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Case Report

Laparoscopic Management of Rare Benign Serous- Cystadenofibroma of Fallopian Tube - A Case Report and Short Review of Literature

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Abstract

Serous cyst adenofibromas of the fallopian tubes are rare benign tumours. Only 18 cases have been reported worldwide [6]. They are usually asymptomatic or are incidentally found during evaluation of pain in abdomen (as in our case) or as co-incidental findings during other surgeries.

We report a case of acute torsion of hydrosalpinx of the right fallopian tube in a 26 year old female. During laparoscopy a large cystic mass measuring 7cms x 8cms was found to have undergone torsion along with the ipsilateral ovary. Laparoscopic right salpingectomy was done and the right ovary was salvaged. Histopathology showed serous cyst adenofibroma of right fallopian tube. We report this case because of its rarity of incidence and short review of literature and also to create awareness of such tumours to avoid aggressive surgery in younger patients and go for ovarian conservation [2].

Keywords: *Benign; Rare Fallopian Tube Tumours; Serous Cyst Adenofibroma; Torsion; Avoid Aggressive Surgery*

Introduction

Serous cyst adenofibroma of the fallopian tube are rare benign tumours. Only 18 cases have been reported worldwide[6]. Most women are asymptomatic and the tumour is discovered as incidental finding at the time of surgery for other gynaecological disorders or during infertility procedures. These tumours are non-neoplastic and of Mullerian origin. Even though these cases are rare, fallopian tube tumours should be kept in mind to avoid aggressive surgery in younger patients and go in for ovarian conservation.

Our case presented with acute abdomen caused due to torsion of the right hydrosalpinx and ovary. It was treated laparoscopically and right salpingectomy was done.

Case Report

A 26 year old unmarried female presented with severe pain in right flank and right buttock for two days. She had inability to pass urine for one day. Vomiting four to five episodes since two days. Her previous menstrual history showed irregular cycles for five years. She would menstruate for six days at intervals of sixty to one hundred twenty days. Her last menstrual period was in November 2019. Past medical history was uneventful. No history of any surgical intervention was there.

Ultrasound whole abdomen showed uterus normal in size. A moderately thick walled clear cystic lesion measuring 7.7 × 4.4 × 4.9 cms was seen in the right adnexa? hydrosalpinx. The lesion showed fine internal septations and no solid components. Gall bladder showed a small 3mm polyp along non dependant walls. Both kidneys are normal in size. Right kidney showed a 3.7 mm calculus in lower calyx. There was minimal pelvicalyceal splitting. Left kidney shows a 3.4 mm concretion in upper calyx and a 4.1 mm calculus in lower calyx.

CECT showed uterus as grossly normal. There is complex right adnexal cyst measuring 5.3 × 6.6 × 6.2 cms showing incomplete internal septations. Bulky ovaries showing hypodense areas in it -likely follicles. Gall bladder was normal. Both kidneys normal. A calculus seen in the lower pole of the left kidney measuring approx. 5.4× 3.5 mm. Concretions are seen in the upper and mid pole of the right kidney and mid pole of the left kidney. Ureters were unremarkable.

Laboratory investigations revealed CA-125 as normal, Beta HCG normal, AFP normal, LDH as normal. Other routine laboratory investigations showed an elevated total leucocyte count with polymorphonuclear leucocytosis.

Laparoscopy revealed a normal uterus. Left ovary was normal but polycystic in appearance. Left tube was normal. Right tube was dilated and tortuous, 7 cms× 8 cms gangrenous, twisted, black in colour. Right ovary was also twisted along with the twisted hydrosalpinx and showed areas of blackish discolouration? gangrene. Right adnexa was de-twisted laparoscopically.

Laparoscopic right salpingectomy was done. Right hydrosalpinx was loaded in a retrieval bag which was brought out through the 10 mm port. Fluid of hydrosalpinx aspirated in- bag and sent for cytology. Specimen retrieved piecemeal from bag without any intraperitoneal spillage. The gangrenous areas on the right ovary were observed for some time - since the ovary started regaining its normal colour decision to leave the ovary was taken.

Patient was discharged on the next day. She was closely followed in out patients department.

Ultrasound pelvis done after three weeks revealed uterus as normal and both adnexa normal. No fluid in POD.

After four weeks patient's recovery was absolutely normal.

Histopathology: Serous cyst adenofibroma right fallopian tubes.

On microscopy of sections from right adnexal cyst branching papillae lined by non- stratified cuboidal to columnar cells showing cilia focally was seen. Underlying wall revealed prominent fibrous stroma. No increase in mitosis or necrosis identified. Fimbrial end was unremarkable.

