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CASE REPORT

DIAGNOSTIC DILEMMA DURING LAPAROSCOPIC CYSTECTOMY OF TWISTED SEROUS CYSTADENOFIBROMA OF OVARY IN ADOLESCENT GIRL – A CASE REPORT WITH LITERATURE REVIEW

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ABSTRACT

Most of the ovarian tumors arises from surface epithelium and serous tumor is the most common histological variant. Serous cystadenofibroma is a rare variant of serous tumor that comprises of both epithelial and fibrous stromal components. Though benign in nature they may often be misinterpreted as malignant due to their solid cystic nature with papillary projections. We herein report a case of a twisted right ovarian cyst in a 17 years unmarried girl that was clinically and radiologically benign and managed laparoscopically. Intra-operatively, the cyst ruptured releasing clear serous fluid. During cystectomy, multiple small to large size (maximum 2x2 cm) papillary projections were noted within the uniloculated cyst without any other solid components. With the possibility of borderline tumor, patient party was recounseled and opted for cystectomy with possible need of second surgery. Final histopathology report was benign serous cystadenofibroma.

Keywords: *adolescent; cystectomy; cystadenomafibroma; laparoscopy*

INTRODUCTION

Serous tumor is the most common epithelial ovarian tumor. Cystic adenofibroma is a variant of serous tumor with prominent fibroblastic stromal component.[1] It is a rare benign tumor of unknown etiology. Because of its solid cystic nature, it may mimic malignant ovarian tumor clinically and radiologically. It is common in the fourth and fifth decades of life.[2] Though uncommon, it can occur in young age group where fertility is also a focus for management without compromising the outcome of treatment .[3] We herein report a case of a 17 years patient who underwent emergency laparoscopic cystectomy for twisted ovarian cyst and solid cystic nature of the cyst was detected intra-operatively only.

CASE REPORT

A 17 years unmarried girl, accompanied by her father, presented to emergency department with the complain of sudden onset of non-radiating severe pain lower abdomen more at right side associated with three episodes of vomiting. The pain was continuous in nature, and not relieved with intake of oral analgesic at home. She gave no history of fever, abdominal distension and was passing flatus. Her menstrual cycles were regular and the last menstrual period was 11 days prior to presentation. She was a student and not sexually active. There was no history of any surgery done in the past.

On examination, she had tachycardia and tachypnea attributable to pain. However, she was not pale, blood pressure was within normal range with normal oxygen saturation at room air. Abdomen was not distended. Right lower abdomen and suprapubic region were severely tender without rebound tenderness. No mass was palpable. Digital rectal examination revealed fullness at right side with suspicious cystic lesion that was tender.

Bedside urine pregnancy test was negative. Urgent transabdominal ultrasound was done with retrograde filling of urinary bladder that revealed enlarged right ovary with increased vascularity measuring measuring 84x78x64 mm located anterior to uterus and superior to urinary bladder with no internal vascularity, calcifications and mural nodules or septations. Left ovary was normal .Clinical and radiological diagnosis of twisted right ovary with benign cyst was made (Fig. 1)

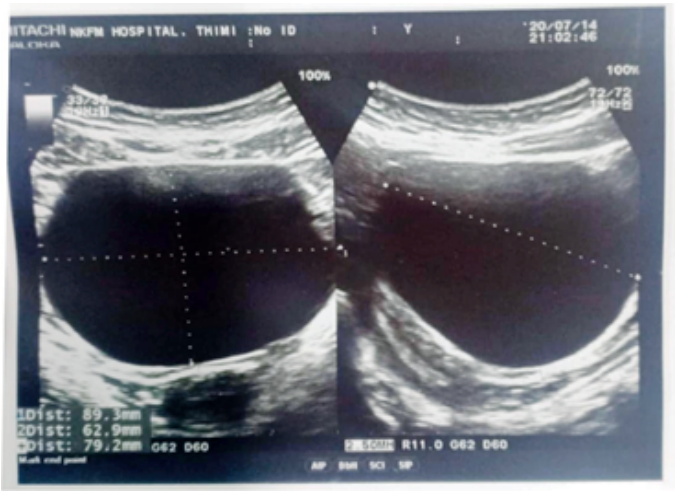

Figure 1: Transabdominal Ultrasound Showing Uniloculated Cyst Without Internal Vascularity, Calcifications, Mural Nodules or Septations

With the clinical and radiological features of benign nature, laparoscopic management was suggested. Our provisional plan was to do cystectomy only. However, informed consent was taken for salpingo-oophorectomy as well if adnexal structures were not viable.

