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Twisted Ovarian Tumor with Cholesterol Granuloma: An Unusual Morphological Entity with Review of Literature

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Abstract: We report a case of ovarian tumor in a 35 year old woman who presented in emergency with acute abdominal pain. She had a history of intermittent abdominal pain and weight loss since 6 months alongwith abdominal distension of 1 month duration. On examination a tender pelvic mass extending upto hypogastrium was palpated. The mass was well defined with restricted mobility mainly on right side of abdomen. Emergency laprotomy was performed which revealed a right sided twisted ovarian cyst ms 20cm in diameter with haemorrhage in the peritoneal cavity. Left ovary also showed presence of cyst 4cm in diameter. Omentum showed firm nodular areas. Both cysts and omental nodules were excised and sent for histopathological examination. Peritoneal fluid was sent for cytological examination as the mass was thought to be malignant. Grossly, the cyst had smooth, deeply congested outer surface. Microscopy revealed benign mucinous cystadenoma ovary with extensive areas of haemorrhage, acute inflammation and formation of cholesterol granulomata. This unusual morphologic entity in ovary is quite rare and hence this case report.
Keywords: Ovarian tumor, Torsion, Mucinous cystadenoma, Cholesterol granuloma.

INTRODUCTION

Ovarian torsion is fifth most common gyanaecological emergency with a reported prevalence of 2.7% in all cases of acute abdominal pain [1]. It is defined as partial or complete rotation of adenexa around its vascular axis that may cause an interruption in the ovarian blood and lymphatic flow [2] which may further lead to secondary changes. Ovarian torsion occurs far more commonly during pregnancy than in non-pregnant state. Torsion of a normal ovary is rare and the typical presentation is usually of unilateral torsion of a pathologically enlarged ovary [3]. Torsion leads to compromise of the vascular supply leading to haemorrhage and necrosis. Extensive intratumoral haemorrhage may lead to secondary changes, one of which is formation of cholesterol granulomas, though it is a rare morphological change associated with ischaemia [4]. Granulomatous inflammation is a common tissue response to a wide variety of stimulus including foreign body, infective agents as well as other substances. Cholesterol granuloma is a foreign body reaction to presence of cholesterol crystals formed during the inflammatory process [5].

CASE REPORT

A 37year old thin built lady reported in the emergency department of our hospital with acute abdomen. She had a history of intermittent abdominal pain on right side since 6 months alongwith weight loss of same duration. Since 1month she had also developed abdominal distension. There was no history of vomiting, loose stools, vaginal discharge, bleeding per vaginum, or dysuria. She was para two with two live issues delivered by caesarean section, last being done 3years back. On examination, her abdomen was distended with a mass of 26weeks size arising from pelvis on right side of abdomen. The mass was extending upto the hypogastrium, was firm, tender, with well defined contours and restricted mobility. There was local rise of temperature. Her blood examination suggested inflammatory process in progress with Hb 8.0gm%; TC-13,700 cells/cumm; DC-P88L08E02M02; ESR-47mm in 1st hr. Urine for routine & microscopy was WNL. Rest of the biochemical parameters were within normal limits. USG showed a large fluid filled cystic mass measuring 20x17cms with multiple septations and internal echoes arising from pelvis and extending superiorly. Her CA125 was within normal limits [7.7IU/ml; RR <35IU/ml]. The patient was subjected to emergency laprotomy.Per op findings

revealed a right sided twisted ovarian cyst (one and a half turn of a circle along the axis of fallopian tube) of size 20x15cms.The left ovary also showed a cystic mass 4x3cm. There was free fluid in the peritoneal cavity. Omentum showed firm nodular deposites of <1cm size. A malignant ovarian tumor was suspected and bilateral ovarian cysts along with omentum were sent for HPE. Peritoneal fluid was sent for cytology.

Histopathological findings

2 cystic ovarian masses of size 20cms and 4 cms respectively in maximum diameter were received alongwith omentum measuring 10x8cms. The larger cystic mass showed a smooth blackish outer surface. Cut surface revealed multilocular cyst filled with haemorrhagic fluid and a solid appearing area measuring 9x6 cms (Fig. 1). The solid area showed yellowish, greasy, shiny areas in between surrounded by microcystic areas filled with mucoid material (Fig. 2). Inner lining was deeply congested (Fig. 2). The smaller cyst showed outer smooth white surface. Cut surface showed unilocular cyst filled with straw colouerd fluid with a peripherally compressed ovarian tissue. Omentum showed firm grayish white nodular thickenings.