Cytology of the aspirated fluid was negative for malignant cells.

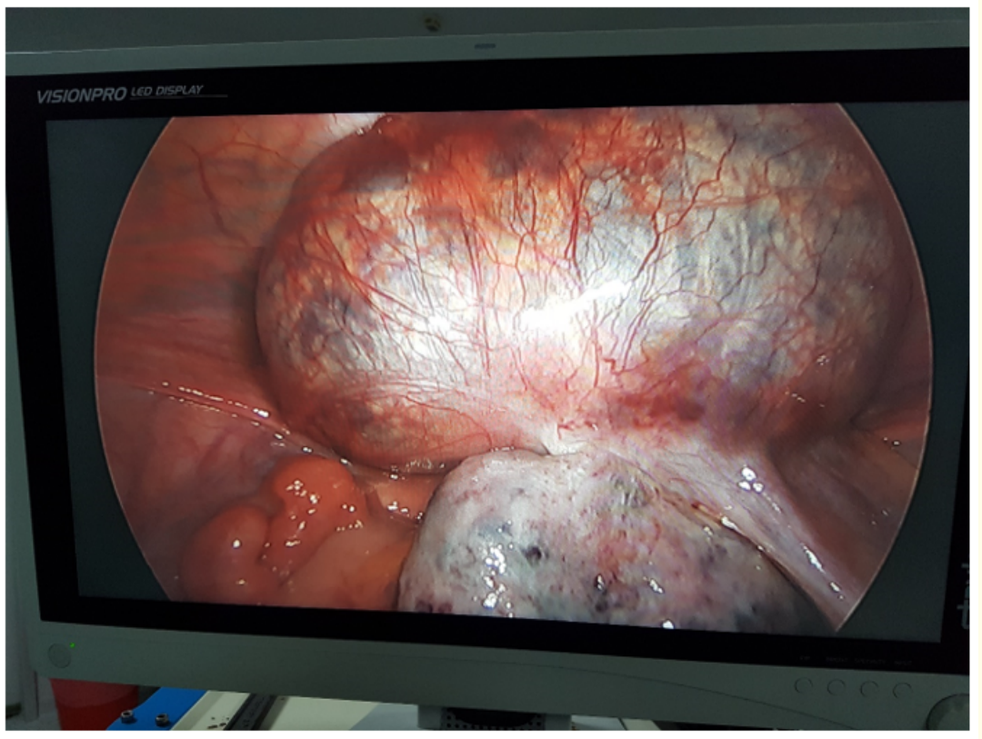

Figure 1: *Laparoscopic view of large hydrosalpinx in mid segment of right Fallopian tube. The fimbrial end appears normal. Right Ovary shows areas of necrosis.*