A primary 10 mm port was placed at umbilicus with Hasson technique and two contralateral secondary 5 mm ports were placed 2 cm medial and superior to anterior superior iliac spine. Diagnostic laparoscopy was done first. There was no free fluid. Right ovarian cyst was noted of size 8x6cm, globular in nature, smooth surface, intact capsule, regular margin without surface nodularities suggestive of benign nature (Fig.2). The cyst along with fallopian tube was twisted four times along its pedicle. However whole structures were viable. Upper abdomen was also grossly normal.

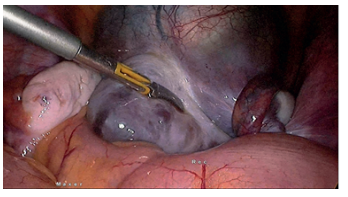

Figure 2: Twisted right ovarian cyst

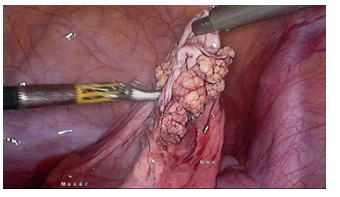

Figure 3: Multiple papillary projections within the cyst

Untwisting of cyst was done and cystectomy was proceeded. There was inadvertent rupture of cyst that released clear fluid. But to the surprise of the surgical team, multiple papillary projections of variable size (5x5mm to 2x2 cm) propped out of the rent that was not seen in ultrasound (Fig 3)

As frozen section was not available and with borderline features, per-operative counseling was done to the father with option of either direct salpingoophorectomy or cystectomy with second surgery if required after the final report. Reconsent was taken for cystectomy only under newcircumstances and cystecotmy was completed.

Serum quantitative tumor markers (CA-125, CEA, AFP, CA-19.9, LDH and HCG) were sent on the first post-operative day only and all were normal. She was discharged on the second post operative day. The clinical events were informed to the pathologist also. Final histopathology was reported as serous cystadenofibroma (Fig.4). Owing to its benign nature, she was kept under regular follow-up only with close monitoring.

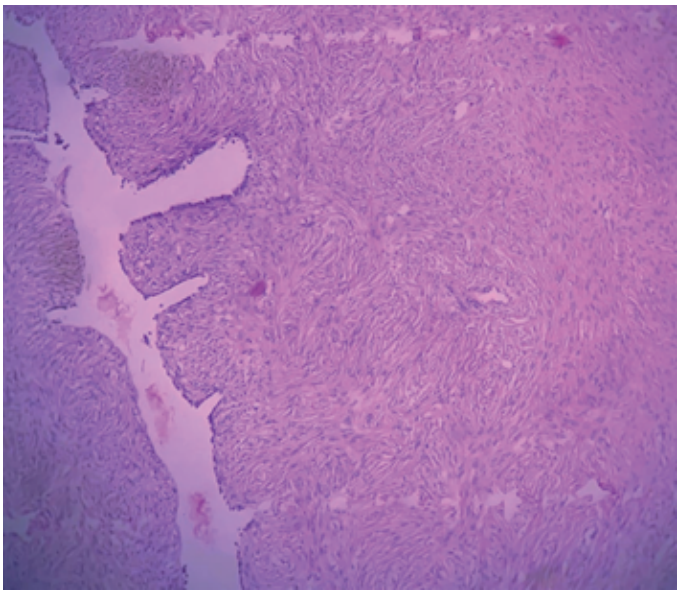

Figure 4: Cyst lined by uniform cuboidal cells surrounded by solid fibrocollagenous tissue

DISCUSSION

Cystadenofibroma is a rare variant of epithelial ovarian tumor with both epithelial and fibrous component that accounts for 1.7% of all benign ovarian tumors. [2] The reported common age group varies from 35-65 years [2] or fourth to fifth decade of life. [3] It is very rare in adolescent girls. Asnat Groutz et al. have reported five cases of serous cystadnofibroma with age group of 19-21 years (two cases of 19 years, two cases of 20 years and one case of 21 years).[3] There are other case reports with viable age group from 34 years,[4] 25 years,[5] 28 years,[6] even in post menopausal lady at age of 50 years.[7] However our case showed that it can occur in young age as early as 17 years.

