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Case report

Paratubal serous borderline tumor in an 85 years old woman: A case report

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ABSTRACT

Paratubal cysts are incidental, common and benign lesions with few cases of borderline tumor and adenocarcinoma been reported in the literature.

Herein, we are discussing a case of an 85 years old woman who visited her Obstetrics and Gynecologist for a chief complaint of post-menopausal bleeding. Physical examination revealed a slightly enlarged uterus. The diagnosis of fibroids were made on ultrasound.

A total hysterectomy with bilateral salpingo-oophorectomy was performed without complications. The gross examination of the specimen was most remarkable for multiple intramural fibroids and a right simple paratubal cyst meausiring 1×1 cm. Microscopical evaluation of the paratubal cyst showed a simple cyst lined by stratified cuboidal epithelium with mild cytologic atypia and no stromal invasion, findings consistent with serous borderline tumor rising in a paratubal cyst. Due to the small size of the tumor and its confinement to the cyst, a follow-up was advised.

Serous borderline tumors arising in paratubal cyst are very rare with only eight cases reported in the literature. Their existence should be acknowledged to plan patient treatment and outcome.

1. Introduction

Paratubal cyst (PC), also known as "hydatid cysts of Morgani", are of Mullerian origin. The vast majority of PC are unilateral, asymptomatic, and benign. Microscopically, PC is lined by single tubal-type ciliated epithelium with no stratification, no cytologic atypia, or mitosis. Due to their derivation from Mullerian origin, and similarly to ovarian surface epithelial tumors, a spectrum of histologic subtypes of epithelial neoplasms can occur, ranging from benign, borderline tumor to adenocarcinoma (Chang et al., 2017; Im et al., 2011). Few cases of serous endometrioid and mucinous borderline tumors arising in a paratubal cyst have also been reported (Im et al., 2011). We will report a case of paratubal serous borderline tumor in an 85 years old woman and we will review the literature and discuss treatment and outcomes.

2. Case report

An 85 year old woman G2P2, was seen by her gynecologist for recent complaints of postmenopausal bleeding. An ultrasound revealed numerous intramural hypoechoic masses and calcifications consistent with leiomyomas. No other lesions were present. Patients had type

diabetes, hypercholesterolemia and hypertension. No family history of cancer. The patient was scheduled for a total abdominal hysterectomy and bilateral salpingo-oophorectomy (TAH + BSO). The surgery was uneventful. The gross examination was unremarkable except for numerous intramural fibroids and a right simple paratubal cyst measuring 1 cm \times 1 cm. On microscopic evaluation, the inner cyst lining was characterized by a hierarchical branching pattern and papillae lined by cuboidal to columnar epithelium with minimal cytologic atypia. No stromal invasion was seen (Figs. 1a and 1b). Subsequently, the entire fallopian tubes and ovaries were negative for any significant benign or malignant lesion upon histologic examination. Sections of the myometrial nodules show leiomyomas. Patient underwent full recovery and no further treatment was needed at her 3 months follow-up visit. Patient was scheduled for another 3 months follow-up visit.

3. Discussion

Partubal serous borderline tumors or serous tumors of low malignant potential are very rare. Using the Pubmed searching engine, only 8 cases were found in the English literature (Seamon et al., 2009; Shin et al., 2010; Puig et al., 2006; Kumbak et al., 2010; Terek et al., 2011;

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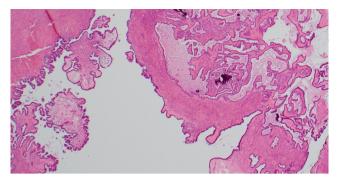


Fig. 1a. Hematoxylin and eosin (HE) section of the cyst shows a cystic lesion. The cyst is lined by broad papillae and few areas of tufting. There is no stromal invasion seen. (\times 20).

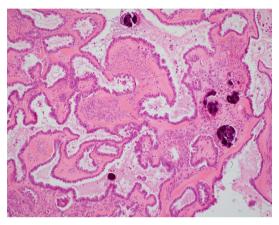


Fig. 1b. HE section sat higher magnification shows the cyst are lined by stratified epithelium with minimal cytologic atypia (\times 40).

Lee et al., 2016; Alaoui et al., 2012; Kiseli et al., 2012). Our case is the 9th of its kind. Based on our review, no age predilection was noted as it can be seen in patients of any age group and ranged from 17 to 85 years, as in our case, with mean age of 35.6 years and median of 26 years. All reported cases were unilateral. The clinical presentations varied from sharp pelvic pain usually due to torsion and large size, to asymptomatic only to be discovered incidentally on ultrasound or at time of surgery. The differentiation between ovarian and tubal and paratubal cysts based on ultrasound imaging alone is very tricky and limited. When a cyst is discovered, a CA125 level would be important to evaluate a potential malignancy and therefore to plan accordingly. The size of could range from 1 cm, as in our case, to 16 cm in diameter as observed in the literature. All paratubal cysts are examined microscopically to exclude malignancy, therefore their rarity is not due to under-reporting but to their rare nature. On gross examination, they usually present as simple cysts and in rare cases as complex cysts. The differential diagnosis is between benign cyst, borderline tumors, adenocarcinoma arising in a paratubal cyst, or even more it could be any of those pathologies arising in organs at proximity such as fallopian tubes and ovaries. Therefore, all paratubal cysts are submitted for microscopic evaluation. The morphologic findings were typical of serous borderline tumor where the cyst is lined by broad, branching papillae covered by stratified epithelium with tufting, minimal nuclear atypia and few mitotic figures which is not seen in benign paratubal cyst. The stroma can be edematous but with no invasion which exclude an

adenocarcinoma that is defined by stromal invasion by tumor cells. Because of the proximity and intimate anatomy of the fallopian tube, paraovarian and mesosalpinx, in most circumstances, the histological diagnosis can be challenging as to evaluate the origin of this tumor. In those circumstances, we recommend the entire ovaries and fallopian tubes be submitted for microscopic evaluation to exclude any alternative diagnoses. Therefore, a pratubal origin is a diagnosis of exclusion. Surgical Management of these masses varied notably depending on the patient's age as well as the surgery's indications. It included simple cystectomy, unilateral salpingectomy, unilateral salpingo-oophorectomy, hysterectomy as well as bilateral salpingo-oophorectomy (TAH + BSO). For young patients, a fertility-preserving surgery was the preferred modality. However, in older patients like ours and because of her pre-op diagnosis of fibroids, a TAH + BSO was done. All reported cases were confined to the cyst with 7 cases staged as FIGO 1A and two cases were ruptured and were staged as FIGO 1C. However, all 9 patients including ours had no further treatment and no recurrences at follow-up ranging from 3 months to 20 months was noted. Herein, our case is unique for two reasons: 1. It is the smallest SBT arising in a paratubal cyst ever been reported and 2. Our patient is the oldest of all patients that has been seen.

CRediT authorship contribution statement

Jordy Mehawej: Writing - original draft, Writing - review & editing. Nicole El Helou: Writing - original draft, Writing - review & editing. Lina Wang: Data curation, Investigation, Writing - review & editing. Paulette Mhawech-Fauceglia: Supervision, Writing - review & editing.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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