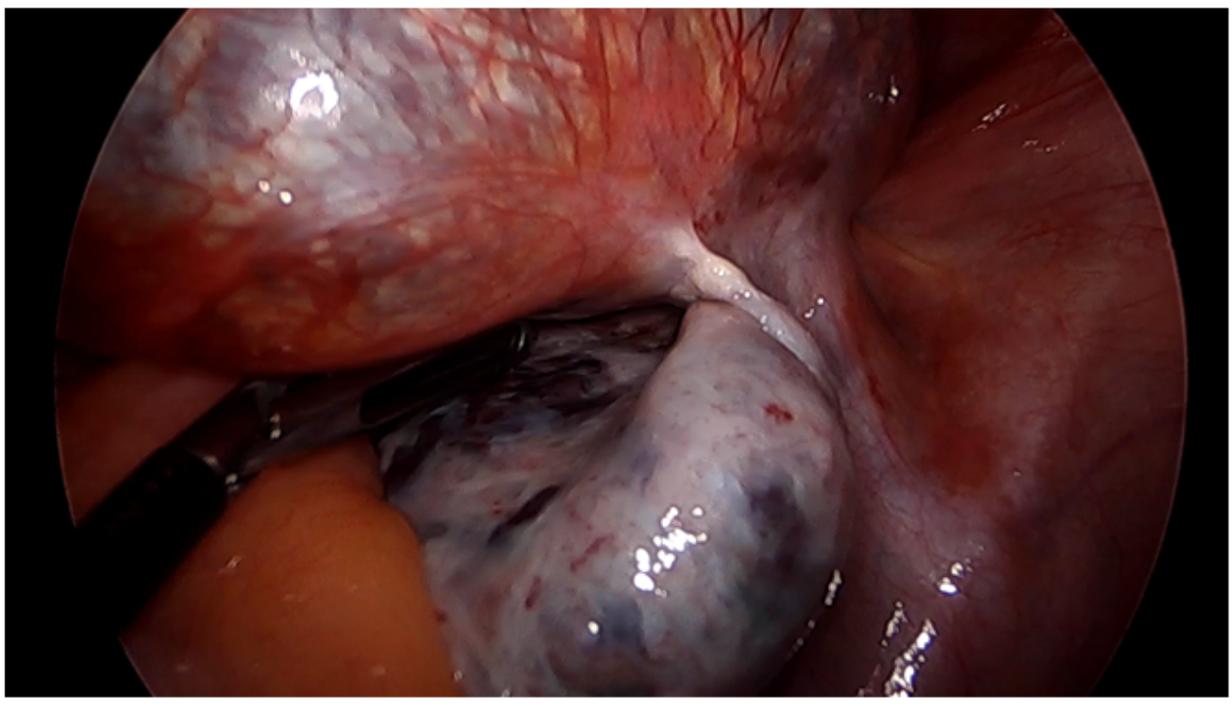

Figure 2: *Laparascopic view of right twisted tube and necrotic right ovary.*

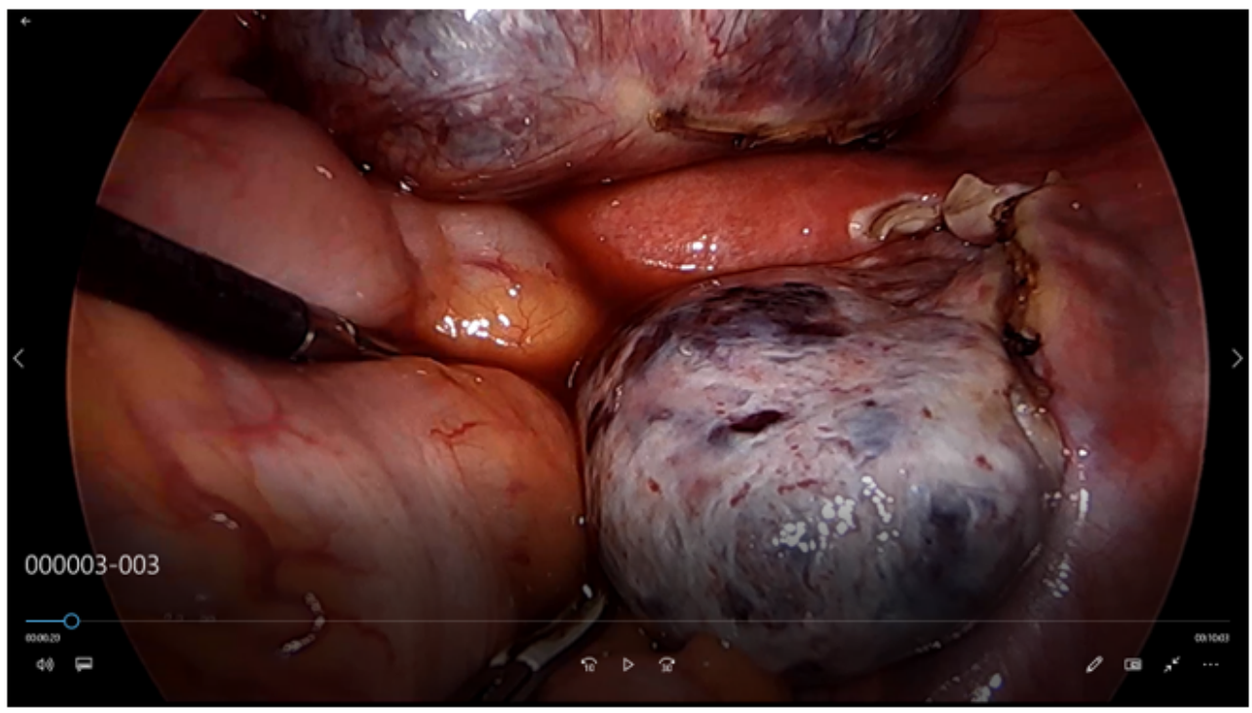

Figure 3: *Right Salpingectomy done. Right hydrosalpinx is parked anterior to the uterus. Right Ovary is regaining its colour after detwisting.*

