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CASE STUDY

A RARE OCCURRENCE OF BILATERAL PRIMARY TUBAL ADENOMYOMA IN A CASE OF PRIMARY INFERTILITY

***Sandhya Deora, Rahul Manchanda and Samina Ashraf**

Department of Gynecology, Pushpawati Singhania Research Institute, New Delhi, India

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ABSTRACT

Adenomyoma of the fallopian tube is a rare entity. We report a rare case of adenomyoma in both the fallopian tubes in a 30-year-old woman, who presented with primary infertility and chronic pelvic pain. Patient underwent laparoscopy, and on gross examination, there were well-circumscribed and well-encapsulated tumours present in both fallopian tubes.Postoperative histopathology showed that foci of endometriosis composed of endometrial glands dispersed in endometrial stroma, were noted within the muscular wall of fallopian tubes suggestive of fallopian tube adenomyoma on both sides.

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INTRODUCTION

Uterine adenomyomas are benign tumors composed of smooth muscles, endometrial glands, and endometrial stroma. They are distinguished from adenomyosis by their sharp demarcation from the surrounding normal tissues, and from leiomyomas by the presence of intrinsic endometrial glandular and stromal elements. However, extrauterine adenomyomas are rare (Choudhrie *et al*., 2007; Stewart *et al*., 2009; Sisodia *et al*., 2012). Rare are those occurring in the fallopian tube (Etoh *et al*., 2012; Aki Miyasaka *et al*., ?: Mardi and Gupta, 2014) and even more rare are those found bilaterally. We report a case of extrauterine adenomyoma of both the fallopian tubesfound in a 30-year-old female on laparoscopy and confirmed by histopathology.

Case report

The patient was a 30-year-old female who presented with primary infertility and chronic pelvic pain with history of two previous laparoscopies and one IVF failure. Previous laparoscopies and ultrasound was suggestive bilateral tubal masses with no conclusive diagnosis. Laparoscopy was done, on which bilateral tubal masses were found, right side of about 3*2 cm, left side of about 5*3 cm containing dark coloured blood.

Corresponding author: **Sandhya Deora,**
Department of Gynecology, Pushpawati Singhania Research Institute, New Delhi, India.

Differential diagnosis included adenomyoma of the fallopian tube and hematosalpinx. On microscopy, foci of endometriosis composed of endometrial glands dispersed in endometrial stroma, were noted within the muscular wall of fallopian tubes suggestive of fallopian tube adenomyoma on both sides.

DISCUSSION

Adenomyomas are circumscribed tumorlike masses most often involving the uterus and consisting of endometrioid glands, stroma, and smooth muscle tissue. They are uncommon in extrauterine sites and in this situation it may be unclear whether such lesions represent foci of endometriosis with marked smooth muscle hyperplasia/metaplasia, uteruslike mass lesions, or leiomyomas with entrapped endometriotic glandular and stromal elements. Extra-uterine adenomyomas may arise from broad ligament, fallopian tube or ovary, even one case in liver has also been reported (Wu Huanwen *et al*., ?). Though, there are a few recorded cases of fibroma and fibromyoma of the fallopian tube in the literature, adenomyoma arising from the fallopian tube is extremely rare (Etoh *et al*., 2012; Aki Miyasaka *et al*., ?: Mardi and Gupta, 2014). So far two theories are proposed to explain the etiology of the extrauterine adenomyoma: (a) the uterine/müllerian duct fusion defect theory and (b) the subcoelomic mesenchyme transformation theory. The first theory explains the abnormality in the development of the female genital tract. Each male or female fetus has two pairs of genital ducts: wolffian (mesonephric) and müllerian (paramesonephric).

