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RESEARCH ARTICLE

MULTILOCULAR PERITONEAL INCLUSION CYSTS: A REPORT OF TWO CASES

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ABSTRACT

Multilocular peritoneal inclusion cysts (MPICs) are neoplasm composed of multiple small cysts filled with serous fluid. It is well defined but rare entity, predominantly in women of reproductive age group. MPICs are cysts of peritoneum or postoperative peritoneal cysts, benign cystic mesothelioma, inflammatory cyst. They have been referred to as multilocular peritoneal inclusion cysts (MPICs). Herewith we are reporting two cases of MPICs, both were belonged to reproductive age group and clinically manifested by lower abdominal pain with palpable mass. So clinically diagnosed as complex ovarian cysts. Final diagnosis as MPICs is made on histopathological examination

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INTRODUCTION

Multilocular peritoneal inclusion cysts are usually occurs in reproductive age group (Julie *et al*., 2013). They occur multiple, small thin walled, translucent cysts in peritoneal cavity. Clinically and intraoperatively these cysts may mimic a cysts of ovarian tumor. Its origin and pathogenesis are uncertain but etiopathological factors like an abdominal surgical procedure, pelvic inflammatory disease or endometriosis are considered.so the term multilocular peritoneal inclusion cysts (MPICs) is preferred to benign cystic peritoneal mesothelioma or benign multicystic peritoneal mesothelioma (Julie *et al*., 2013). Surgical complete removal of the tumor is the best treatment strategy for MPICs (Santosh Shetty, 2014). It has shown no evidence of malignant transformation but as many as 50% of lesion have recurred locally. Probably due to newly formed postoperative adhesions (Santosh Shetty *et al*., 2014 Baddoura *et al*., 1980).

Herewith we are reporting two cases of MPICs

Case1: A 29years multipara female came with complaints of abdominal distension and mild pain of one year duration which was clinically diagnosed as complex ovarian cysts.

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She had also history of tubectomy 2 years back. Ultrasonography showed multicystic large intraperitoneal mass occupying entire lower abdomen. There was no connection of the mass and other viscera. Fine needle aspiration cytology of this mass was done which revealed yellowish serous fluid. Microscopically it showed benign mesothelial cells. Routine biochemical and haematological examinations were within normal range. Laparotomy was done and large multicystic mass was found which was occupying lower abdomen and loosely adherent to the uterus, fallopian tubes and ovaries, small and large bowel, and urinary bladder. The entire mass was resected and sent for histopathological examination. On gross examination, specimen was consisted of yellowish watery fluid filled multilocular cystic gelatinous mass measuring 15x12x10cm in diameters (Fig. 1). External surface was smooth.

On cut open multilocular cysts of varying size, filled with serous fluid were noted. One area showing small papillary excrescences was seen. Microscopically, multiple sections from cyst wall showed thin fibrovascular wall lined by single layer of cuboidal to flattened mesothelial cells At places papillary structures lined by benign cuboidal mesothelial cells are also noted. There was no evidence of dysplasia or malignancy. (Fig. 2, A and B) Diagnosis of Multilocular peritoneal inclusion cysts was made. After surgery patient had no any complaints.

