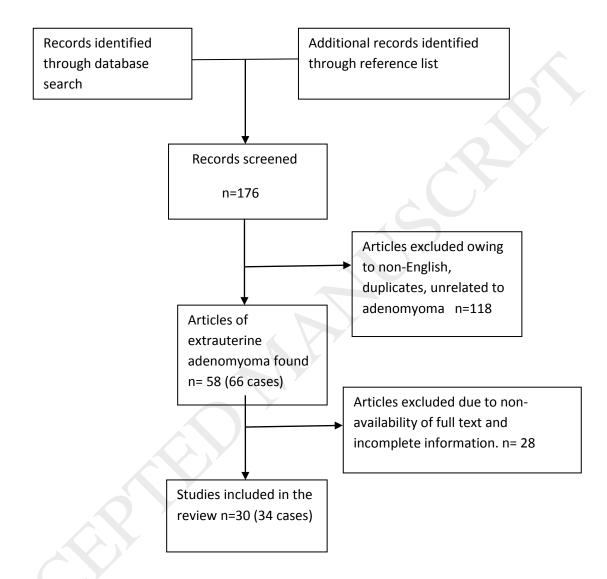


Table No. 2 - Description of extrauterine adenomyomas

### Table no 2

Sr.n	Study	Size and location	Ag	Past	Presenting	Imaging	Surgical
o	(Year)		е	history	complaints	modaliti	intervention
						es	/
1	Rahilly	5cm, right ovary	38		RIF and pelvic	IVP	TAH with BSO
	et al				Pain		
	(1991) <sup>13</sup>				15		
2	Horie et	14x11cm, small	59		Mass lower		Surgical excision
	al	bowel mesentery			abdomen		
	(1999) <sup>30</sup>				>		
3	Redman	5cm, Pararectal	50	TAH with	Dysuria and	CT,	Excision + left
	et al			BSO +	suprapubic,	USG,	ureteric stenting
	(2005)3			HRT	pelvic pain	IVP	
4	Bayar et	7.5cm, left ovary	38	Gonadotro	Infertility and	USG	Laparoscopic
	al			pin	pelvic pain		excision
	(2006)10			treatment			
5	Choudhr	0.8cm, Left ovarian	57		Lump lower	USG,	TAH with BSO
	ie et al	ligament			abdomen &	IVU	
	(2007)8				pelvic pain		
6	Mi Jin	10.5X9.5cm,	42		Lower	СТ	Surgical excision
	Kim et al	pararectal			abdominal pain		
	(2007) <sup>7</sup>						
7	Menn et	6x4cm, right broad	37	Myomecto	Right quadrant	USG,	TAH

	al	ligament		my and	pain and	MRI	
	(2007) <sup>16</sup>			polypecto	intermenstrual		
				my	spotting		
8	Kaufma	7x5cm, right pelvic	39	Subfertility,	Dysmenorrhea,	USG,	Laparoscopic
	n et al	wall, absent right		PID	pain,	СТ	excision
	(2008) <sup>21</sup>	kidney, absent			menorrhagia,		
	Case 1	right ureter					
9	Kaufma	10.5x 9cm, right	57	RSO, TAH	RIF pain,	USG,CT	Laparoscopic
	n et al	pelvic wall		+ LSO for	suprapubic	,IVP	excision
	(2008) <sup>21</sup>			Endometri	pain and		
	Case 2			osis +	backache		
				HRT			
10	Stewart	6x4.5cm left	40	TAH for	Left iliac fossa	USG	Laparoscopic
	et al	paraovarian mass		DUB	pain		excision
	(2008) <sup>20</sup>						
	Case 1						
11	Stewart	6.3x4cm right	65	PID,	Pelvic mass	USG	Hysterectomy
	et al	parametrial mass		breast			with BSO with
	(2008) <sup>20</sup>	$\langle \rangle$		cancer			mass excision
	Case 2	1					
12	Carinelli	10cm sigmoid,	46	Myomecto	Abdominal pain	USG,CT	Excision,
	et al	6cm pelvic, 4cm		my	and		Hysterectomy
	(2009)4	ileal, 1cm paraileal			constipation		with partial
	Case 1	and paravesical					colectomy and
							Meckel

							diverticulum
							resection +
							GnRH agonist
13	Carinelli	3cm sigmoid,	39	Left	Dysmenorrhea,	USG,CT	Laparoscopic
	et al	3.5cm right ovary		ovariectom	Chronic	,MRI	excision. Partial
	(2009)4			y for	abdominopelvi		colectomy with
	Case 2			endometri	c pain		colostomy 7
				osis			days later +
							GnRH agonist
							for relapse
14	Liang et	4cm, left broad	17	Mesosalpi	Dysmenorrhea	USG,CT	Excision
	al	ligament		nx	and pelvic pain		
	(2010)14			cystectom			
				у			
15	Sisodia	5.5x5.3cm, Right	56		Dysuria, lower	USG,IV	TAH with BSO
	et al	ovarian ligament			abdominal	Р	
	(2011) <sup>9</sup>				pain, bleeding		
					per vaginum		
16	Moon et	7x6cm, pararectal	41	SCH and		USG,M	Excision and
4	al	) _		right		RI	LSO
	(2011)5			salpingect			
	7			omy			
7							
17	Seki et	3.8 x 2cm, left	44	Left	Abdominal pain	USG,M	Surgical excision
	al(2011)	inguinal region		oophorect		RI	