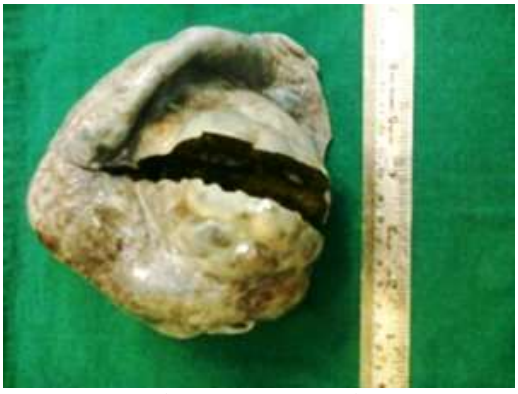

Fig 1: Large ovarian mass measuring 20x18x12cms. Outer surface is smooth well defined showing areas of congestion

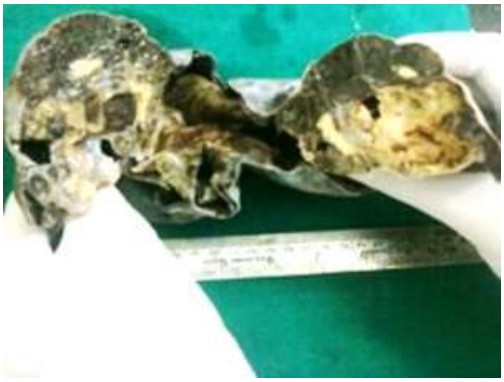

Fig. 2: Cut surface of tumor mass showing yellowish areas of cholesterol deposition surrounded by congested solid appearing areas with small mucin filled cysts

Microscopy

Sections from varied areas showed thick fibrocollagenous wall tissue lined by tall columnar mucin secreting epithelium (Fig. 3) with underlying glands of similar lining. The glands showed mucin admixed with neutrophilic infiltrate in the lumen (Fig. 4). Large areas of haemorrhage with intense neutrophilic infiltrate were seen in majority of the sections (Fig. 4). Sections from yellowish areas showed presence of cholesterol clefts surrounded by numerous multinucleated foreign body type giant cells, neutrophils and lymphoplasmacytic infiltrate (Fig. 5). Other areas showed mucin pools with detached lining epithelium and neutrophilic exudates.Sections from omental nodules also showed areas of fibrosis and intense neutrophilc exudate. No evidence of malignancy was seen in any of the sections processed. Left sided ovarian cyst was compatible with a follicular cyst. Peritoneal fluid cytology also revealed inflammatory cells, reactive mesothelial cells and debris.

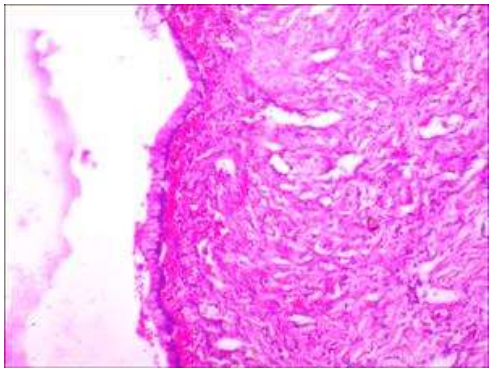

Fig 3: Section from tumor mass showing thick fibrocollagenous wall lined by tall columnar mucin secreting epithelium (H&E; 100x)

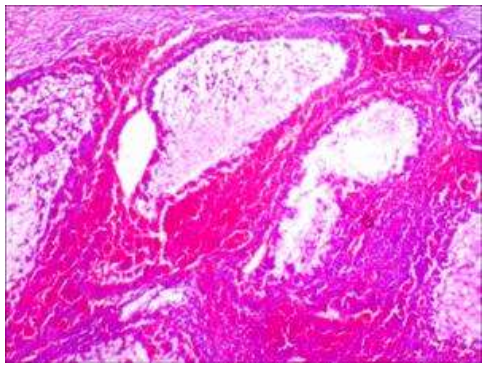

Fig. 4: Section showing mucin secreting glands with extensive haemorrhage and inflammation in surrounding areas (H&E; 100x)