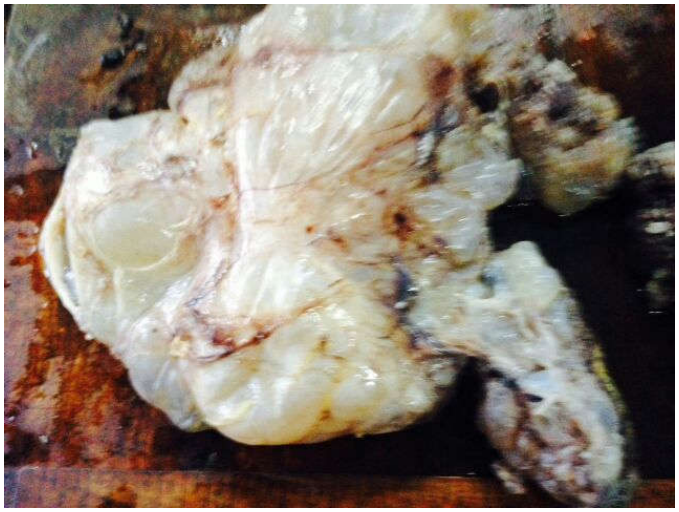
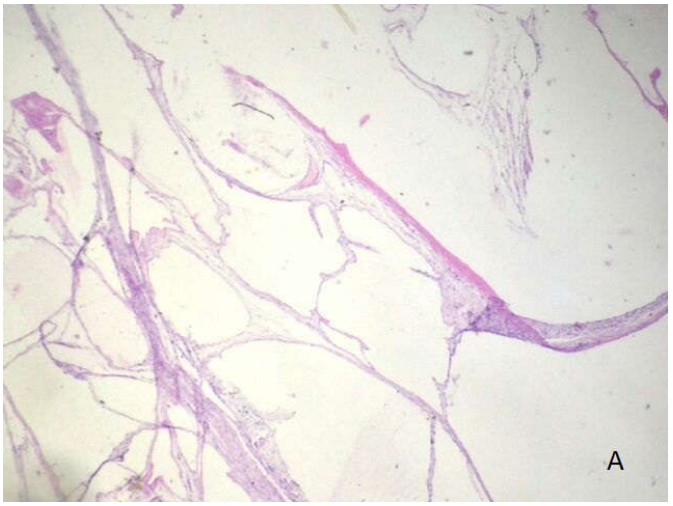


Figure 1. Photograph of gross specimen of MPICs with smooth gelatinous external surface



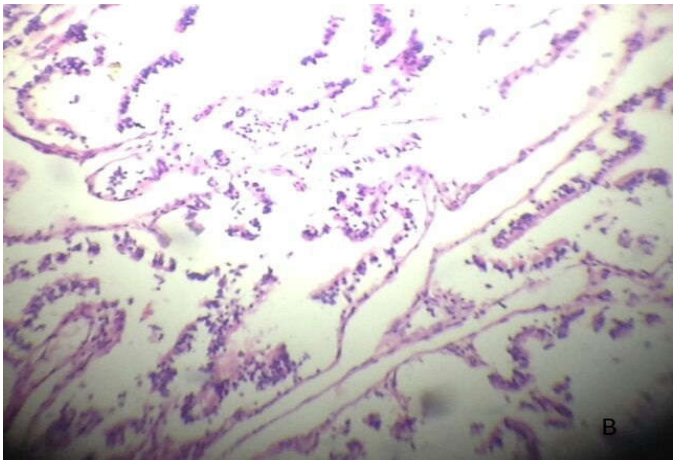
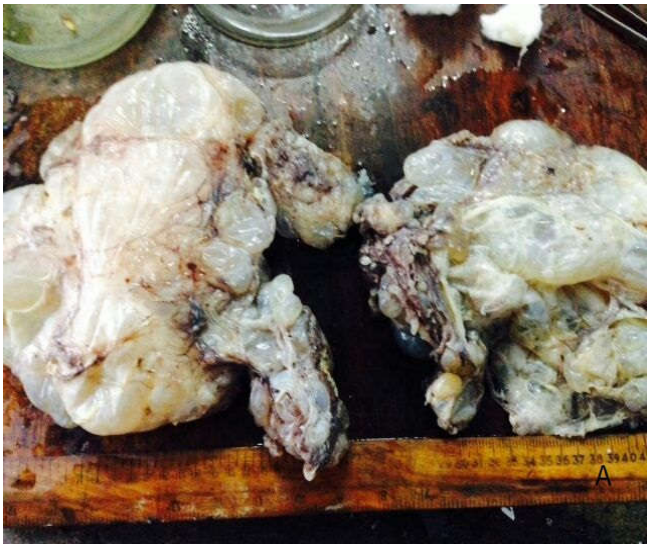


Fig. No. 2A showing - Photomicrograph of MPICs showing thin fibrous wall lined by flat mesothelial cells and fig.no.2 B showing at places papillae formation

Case report 2

A 32 years multipara female with vague complaints of pain in abdomen, distension of abdomen. She had undergone tubectomy surgery 06 years back. On clinical per abdominal examination, clinician felt a large cystic mass in right iliac quadrant extending into hypogastric region and similar mass was also felt in left iliac quadrant.