	28			omy,			
				Endometri			
				osis			
				0515			
18	Takeda	3.8x3.7cm, left	39		Pain lower	CT,	Laparoscopic
	et al	ovarian ligament			abdomen	MRI,IVP	excision
	(2011) <sup>11</sup>						
19	Moghad	6cm pararectal,	39	SCH,	Abdominal pain	СТ	Surgical Excision
	amfalahi	7.5cm upper		Cervical	and bleeding		
	et al	abdomen		myomecto	per rectum		
	(2012) <sup>6</sup>			my,			
				endometri			
				osis			
20	Carvalh	Four mm to 50mm	32	Hyotorogo		LISC	Excision +
20	Carvain	Few mm to 50mm,	32	Hysterosc		USG,	Excision +
	o et al	pelvic and		opic		CT,MRI	Goserelin +
	(2012)12	abdominal		myomecto			Anastrazole
	Case 1	peritoneum and		my			
		omentum, left					
		ovary					
		$\lambda$					
21	Carvalh	Few mm to 20mm,	41		Dysmenorrhea		LSO with partial
	o et al	pelvic and			and pelvic		excision of
	(2012)12	abdominal			pain, proctalgia		nodules +
	Case 2	peritoneum and					Medroxy
		omentum					progesterone
							acetate
22	Kim et al	2x1.5cm, appendix	46	Supracervi	Right lower	USG,	Surgical excision

	(2012)29			cal	quadrant pain	СТ	
				hysterecto			
				my			
23	Huanwe	3.6x2.6cm, liver	29	Myomecto	Back pain	USG,CT	Surgical
23		3.0x2.0cm, liver	29		васк раш	030,01	
	n et al (			my			resection
	2013) <sup>23</sup>						
24	Bulut et	5-10cm, Bilateral	56		Menorrhagia	USG,M	TAH with BSO
	al	broad ligament			and pelvic pain	RI	and excision of
	(2013) <sup>1</sup>	with pus, ectopic					intraligamentary
		adrenal tissue					masses
25	Ki Yong	Caecum,	39	Total	Right lower	USG,CT	Colonoscopic +
	Na et al	descending colon		hysterecto	quadrant pain		Laparoscopic
	(2013) <sup>22</sup>	and mesocolon		my with			resection
				LSO, RSO			
				for			
				endometri			
				osis			
26	Ulm et al	3cm, left round	49		Metromenorrhh	СТ	TAH with BSO
	(2014) <sup>19</sup>	ligament			agia		and lymph node
							dissection
27	Torres	4cm, right broad	58		Post	USG,CT	Total Robotic
	et al	ligament			menopausal	355,51	
		ilgani <del>e</del> ni					hysterectomy
	(2015) <sup>18</sup>				bleeding		with bilateral
							salpingo-
							oophorectomy

28	Sopha	1.4cm, Liver	47	RSO for	Right quadrant	СТ	Laparoscopic
	et al			teratoma,	and back pain		excision biopsy
	(2015) <sup>25</sup>			SCH+			
				HRT			
29	Jennifer	4cm, right adnexa	64		Recurrent thigh	MRI	Laparoscopic
	et al				sarcoma		BSO
	(2015)35						
30	Jian He	7x4.6cm, Left	43		Acute lower	USG	Surgical excision
	et al	broad ligament			abdominal pain		
	(2016) <sup>15</sup>				and		
					hypomenorrhe		
					а		
31	Khurana	13x9cm,	47	Subtotal	Bleeding per	СТ	Surgical excision
	et al	Abdominopelvic		Hysterecto	vaginum		
	(2017) <sup>26</sup>			my for			
				fibroids.			
		Y		Bilateral			
				oophorect			
				omy for			
		/ *		endometri			
				osis			

32	Tandon	6x4.5cm, Liver	50	Laparosco	Lower	СТ	Surgical
	et al			pic	abdominal pain		resection
	(2017) <sup>24</sup>			hysterecto			
				my with			
				unilateral			
				salpingect			<i>y</i>
				omy			
33	Sampaio	5cm, abdominal	70	Melanoma	Backache	СТ	USG guided
	et al	wall					core biopsy
	(2017) <sup>27</sup>						
34	Goswam	20x8cm, right	46	7	Swelling and	USG,	TAH + BSO
	i et al	broad ligament			pain abdomen	СТ	
	(2017) <sup>17</sup>			Vr.			
35	Present	10cm, pararectal	39	Laparosco	Heavy	USG	TLH with right
	study			pic right	menstrual		oophorectomy,
	Case 1			ovarian	bleeding, mid-		left ovarian
				cystectom	cycle pain and		cystectomy and
				y 2 years	difficulty in		excision of
				back	initiating		pararectal mass
		1			micturition		

36	Present	3cm, right round	45	Laparosco	Right lower	USG	Laparoscopic left
	study	ligament		pic left	quadrant pain		oophorectomy
	Case2			ovarian			with excision of
				cystectom			round ligament
				y 20years			mass
				back, SCH			
				12yrs back			
				and			
				laparoscop	15		
				ic RSO			
				and left			
				salpingect	>		
				omy 4			
				years back			
37	Present	6cm pararectal	37	Laparosco	Subfertility,	USG	Laparoscopic
	study	mass, 3 cm		pic	intermenstrual		excision with left
	Case 3	ovarian mass		myomecto	spotting,		ovarian
				my 5 years	dysmenorrhea,		cystectomy
				back	constipation		

RIF- Right iliac fossa, TAH with BSO- Total abdominal hysterectomy with bilateral salpingooophorectomy, HRT-Hormone replacement therapy, CT-Computerized tomography, USGUltrasonography, IVP-Intravenous pyelography, MRI-Magnetic resonance imaging, IVUIntravenous urography, TLH with BSO-Total laparoscopic hysterectomy with bilateral salpingooophorectomy, PID-Pelvic inflammatory disease, LSO-Left salpingo-oophorectomy, RSO- Right salpingo-oophorectomy, DUB-Dysfunctional uterine bleeding, GnRH- Gonadotropin releasing hormone, SCH-Supracervical hysterectomy