CASE REPORT

Benign Cystic Mesothelioma of Uterus: An Unusual Cause of Pelvic Pain

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Introduction

Benign cystic mesothelioma also known as peritoneal inclusion cyst is a rare tumour which was first reported in 1979. It can occur at any abdominal peritoneal surface, and till date only 140 cases have been reported from round ligament, mesentery, peritoneum, adnexa, etc. [1, 2]. Most common presenting symptom is pain, and surgery forms the definitive management in most of the cases.

Case Report

A 40-year-old para 4 woman presented to our OPD with history of pain abdomen for 6 years which was progressive. The pain initially was mild discomfort in lower abdomen, but for the last 2 years pain was continuous and

moderate in intensity. Pain was controlled on NSAIDs and opiates, interspersed with episodes of severe pain needing hospitalisation. Her menstrual cycles were regular although flow was heavy. On examination, patient was moderately anaemic with stable vital parameters. Abdominal examination revealed only suprapubic tenderness on palpation. Cervix and vagina were healthy on per speculum examination. Per vaginum examination revealed a normal-sized retroverted mobile and tender uterus. Both fornices were free and tender. All routine investigations were within normal limits. Ultrasonography revealed left-sided ectopic hypoplastic kidney and mild splitting of pelvicalyceal system in right kidney. Liver, pancreas and gall bladder were normal. There were suspected endometriotic changes in uterus with peripheral myometrial cysts with pelvic congestion. Endometrium was 11 mm in thickness and echogenic. Her contrast-enhanced CT scan revealed that left kidney was ectopic and malrotated and there were concretions in right kidney. Uterus was retroverted, normal size, and few small cysts were present at the periphery of the uterine margins with normal adnexa. No free fluid was present in POD. Suspicion of endometriosis was expressed. Her urologic evaluation and upper GI endoscopy were normal. Clinical diagnosis of endometriosis was made, and laparoscopic-assisted vaginal hysterectomy was performed.

On laparoscopy, uterus was normal size, mid-position with a small intramural fibroid of 2×2 cm on posterior wall. To our surprise, there were 9–10 tense clear cysts of

Fig. 2 Cyst lined by cuboidal epithelium

2–2.5 cm in size on posterior serosal surface of uterus (Fig. 1). There was no such cyst on adnexa or on any other site. Patient recovered well and is asymptomatic in follow-up.

Histopathological examination revealed chronic cervicitis, proliferative endometrium and a small leiomyoma in uterus. There were multiple mesothelial inclusion cysts near the serosal aspect of uterus. On histopathology, cyst was lined by cuboidal epithelium (Fig. 2). Diagnosis of benign cystic mesothelioma was further confirmed by immunohistochemistry. The specimen slides were strongly positive for mesothelin and calretinin stains. This report confirmed the diagnosis of benign cystic mesothelioma on the serosal surface of uterus (Figs. 3, 4)

Fig. 1 Gross appearance of benign cystic mesothelioma

Discussion

Review of the literature revealed only four cases reported earlier from surface of uterus [3]. Benign cystic mesothelioma is considered as a developmental disorder, but there is some controversy regarding its origin. The other school of thought considers it as either reactive or neoplastic. It seems to be associated with previous abdominal surgery, pelvic inflammation and endometriosis [1, 4]. It predominantly affects women of reproductive age group. Ultrasonographic picture of benign cystic mesothelioma is multicystic, vascular lesion without calcifications. There is a controversy over the presence of oestrogen and progesterone receptors in these tumours. Few authors believe that it is stain negative for oestrogen and progesterone receptors and there is no sex hormone dependence [5]. Other authors disagree and demonstrated hormone receptors on peritoneal surfaces of the tumour. Point in favour of hormone dependence is that it typically occurs in women of reproductive age group and there are reports of increase in size of tumour after administration of gonadotropins during IVF

Fig. 3 Cyst-lining cells positive for mesothelin

Fig. 4 Cyst-lining cells positive for calretinin

cycles. In fact some authors had reported management of such tumour by GnRh analogues. This tumour can be distinguished from other entities by demonstration of immunohistochemical staining specific to mesothelial cells, such as calretinin and cytokeratin 5/6.

As the tumour itself is very rare, there is no definitive protocol for treatment yet. Various modalities have been

used depending on the presentation. It can be monitored by serial ultrasound for the size of cyst in case the woman is asymptomatic. Surgical excision is the definite treatment. Aspiration of cyst has been tried for solitary lesions. It may provide temporary relief, but eventually these tumours become symptomatic again [5]. Most authors consider it a benign tumour with negligible recurrence and malignant potential [5]. Contrary to that, there are some reports of recurrence as high as 27 % and even malignant transformation of these tumours is reported [1]. Gynaecologists should be aware of this entity while evaluating any pelvic pain in women of reproductive age group.

Compliance with Ethical Standards

Conflict of interest None of the authors had conflict of interests.

Ethical Approval This article does not contain any studies with human participants or animals performed by any of the authors. It is a case report, and informed consent was obtained from subject and parents before reporting.

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