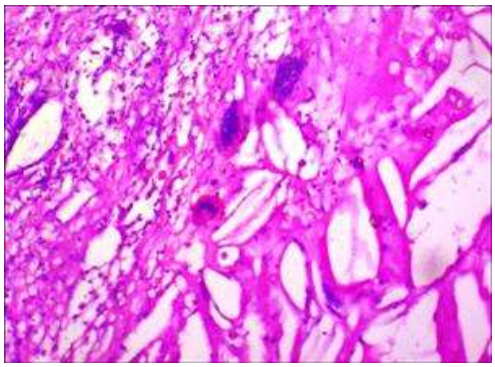

Fig. 5: Section from the tumor mass showing areas of cholesterol clefts surrounded by multinucleated giant cells and chronic inflammatory cells (H&E; 200x)

DISCUSSION

American studies have reported that ovarian torsion is the 5th most common cause of surgical emergency in gynaecology and most reported cases occur in the reproductive age group. Median age varied from 28 to 33 years as reported in literature [6, 7]. Malignant tumors are less likely to undergo torsion due to the presence of cancerous adhesions that fix the ovary to the surrounding structures. Thus, tumors which undergo torsion are more likely to be benign, as was in our case. The most common benign tumor reported to have undergone torsion is dermoid which was contrary to our case which was a mucinous cystadenoma. The typical clinical history in ovarian torsion is of sudden, severe, unilateral lower abdominal pain that worsens intermittently [3]. Nausea and vomiting are seen in 70% cases [3], which was not reported in our case. USG is the most important investigation of choice for confirmation of diagnosis.

Cholesterol granuloma is a histopathologic entity describing large number of clefts present after cholesterol crystals have dissolved during processing with surrounding foreign body type giant cells, foam cells and macrophages filled with hemosiderin in a fibrous granulation tissue [8]. Extensive search of literature revealed that no case of twisted ovarian tumor with this morphological entity has been described as far as we know. Though presence of cholesterol granulomas have been reported in pituitary tumors [4] and inflammation of maxillary sinus [5]. A single case of ovarian cholesterol granuloma has been reported in a 15yr old Yorkshire bitch in a veterinary journal though it was not associated with any tumor or torsion [9]. The authors hypothesized the lesion to be secondary to haemorrhage and congestion. In addition they included high serum lipid levels too in the predisposing factors. In our case S. lipid levels were not done. Grignon DJ *et al*. [10] reported cholesterol granuloma in iliac and paraaortic group of lymph nodes which were excised alongwith ovarian tumor in an elderly lady, considering the tumor to be malignant. But later on histopathology showed it to be a benign mucinous cystadenoma. They speculated that fluid from ovarian neoplasm drained into the regional lymph nodes causing this unusual granulomatous response.

It has been postulated that the process starts with haemorrhage. The torsion leads to obstruction of venous and lymphatic drainage, leading to microhaemorrhages [11]. Arterial obstruction causes infarction which leads to extensive haemorrhage and acute inflammatory cell response which was seen in our case. As a result, large amount of RBC breakdown takes place causing membrane damage and lipid accumulation. Normally, if the lymphatics are not obstructed completely these lipids are removed from the site but with complete obstruction of lymphatics these are not cleared leading to formation of cholesterol crystals and their esters [11]. These in turn give rise to a granulomatous reaction with formation of foreign body type giant cells. We postulate that this is what happened in our case. The extended duration of complaint seen in our patient might have led to amplification of secondary response along with subsequent spread of inflammation to omentum and also explains the weight loss in the patient.

In experimental works done by Niho [12] and Maine [13], the isolation of haemorrhage in a small area without ventilation was important in pathogenesis of formation of cholesterol granulomas. Niho [12] suggested that cholesterol deposits were a fatty degeneration of connective tissue in a cavity obstructed by inflammation.

CONCLUSION

Based on available clinical information and experimental studies, key factors for formation of cholesterol granulomas are prolong inflammation and obstruction secondary to haemorrhage as was seen in our patient. In our case, the association of cholesterol granuloma with mucinous cystadenoma is not so important as the fact that these changes were precipitated because of torsion of ovarian tumor itself. Torsion led to compromised blood and lymphatic flow leading to haemorrhage and inflammation which secondarily led to formation of cholesterol granulomas. This process occurs rarely and morphologically gives rise to an unusual entity, that is why this case is being reported. Additional studies based on experimental model may improve our understanding of this entity.

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