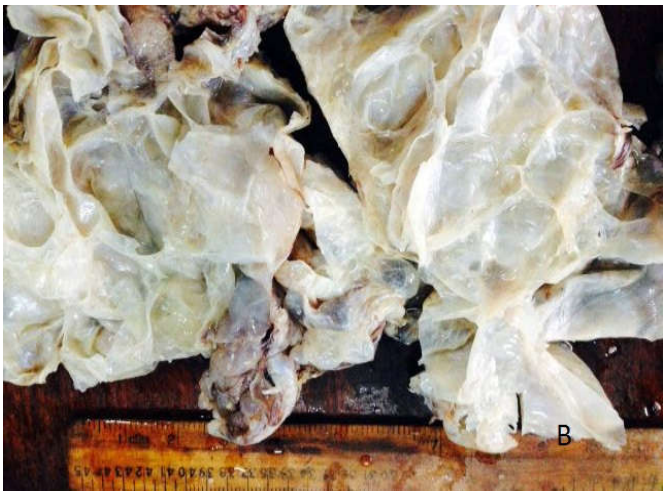
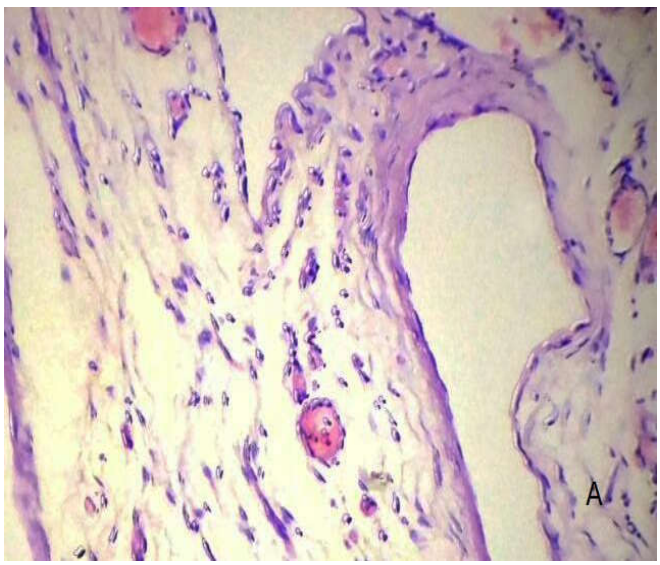


Fig. No. 3. A - Photograph showing gross appearance of Multilocular cystic mass. B showing cut surface with multilocular serous fluid filled cystic mass



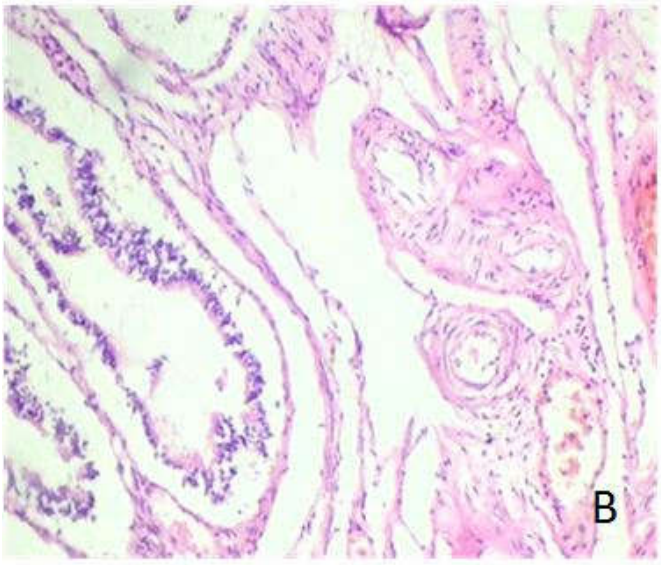


Figure 4. Photomicrograph. A, showing fibrovascular cyst wall with sparse mononuclear cell infiltration. B, showing cyst wall lined by flat mesothelial cells with at places papillae formation

Ultrasonography revealed a large multicystic mass occupying lower abdomen and probably arising from peritoneum not attached to any abdominal solid organs. So clinically and radiologically it was diagnosed as multicystic tumour suggestive of angiolymphangioma. Routine biochemical tests and haemogram were normal. Patient underwent laparotomy surgery. Removed a large multicystic gelatinous mass measuring 40x35x10 cm, submitted for histopathological examination. (Fig.no.3 A and B). Cells, with focal multilayering and papillary infoldings. Cyst wall and septae were composed of fibrovascular connective tissue with sparse mononuclear cell infiltration (Fig.no.4). So final diagnosis of MPICs was made.