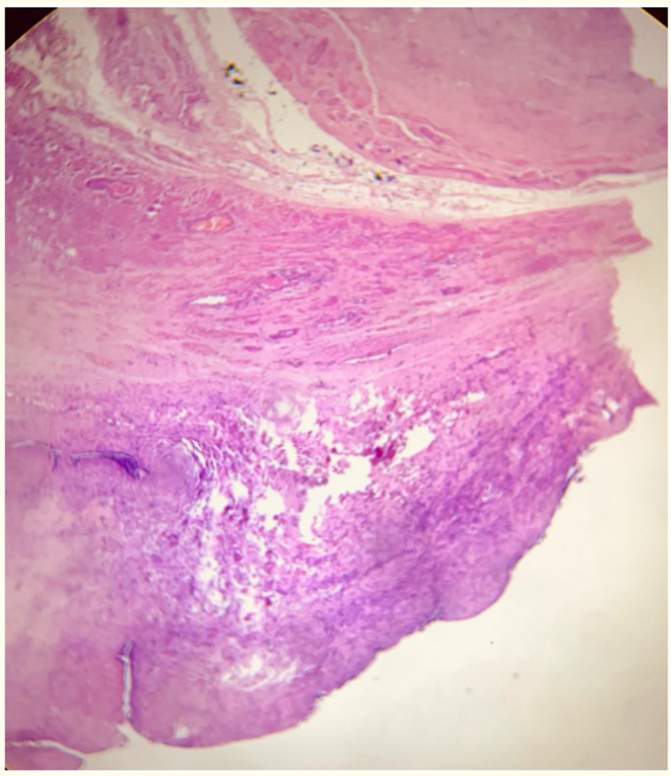

Figure 4: *HE 10x. Cyst wall lined by cuboidal epithelium with fibrous stroma in the wall overlying smooth muscle of fallopian tube.*

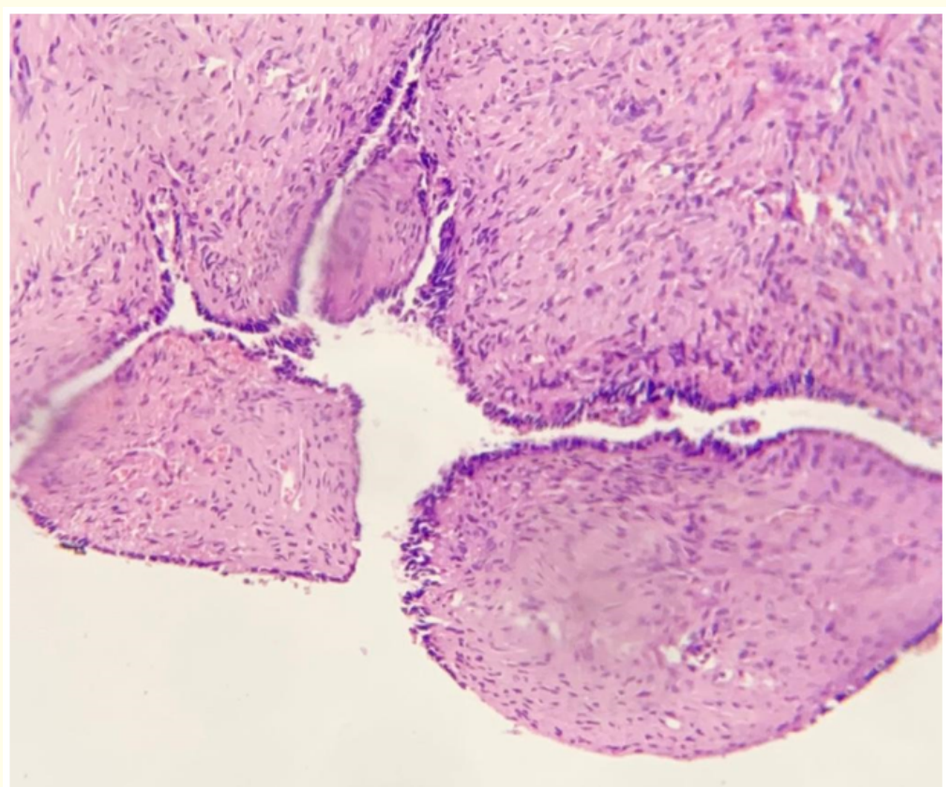

Figure 5: *HE 40 X. Cyst wall showing villi lined by cuboidal to columnar epithelium.*

Discussion

According to the World Health Organization (WHO), benign epithelial tumours of the fallopian tube can be classified into papilloma, cystadenoma, adenofibroma, cyst adenofibroma, metaplastic papillary tumour and endometroid polyps [4]. Iwanow described in 1909, the first case of cyst adenofibroma of the fallopian tube. These tumours arise from germinal lining and ovarian stroma. Tumours of fallopian tube pathologically can be serous, endometroid, clear cell or mucinous type [1]. The clinicopathological presentation of all the cases reported worldwide along with our case is summarized in the below table 1 [6]. Cyst adenofibromas commonly present in the fourth and fifth decade of life however in a subset of patients it is also reported in the reproductive age. These tumours commonly present as a case of acute abdomen. Diagnosis is usually post surgical and clinched on histopathological examination. Differential diagnosis include ovarian torsion, twisted hydrosalpinx, ectopic pregnancy, acute appendicitis. In this case it was confused with renal colic. Mode of treatment is always surgical and nowadays preferably laparoscopy.