Clinically, they present with features of abdominal

discomfort, pain abdomen and palpable mass. Uncommon features like nausea, loss of appetite, vaginal bleeding and feminization may sometimes mimic malignancy clinically.[1,8] Vaginal bleeding and feminization may be due to estrogen section from the tumor that makes the endometrium thick. Bilateral cysts are seen in 6-30% of the cases. [3] Ultrasound is the preferred modality for the detection of ovarian cyst. Usual findings are uniloculated or multiloclated cystic solid nodules or papillary projections with increased vascularity in 50% of the cases.[8] Ultrasound can miss papillary projection especially when they are exocystic in nature. It has been reported that even with both transabdomial and transvaginal ultrasound study, it has sensitivity and specificity of 78%, and 96% respectively with positive predictive value of 84%and negative predictive value of 94%. Overall accuracy is 92% in detecting papillary projections.[9] In our case also, transabdomial ultrasound did not detect papillary projection.

When ultrasound is suspicious, further imaging can be done with Computed Tomography (CT) or Magnetic Resonance Imaging (MRI). Cho et al. has reported that there is higher chance (50%) of being reported as malignancy in CT and MRI (16 / 32).[2] MRI findings may vary from pure cystic to cystic solid in the form of trabecular or nodular solid areas. Thick and irregular septa are also common that may mimic malignancy. In our case further radiological evaluation was not done due to emergency presentation and suspicion of torsion.

Tumor markers were not raised in a case series report by Groutz.[3] But CA 125 was reported raised by nearly two folds (66.9 IU/L) in a case report of bilateral serous papillary cystadenofibroma of ovary.[1] In contrary CA 125 was reported markedly raised (995.4 U/ml) in a case report by Deepali Jain, but it was a case of primary psammocarcinoma of ovary with serous cystadenofibroma of contralateral ovary.[10] In this case also, tumor markers were sent on the first post operative day and found to be normal.

Surgical removal can be both via laparotomy or laparoscopy. Being benign in nature, cystectomy is optimal.[11] Depending upon age and parity; salpingoophorectomy can be done. There is a practical chance of extensive surgery being carried out due to misleading possibility of malignancy specially when there are no services for frozen section. Over-treatment should be avoided specially in a young patient like in our case. Frozen section should be carried out wherever available. But in settings with limited set-up, two stage surgery is also good option as we initially planned in our case. The girl was fortunate to have benign lesion and hence did not require second surgery as well we could preserve her ovaries. Laparoscopic management has been reported for cystadenofibroma in other literatures also without any further complications.[12] Contralateral ovarian wedge biopsy is an option considering the bilateral nature of the pathology .[3]

Papillary projections are also common in borderline ovarian tumors.[13] Histologically, they are neoplasms with high degree of architecturally complex epithelial proliferation with hierarchically branching papillae and pseudopapillae. Stroma is paucicellular and edematous and may be hyalinised. There may be mild to moderate nuclear atypia and hyperchromasia.[14] But in cystadenofibroma, there are prominent fibroblastic stromal component that is composed of spindled cells arranged in fascicles and storiform patterns. Though they are benign with rare chance of malignancy, coexisting adenocarcinoma has also been reported. [15-17]

Due to few case reports, recurrence rate is still unknown especially after fertility sparing surgery. [7] But in a report of 34 cases of serous adenofibroma, outcome was found to be similar regardless of nature of management. [11]

CONCLUSION

Serous cystadenofibroma is benign in nature with rare chance of malignant potential and clinical /radiological and operative findings may mimic malignancy. The surgeon should be aware of chance of being benign in nature, despite solid cystic nature of the cyst, before choosing aggressive surgery especially in younger age. When in doubt, two step surgery is a better option when frozen section is not available.

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