DISCUSSION

In 1889, Henk reported a case of Multiple cystic lymphangioma like tumor (Baddoura, 1980). Baddoura Smith and Mennenmyer, 1979 reported the first case of benign cystic mesothelioma of peritoneum (BCM), since then about 146 cases reported until 2009 (Tian Bao Wang) The designation "multilocular peritoneal inclusion cysts" (MPICs) is preferred to benign cystic mesothelioma. Postoperative follow up, local recurrence is common in 50% cases which are probably due to newly formed postoperative adhesions (Julie, 2013) MPICs are rare benign lesions with very rare malignant transformation. Peritoneal inclusion cyst is single or multiple cystic tumor arising from peritonium of pelvic organs. Symptoms like abdominal pain, tenderness and distension with lower abdominal mass are common findings. These presentation are not specific so preoperative diagnosis is not possible. This lesion is common in women with reproductive age group in 80% of cases with an average age of 34years (Tian Bao Wang; Bhandarkar *et al*., 1993 and Raafat, 1988). There have been only 20 cases reported in males. It is believed that female sex hormones play a role in its pathogenesis (Santosh Shetty *et al*., 2014). Radiological investigation like ultrasonography and computed tomography are useful. Radiologically guided aspiration cytology is also very useful as it reveals benign mesothelial cells. With the clinical, radiological and cytological modility the differential diagnosis are peritoneal cystic lymphangioma, pseudomyxoma peritoni and cystadenoma of ovary. In the present study, both cases of MPICs were female of reproductive age group with history of tubectomy. Previous abdominal surgery like oopherectomy, endometriosis and colonic adenocarcinoma are considered as a risk factor for MPICs (Gonzalez-Moreno *et al*., 2002; Gonzalez-Moreno *et al*., 2003 and Nozawa *et al*., 2000).

As MPICs are extremely rare lesion, without histopathological examination definitive diagnosis is difficult. On microscopy it reveals multiple cysts composed of thin fibrovascular septae lined by single layer of flattened or cuboidal mesothelial cells. Special stain like Masson trichrome shows absence of muscle fibre in MPICs. Presence of muscle fibres is demonstrated in cystic lymphangioma. Cystic lymphangioma consists of cyst filled with chylous fluid and walls show lymphoid aggregates, smooth muscles. On immunohistochemistry cystic lymphangioma shows CD2- 40 positive (Smith, 1979 and Raafat, 1988). Mesothelial cells show positive expression for calretinin and negative expression for CD2- 40, CD 31 and CD 34 (Tian Bao Wang and Raafat, 1988).

On electron microscopy, MPICs shows the characteristics of mesothelial cells with slender microvillous on the luminal surface of the cells, desmosomes, intracytoplasmic intermediate filaments, endoplasmic reticulum and mitochondria (Smith, 1979 and Raafat, 1988). Immunohistochemistry shows strong staining for cytokeratin in the cystic lining and for vimentin in subepithelial cells (Raafat *et al*., 1988). Increased level of serum CA 19-9 concentration in cases of MPICs have been reported (Tian Bao Wang and Gonzalez-Moreno, 2002). Malignant mesothelioma is known to develop after exposure to asbestos and is frequently metastasising, fatal tumour. In contrast to this, MPICs is benign and very rarely associated with asbestos exposure (Santosh Shetty, 2014). Occasional MPICs have been immunoreactive for estrogen and/or progesterone receptors (Sawh *et al*., 2003). This tumors respond to treatment with gonadotropin releasing hormone (GnRH) agonist (Sawh *et al*., 2000).

Surgical enbloc removal of the MPICs is the best treatment. Its complication like recurrence of cysts is more common is about 50%. Because of malignant transformation is very rare, adjuvant chemotherapy or radiotherapy are not required. Hormonal therapy, sclerotherapy and thermotherapy have not been proven useful treatment. In all cases of MPICs post-surgical follow up including physical examination and imaging studies is done. In MPICs prognosis is excellent [(6)].

Conclusion

Multilocular peritoneal inclusion cysts (MPICs) is rare benign tumor with high local recurrence rate. Enbloc removal and post-operative follow up should be done.

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