CASE REPORT

Primary mucinous cystadenoma of the retroperitoneum: a case report

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Introduction

Mucinous cystadenomas and cystadenocarcinomas of the ovary are well-recognized and common tumours, both clinically and histopathologically [1]. However, Primary mucinous retroperitoneal cysts (PRMCs) are exceptionally rare tumours, with only fifty-five cases documented in the English literature up to 2018 [2,3]. They usually lie in the retroperitoneum without any connection to abdominal organs except areolar tissue. The first description of a primary retroperitoneal mucinous cystadenoma (PRMC) was provided by Handfield-Jones in 1924, in his study on retroperitoneal cysts [4].

The precise origin of PRMCs remains uncertain, though hypotheses suggest that they may arise from ovarian tissue or mesothelial cells undergoing metaplasia [1,2]. Generally, these PRMCs are asymptomatic and are found incidentally during routine assessment [2]. This is because the retroperitoneum is a large potential space and therefore they can become considerably large before becoming symptomatic. Accurate preoperative diagnosis is challenging due to the lack of established diagnostic methods. Preoperative radiological diagnosis of benign nature is useful though not essential [5]. Delayed diagnosis and treatment can be life-threatening, as complications such as rupture, infection, recurrence or malignant transformation may occur despite the benign nature of these cysts [1]. The exact diagnosis of PRMC is predominantly depend on pathological rather than radiological findings, though radiological imaging may suggest malignancy especially in the presence of calcification or enhanced mural nodules [4,1]. This emphasis the need for surgical intervention for both diagnosis and treatment [2]. Radical resection either through laparotomy or laparoscopy is the preferred treatment approach. Surgery, not only confirm the diagnosis but also prevent complications mentioned above. We report a case of a 28-year old female in whom excision of a PRMC was successfully performed via open laparotomy.

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Case presentation

A 28-year old female presented to our GI surgical clinic with progressive abdominal distension over four months duration. There was no abdominal pain. Her bowel habits were regular and there were no urinary symptoms. Her appetite was normal and there was no recent unintentional loss of weight. She had regular menstrual cycles. Her past medical and surgical history was unremarkable.

She was of average build and there were no nutritional deficiencies. Abdominal examination revealed a fullness mainly on the right side of the abdomen. There was no tenderness or organomegaly. Digital rectal examination was normal.

Routine haematological and biochemical examinations were within normal limits. Trans abdominal ultrasonography revealed a markedly dilated right colon. Therefore, a double contrast barium enema (DCBE) was performed as a colonic pathology was suspected and the DCBE was the only available investigation for this purpose in our institution at that time. Since it showed bowel displacement towards the left upper quadrant extra-colonic pathology was suspected therefore a contrast enhanced CT scan was performed which revealed a retroperitoneal cyst extending on either side of the abdominal aorta with an isthmus at the level of the pelvis (Fig-1a,1b).

Laparotomy was considered as the patient was symptomatic. A midline laparotomy was performed under general anaesthesia. There was a large retroperitoneal cystic lesion occupying both paracolic gutters. The cyst was completely dissected off the retro peritoneal structures starting from the right side. The right colon, caecum and the ileum were overlying the cyst was carefully mobilised starting from the right side. A peritoneal incision was made below the ileocolic pedicle and the cyst was gradually dissected off the

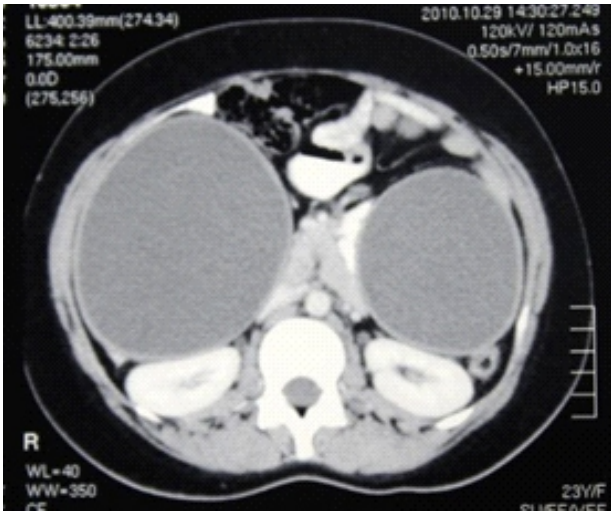

Figure 1 : A. Retroperitoneal thin walled cystic structure with bi- lobed configuration

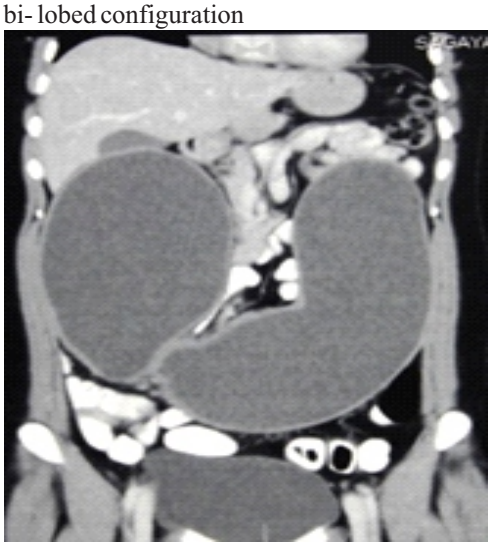

Figure 1: B. Isthmus of the cyst at the lower pole

retroperitoneum with the combination of blunt and sharp dissection. In the midline the cyst had to be carefully dissected off the aorta and IVC before moving on to the left side. As we were able to enter the correct plane at the beginning we were able to completely remove the cyst with no spillage of the cystic fluid and minimal bleeding. The duration of surgery was 3 hours and the blood loss were 400 ml. The cyst measured 40 cm in length and the widest diameter was 11 cm (Fig-2). She made an uneventful recovery and was discharged from the hospital on the 6th postoperative day. Histology of the specimen revealed cyst wall consisting of smooth muscle lined by mucinous epithelial cells. At 47 months follow up patient remained asymptomatic and there was no evidence of recurrence on follow up ultrasound scan.

