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A Rare Case Presentation of Papillary Serous Cystadenofibromain Paraovarian Cyst

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Abstract: ***Papillary serous cystadenofibromais a rare presentation in paraovarian cyst. We report a case of 38year old female with abnormal uterine bleeding with incidental scan finding of cystic mass in right adnexa with septations, who was subjected fortotal abdominal hysterectomy and right salpingectomy with paraovarian cyst excision.Histopathological examination showed papillary serous cystadenofibroma of paraovarian cyst.***

Keywords: Paraovarian cyst, Papillary serouscystadenofibroma, adnexal mass, reproductive age, asymptomatic

1. Introduction

Paraovarian cystsrepresent approximately 10- 20% of adnexal masses.They are usually asymptomatic and are found incidentally on scanning or \at laparotomy. Papillary serous cystadenofibroma is a rare benign serous tumour.This case is presented on account of its rarity and signifies the importance of including paraovarian cysts and its rare presentations in the differential diagnosis of adnexal masses.

Literature Survey: Past review of para ovarian cysts suggested the rarity of the neoplastic lesions of para ovarian cysts and non-availability of clinical studies on our case except one study by histopathologist.

Case report: A38year old parous P_2L_2 with onecaesarian delivery, tubectomised,presented with continuous bleeding pervaginum for 1 month preceeded by 3 months amenorrhea, prior cycles were menorrhagic for 9 months. Bleeding was associated with dysmenorrhoea and passage of clots. There was no pain abdomen in between the cycles. On physical examination, General condition fair.Per abdomen showed subumbilicalparamedianscar, no palpable masses.Per Speculum-posterior lip of cervix nabothian follicles and mild erosion.Bimanual pelvic examination-cervix firm, uterus normal size,anteverted,mobile, fornices free.Her Hematology and Biochemistry results within normal limits.Serum CA 125 - 5µg/dl.Ultrasonography of whole abdomen - uterus 7.3 x 3 x 4.1cm. Endometrial thickness 9mm, Bilocular cyst with anechoic content in right adnexaof 9.8 x 4.5cm with septations. No Ascites.Rest of the organs normal. Hysteroscopy guided endometrial biopsy and cervical biopsy done. Endometrial biopsy revealed non-secretory endometrium with cystoglandular hyperplasia, no evidence of malignancy or endometritis,Cervix-chronic non-specific cervicitis with no evidence of malignancy.

Patient was advised for laparoscopic management of cyst, but denied and insisted for hysterectomy along with cyst removal.

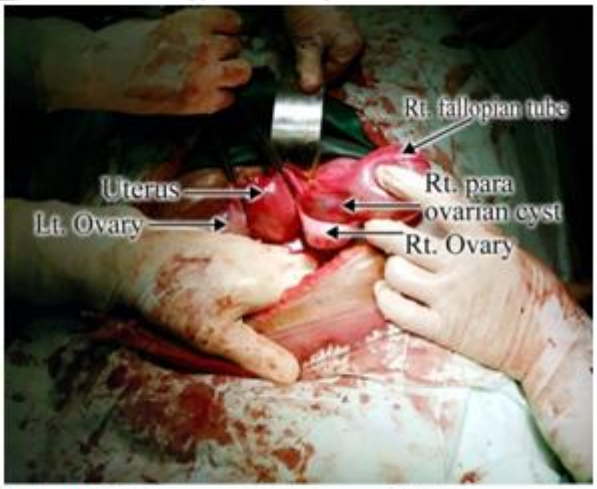

Fig1. Right paraovarian cyst in laparotomy

Laparotomy was performed. Intraoperatively peritoneal cavity was obliterated by layer of Omental adhesions to general peritoneum. Same released and approached the peritoneal cavity. Uterus found to be normal size with thick band ofomental adhesions on to left upper lateral wall ofuterus. Same released after excluding bowel. Both ovaries normal size.Left side fallopian tube normal.On right side, fallopian tube seen compressed.Paraovarian cyst, dumb bell shaped seen in the broad ligament between the medial border of ovary and lateral aspect of right lateral wall. Right fallopian tube was not only compressed but seen above the cyst forming the upperrelation to cyst. Right ovary was away from cyst. Cyst of size 8x4cm, 6x3cm.smooth walled, transparent(Fig.1).Paraovarian cyst excision along with right

salpingectomy andAbdominal hysterectomy done.Biopsy report revealedunilocculated cyst filled with clear fluid, Inner surface showed papillary excrescences. Uterus - proliferative phase, cervix-chronic nonspecific cervicitis with immature squamous metaplasia of endocervical surface lining epithelium and glands with no evidence of malignancy in uterus and cervix.Fallopiantube normal. Thecyst was reported as papillary serous cystadenofibromaof paraovarian cyst. Postoperative period uneventful.

Discussion: Paraovarian cysts represent approximately 10-20% of adnexal masses. They are common in childbearing women.These are hormone sensitive andare generally asymptomatic. Paraovarian cysts are found in the broad ligament between ovary and fallopian tube[1],[2]. They may be either non neoplastic simple cysts (or) cysts of neoplastic origin[1],[3]. The simple paraovarian cyst originates from the embroyonic remnants of the urogenital system (i.e., the mesonephric and paramesonephric duct) or from the invagination of the fallopian tube'sserosa (creating a mesothelialcyst)[(4].Paramesonephric cysts develops as follows: the paramesonephric duct (or mullerian duct) forms the fallopian tube at about 9 weeks gestation. Multiple invaginations that do not connect may form a blind sac and enlarges to form a paraovarian cyst.The neoplastic paraovarian cyst originates from neoplastic transformation of a paraovarian simple cyst (or) from the adjacent ovary[3].In both cases, neoplastic paraovarian cysts are generally benign serous cystssimilar to benign ovariantumors (i.e, cystadenomas or cystadenofibromas). While borderline or even malignant paraovariantumors are encountered less often [5].