Authors	Year of Publication	Age (in years)	Clinical findings	Treatment	Site and size	Microscopy
Kanboer., *et al*. [13]	1973	63	Incidental findings during surgery for prolapse	Vaginal hysterectomy	intramural part of left uterine cornea, 2 cm	Cystic papillary adenofibroma
Silverman., *et al*. [14]	1978	36	Incidental finding during tubal ligation following termination of pregnancy	Bilateral partial salpingectomy	Cystic mass at fimbrial end of left tube, 3.5 cm	Serous cyst adenofibroma
de la Fuente [8]	1982	73	Incidental finding during surgery for uterine leiomyomas	TAH with BLSO	Fimbrial end of right fallopian tube 2.5cm x 2cm x 2cm	Mixed Mullerian tumour-adenofibroma
Casasola and Mindan [12]	1989	32	Incidental finding during operation for multiple uterine leiomyomas	Hysterosalpingoopherectomy	NA	Cyst adenofibroma
Chen [17]	1994	24	Primary infertility	BL tubal cystectomy with wedge biopsy of the ovary	Fimbrial end of right and left fallopian tube, 2.5cm x2cm and 0.3cmx0.2cm respectively	Bilateral papillary adenofibroma
Sills., *et al*. [1]	2003	NA	Incidental finding during IVF embryo transfer	Laparoscopic tubal cystectomy	Distal end of right fallopian tube, 5.5 cm	Serous cyst adenofibroma

Gurbuz and Ozkara [7]	2003	48	Irregular vaginal bleeding with uterine leiomyomas	TAH with BLSO	Serousal surface of right fallopian tube, 0.4cm	Serous cyst adenofibroma
de Silva., *et al*. [2]	2010	19	Pain in right iliac fossa	Right SO	8cm	Cyst adenofibroma
Mondal [16]	2010	27	Ectopic pregnancy	Left salpingectomy	Fimbrial end of left fallopian tube, 2cm x 1.5cm	Adenofibroma with ectopic pregnancy
Erra and Costamagna [4]	2012	50	Incidental finding during operation for leiomyomas	TAH with BLSO	Fimbrial cyst, 3cm	Serous cyst adenofibroma
Pandey., *et al*. [3]	2012	20	Incidental finding during emergency LSCS with BLTL	LSCS with BLTL	Cystic mass in the fimbrial end of left tube, 4 cm x 3 cm	Serous cyst adenofibroma
Fukushima., *et al*. [15]	2014	32	Incidental finding during operation for a suspected case of ectopic Pregnancy	Linear salpingostomy and evisceration	Solid cystic mass bear ampule of left fallopian tub, 20mm	Papillary cyst adenofibroma
Khatib., *et al*. [6]	2015	30	Incidental finding during operation for suspected case of ovarian neoplasm	Left tubal cystectomy	Solid cystic mass on the serosal aspect of left fallopian tub, 12 cm x 10 cm	Serous cyst adenofibroma
Rai., *et al*. Present case	2020	26	Incidental findings during laparoscopy for twisted Hydrosalpinx	Laparoscopic right salpingectomy	Solid cystic mass in right fallopian tube 7cms by 8cms	Serous Cyst adenofibroma

Table 1: *Reported cases of cyst adenofibroma of the fallopian tube.*

TAH with BLSO: Total abdominal hysterectomy with bilateral salpingooophorectomy, LSCS with

BLTL: Lower segment caesarean section with bilateral tubal ligation, NA: Not available, IVF: In vitro fertilization.

Conclusion

The origin of serous cyst adenofibroma of the fallopian tube is not clear. The tumour is considered an embryological remnant of the Mullerian duct rather than a proliferating neoplastic process. These tumours seem to have a benign course and a malignant potential has not been described. Hence in younger patients a conservative approach should be considered, while in patients who have completed their family and are above 50 years a more definitive surgery can be done.

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Conflict of Interests

There is no conflict of interest.

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Authors Contributions

Dr Birbala Rai has a major contribution in operative management of the case. She also conceptualised the case report and planned the article, writing, formatting, finding references and putting the details together. The order of authorship was decided according to the above facts. Dr B Swathi has helped in researching the reference articles and analysing the data, She has also helped in writing the article.

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