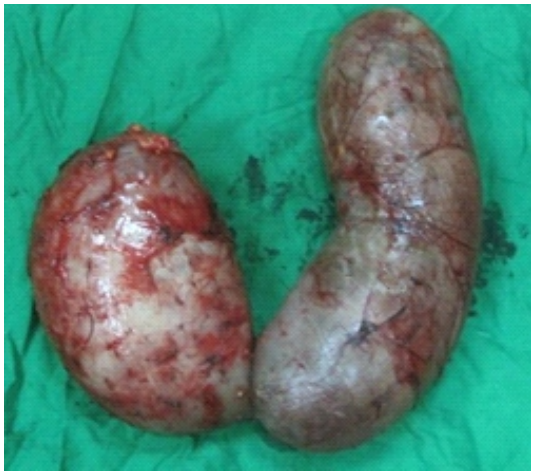

Figure 2: The mesenteric cyst

Discussion

In this patient the cyst was large enough to cause abdominal distension or discomfort. Literature shows that symptoms and signs are usually due to its huge size and compression effects [2]. Some patients have even presented with acute symptoms due to rupture though it is uncommon. Lage cysts are palpable on abdominal examination [1] as in this patient. We decided to investigate her with ultrasonography initially as this patient's clinical features were more in favour of an ovarian pathology. Since ultrasonographical appearance suggested dilated colon, DCBE was arranged as it was the only means of investigating the colon at that time in our set up. Given rarity of the condition, pre-operative diagnosis was difficult despite the appearance of DCBE showing a largely displaced right Colon. Abdominopelvic CECT scan revealed a detailed view of the retroperitoneal cyst extending from either side of the aorta with an isthmus at the level of the pelvis. It was very useful in both making the diagnosis as well as in the assessment of its extent to plan surgical management of this patient.

Complete resection of retroperitoneal mucinous cystadenoma is usually recommended whenever possible due to the concerns regarding potential malignancy or infection within the tumour and due to recurrence [2,3]. Though radical resection of the cyst can be done laparoscopically [1] we did not have facilities or expertise and our spontaneous choice was open surgery. However due to its anatomical location and close proximity to the vital structures complete excision was difficult. It was important not to allow cystic fluid spillage because of the unclear pathology of the lesion as in most of the cases [3]. In some cases, percutaneous CT-guided drainage had been performed prior to laparotomy due to the large size to minimise the cystic fluid spillage during surgery [5]. In

such circumstances, marsupialization of the cyst wall and drainage of the cyst has also been suggested as an alternative. However, it is not impossible to excise a large cyst intact even if it occupies the retro-peritoneal space on either side of the midline provided the correct tissue plans are followed as in this instance.

Retroperitoneal cysts are almost exclusively found in females [1,2,3,5] and they not only resemble ovarian cystadenomas clinically as in this patient but also histologically and macroscopically [3]. Sizes of the reported cysts vary from 7 cm to 20 cm but the cyst of this female patient was 40 cm in length and 11 cm in maximum diameter, therefore probably making it the largest PRMC reported so far [4,5].

Microscopically, these tumours are characterised by a combination of tall columnar cells and low cuboidal cells situated within a collagen-rich stroma. In most cysts neither atypia nor stromal invasion were present histologically [5]. Because of its mullerian origin, the immunochemistry for K7 and CA125 were also found to be positive in some patients (3). However, in most cases, serum Cancer antigen 125 (CA 125), Carbohydrate antigen 19-9 (CA19-9) and Carcinoembryonic antigen (CEA) were also found to be within normal ranges [1,3]. In this patient these biochemical markers were not carried out as they were not available at that time in our institution.

Most of these patients had been discharged without any complications after surgery [1,3,5]. In this particular patient, even though she had a very large cyst, probably the largest RPMC so far, excision of the cyst via laparotomy was done successfully and the recovery was uneventful.

Conclusion

Mucinous cystadenoma rare cystic lesion in the retroperitoneum. Differential diagnoses should include malignant cystic neoplasms of the ovaries as well as RPMC since they occur exclusively in females. This is especially because different surgical approach may be required in case of RPMC. These patients need to undergo proper evaluation and management preferably by a group of experts. Once the tumour is diagnosed, radical resection is indicated either through laparotomy or laparoscopy.

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Learning Points:

- Primary retroperitoneal mucinous cystadenoma is an uncommon benign cystic lesion almost exclusively found in females.
- PRMC should be included in the list of differential diagnosis in patients presenting with abdominal pain & distension.
- The recommended treatment is surgical resection.