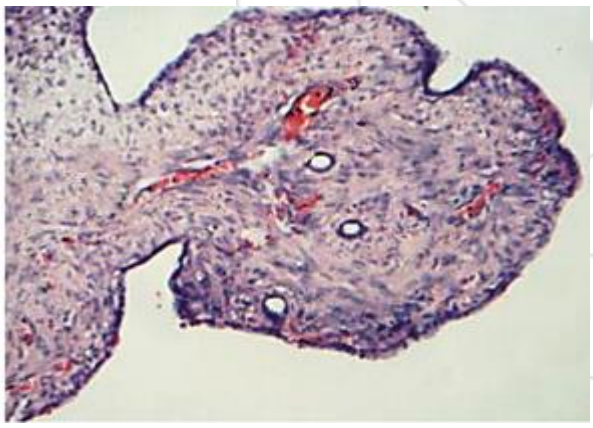

Figure 2: H & E stained Section

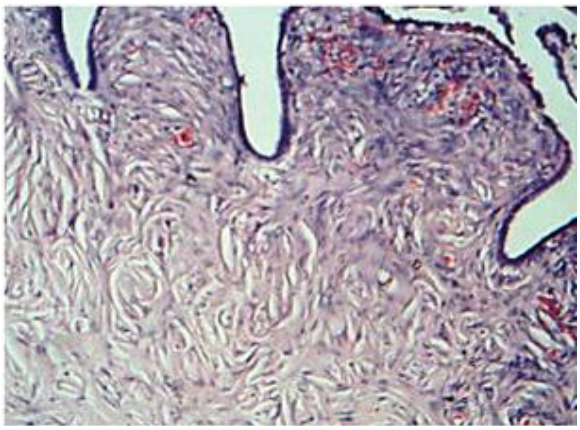

Figure 3: H & E stained Section

Paraovarian cystadenoma histologically resembles with increased frequency in the epididymis of men with Von Hippel Lindau Syndrome.[6],[7]

The prevalence of neoplastic lesions from all types of paraovarian cysts has previously been reported to be low. Grenadry et al.[1] found that 8/140 paraovarian cysts they studied had been neoplasms (4 benign cystadenomas and 4 malignant cystadenomas) 5%.Pepe et al –[8] found only one case of benign cystadenoma in a series of 59 paraovariancysts (1.7%).Stein et al –[9] established that the overall incidence of malignancy in paraovariantumor is 2%.More recently, Savelli et al –[10] found a higher prevalence of neoplastic paraovarian cysts. (15/50 cases, 30%).

Types of paraovariancysts[3],[4]: Paramesonephric cysts, hydatid cysts of morgagni,wolffian cyst, kobeltcyst,cysts of the organ of rosenmuller.Complications of paraovarian cysts being torsion (2-16%)[11],hemorrhage, rupture, secondary infection, neoplastic transformation (2.9%).Neoplasms are papillary serouscystadenoma, endometrioidcystadenocarcinoma, serous cystadenocarcinoma, mucinous cystadenocarcinoma. .

Differential diagnosis of extraovarian masses in female pelvis: Predominantly cystic masses – common are hydrosalpinx,paraovariancyst, peritoneal inclusion cyst. Uncommon are paraovarian cystadenoma, tarlov cyst, bladder diverticulae, lymphocele, abscess, varices. Predominantly solid masses – common are pedunculated fibroid, Uncommon are neural neoplasms, lymphadenopathy,rudimentary horn of unicornuate uterus.

In ultrasound, Paraovarian cysts appear as simple cysts with variable sizes, thin wall, unilocular, Smooth margins usually with anechoic content and are indistinguishable from ovarian cysts if one does not recognize the extraovarianlocation.The corresponding ovary if visualised is usually normal. Occasionally,paraovarian cysts have internal echoes due to hemorrhage.Paraovarian cystadenomas or paraovariancystadenofibromas are uncommon,typically have a mural noduleor septation.[10],[12],[13]Avery uncommon and rare presentation quoted in radiological studies [12].

Histopathology of Papillary serouscystadenofibroma(Fig.2 &Fig.3): contains epithelial and fibrous components with papillary excrescences. Cyst lined by a single layer of flattened to cuboidal cells with uniform basal nuclei, and stroma is highly cellular and fibrous.[14].Though studies are available on papillary serous cystadenofibroma of ovary[15]and on benign cystadenofibroma of fallopian tube[16], clinical studies on papillary serous cystadenofibroma of paraovarian cysts are not available except one case reported by histopathologist [14].A Case report on papillary cystadenofibroma of epididymis is also available [17].This indicates the rarity of the case. Management: depends not only on the size but also presence, number of septations and loculi and anechoic content.

2. Conclusion

Any adnexal mass with septations and anechoic content in sonography should be suspected to have a neoplastic component and thus shouldbe evaluated completely and treated appropriately.

3. Future Scope

As neoplastic lesions of para ovarian cysts were under reported, with our case it emphasizes the need of careful histopathological examination of adnexal masses, in particular paraovarian cysts for any neoplastic component.

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