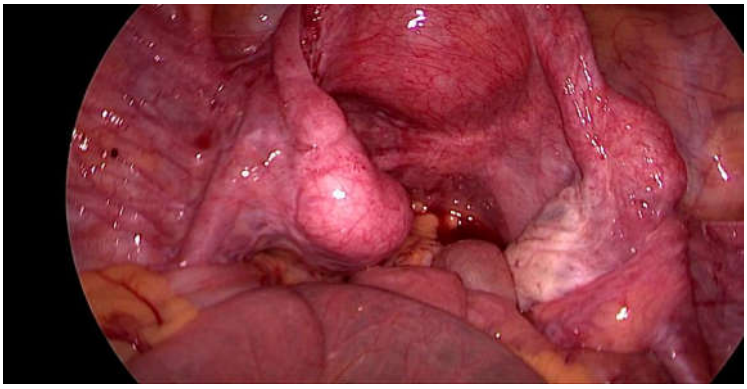

Fig. 1. Bilateral tubal masses visualised on laparoscopy

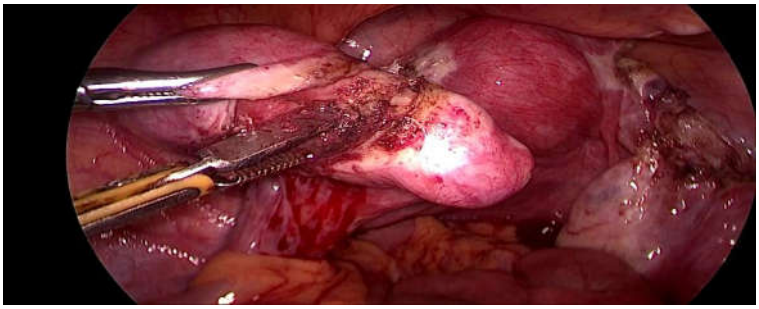

Fig. 2. Dissection of the left large tubal mass

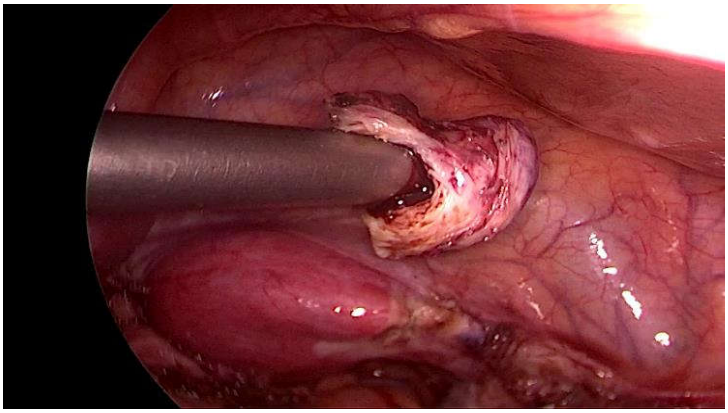

Fig. 3. Morcellating and removing the left tubal mass with dark colored blood inside

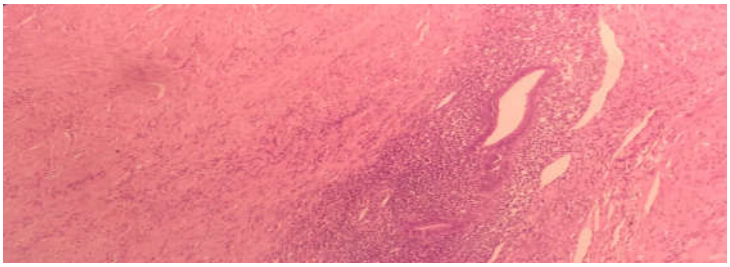

Fig. 4 Endometrial glands and stroma, surrounded by muscle (H&E, 40x)

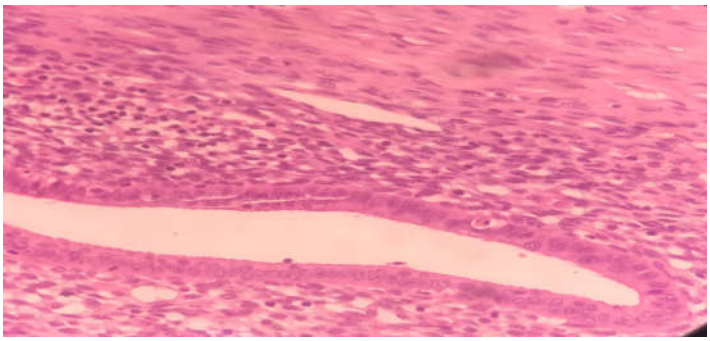

Fig. 5. Endometrial glands and stroma, surrounded by muscle(H&E, 400x)

The müllerian duct, as the main female genital duct, begins as a longitudinal folding of the coelomic epithelium on the anterolateral surface of the urogenital ridge. Lack of fusion of the müllerian duct system may explain various duplications or atresias of the uterus (Redman *et al*., 2005). The same etiology has been proposed to be related to uterine-like mass lesions. The subcoelomic mesenchyme is a layer of tissue that lies underneath the mesothelial surface of the peritoneum. In fetus, this layer of tissue gives rise to the mesenchyme of the urogenital ridge that surrounds the early müllerian and wolffian ducts. In adults, the subcoelomic mesenchyme presents as an inconspicuous layer of flattened cells that lie immediately underneath the subserosal stroma of the uterus, ovaries, tubes, and uterine ligaments. The cells of this layer, which is also called secondary müllerian system, are thought to be multipotential and can proliferate in response to hormonal stimulation (Redman *et al*., 2005). In this case, it is unlikely that the patient had a structural uterine abnormality consistent with a müllerian fusion defect, because she had a normal uterus, cervix, fallopian tubes, ovaries, and was also without any renal abnormality. It is most likely that this extrauterine adenomyoma/uterus-like mass of the fallopian tube arose from the tissues of the secondary müllerian system, which was derived from the subcoelomic mesenchyme.

Conclusion

To the best of our knowledge, this is the first reported case of bilateral tubal adenomyoma. Hence, even though very rare, this entity along with hydrosalpinx due to other causes should be kept in mind while dealing with cases of bilateral tubal masses.

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Author's contributions

R.M; contributed as the surgeon performing and was responsible for overall supervision. S.D; assistant in surgery. Drafted the manuscript and did all the research. S.A; assistant in surgery. Helped with research and revision.

All authors read and approved the final manuscript.

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