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Extrauterine Adenomyomas: A Rare Case Report in a 41 year old female

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Abstract- Smooth muscle cells, endometrial glands, and stroma are the main components of benign tumours called adenomyomas. Extrauterine locations are relatively uncommon and have an unidentified aetiology. It might be difficult to make a preoperative diagnosis, and a pathologic examination is required to confirm the diagnosis. Extrauterine adenomyomas are quite rare. This case report describes a rare instance of a 41-year-old lady who underwent a hysterectomy six months prior due to multiple fibroids and later post operatively developed a painless non-reducible swelling in her right inguinal area without any association with fever, vomiting or any other concerning symptoms. The lump was surgically removed as part of her treatment with herniorrhaphy. The diagnosis was confirmed after histological examination.

Keywords: Extrauterine adenomyoma, Right inguinal hernia, Adenomyoma

INTRODUCTION

Extrauterine adenomyomas are incredibly uncommon [1]. It is a benign tumour lined by thick-walled smooth muscles and made up of endometrial glands and stroma [2]. The ovaries are considered to be the most typical location for extrauterine adenomyoma. The aetiology of extrauterine adenomyoma is still unknown. Nonetheless, there is some evidence that these tumorous abnormalities are caused by hormonal influences, embryologic developmental errors, and mechanical/iatrogenic mechanisms [3, 4]. Establishing a preoperative diagnosis for extrauterine adenomyoma is difficult. Surgical excision followed by histological evaluation offers a conclusive diagnosis and is typically curative in cases of benign pathology [1, 2].

CASE REPORT

A 41-year-old multiparous woman (P2L2A0) presented with a single painless non-reducible mass in the right inguinal area since 2 months which has gradually increased in size and is not associated with fever, nausea, vomiting or any other concerns .Her gynecologic history included two normal vaginal deliveries. Due to multiple uterine fibroids, a total hysterectomy (without a salpingo-oophorectomy) was performed 6 months back. The entire uterus was removed without any difficulties and post operative course was uneventful. No history of any ocp intake.

On clinical examination, a single oval non reducible swelling of 3.0 x 2.0 cm was present in the right inguinal region. Cough impulse was absent. Skin surrounding and over the swelling was normal. Lower Midline scar mark was present.

There was no local rise in temperature, no tenderness. Swelling was uniform, firm to hard in consistency with well defined margins. Swelling was mobile and free from skin.

An ultrasound whole abdomen was performed which showed a defect measuring 13mm through which omental fat was seen herniating with internal echoes, thickening and entrapment along with soft tissue mass as contents. All relevant blood investigations were done.

Thus, based on these findings patient was planned for Right Inguinal Herniorraphy. Intraoperatively there was an Indirect Hernial sac along with omentum and soft tissue mass as content. Omentum was reduced and mass excised. Cut surface was: Grey-white, nodular, firm(Figure 1). On microscopy—sections showed lobulated mass consisting of endometrial tissues surrounded by fascicles of smooth muscles(Figure 2). Features were consistent with Adenomyoma. The patient was discharged within 24 h after surgery.



FIGURE 1

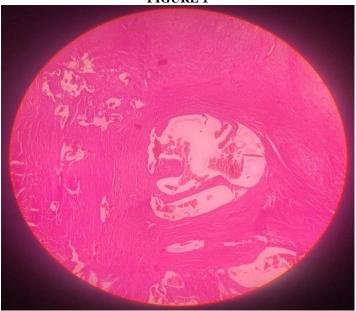


FIGURE 2

DISCUSSION

Extrauterine adenomyomas are quite rare. The recent literature has described only few cases. The pararectal space, ovaries, and wide ligament were the sites of the majority of cases of pelvic adenomyomas [1]. Adenomyomas that are found outside of the pelvis have also been documented, for example in the caecum, descending colon, and mesocolon [2]. There have also been reports of extrauterine adenomyomas in the upper abdomen [5]. But there is no existing report/literature suggesting adenomyomas as content of inguinal hernial sac.

Due to its rarity, the pathogenesis of extrauterine growth of adenomyomatous tissue is still poorly known. Nonetheless, a number of ideas contend that multiple factors contribute to the emergence of extrauterine adenomyomas. A faulty Mullerian duct fusion was the explanation, Rosai et al. [6] offered to account for the extrauterine location of adenomyomas. In a few instances, the association of adenomyomas with congenital malformations, such as renal agenesis, a double excretory system, and anomalies of the lower genital tract [7], lends weight to this notion. For a theory of smooth muscle cell metaplasia, Cozzuto et al. also proposed that the pathophysiology of adenomyoma is explained by the metaplasia of an existing foci of endometriosis into smooth muscle [3].

The sub-coelomic mesenchymal metaplasia idea was developed by Redman et al that under the influence of hormones, the multipotent cells that make up this layer, which is located underneath the mesothelial surface of the peritoneum, can specialise and multiply to form the endometrial stroma, smooth muscles, and even decidua [4]. The Mullerianosis theory, the final theory of embryological genesis, was put forth by Batt et al. It states that a heterotropic organoid structure of embryonic origin consisting of Mullerian cell resting may get incorporated into regular organs at the time of organogenesis. This hypothesis offers an explanation

for unique extrapelvic placements of adenomyoma [8]. The spread of cells during a myomectomy or (morcellated) hysterectomy is another frequently discussed process [9].

Abdominal pain, which is frequently accompanied by menstruation-related symptoms such intermenstrual spotting, menorrhagia, dysmenorrhea, or metromenorrhagia, is the most typical presenting symptom of extrauterine adenomyomas [1, 2]. A history of gynecologic surgery, such as a hysterectomy or myomectomy, may be pertinent as it was there in our case [1]. Unfortunately, no clear-cut risk factors for the development of extrauterine adenomyomas have been identified.

A well-defined lesion with benign endometrial gland foci and endometrial stroma encircled by hyperelastic smooth muscle bundles is what the histopathology looks like. In this instance also lobulated mass consisting of endometrial tissues surrounded by fascicles of smooth muscles was found.

A woman in this intriguing and unusual instance had an extrauterine adenomyoma in her inguinal region as content of hernial sac six months after having a surgical hysterectomy. The preoperative diagnosis was difficult because of its location and presentation; the diagnosis was confirmed through histological examination after surgical excision. Surgery is still the preferred form of treatment for adenomyoma.

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