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Two cases of ovarian mature cystic teratoma with bowel fistulation

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Abstract

Chronic diarrhoea is one of the most common reason for referral to a gastroenterology clinic, estimated prevalence of chronic diarrhoea in a Western population are 4–5%. Ovarian teratoma is a benign ovarian neoplasm, however it can be associated with various complications, and bowel fistulation is a very rare phenomenon. We hereby presented two young ladies with chronic diarrhea due to bowel fistulation complicating a benign ovarian mature teratoma.

Keywords: Two cases, mature cystic, bowel fistulation

Introduction

Benign cystic teratomas or dermoid cysts comprise 20–25% of all benign ovarian neoplasms. They arise from totipotent stem cells and are neoplastic sequelae from a transformed germ cell. Histologically, benign teratomas are composed of mature cells, usually from all three germ layers, and often contain sebaceous fluid and hair [1].

The clinical manifestations of ovarian teratomas vary from incidentally detected small masses to malignantly transformed tumors leading to high mortality [2]. Ovarian teratomas can be associated with various complications, including torsion (16%), rupture (1%–4%), malignant transformation (1%–2%), infection (1%), and autoimmune hemolytic anemia [2, 3].

Herein, we present two cases benign ovarian dermoid cyst complicated with bowel fistula.

Case one demonstrated a 25 years old lady who presented with chronic diarrhea for 3 months with a weight loss of 5 kg. Initial colonoscopy showed non-specific inflammation over rectum only. CT enteroclysis was ordered to rule out small bowel pathology. CT abdomen and pelvis revealed two pelvic masses. One 5.3x5.9cm (WxAP) mass in right adnexa is predominantly soft tissue attenuating with internal fat densities and tooth like calcifications in its superior aspect. A second mass measures 7.9 x 7.1cm (WxAP) and contains a gas fluid level with internal fat containing mural nodule located in central pelvis superior to the uterus and lateral to sigmoid colon (Figure 1). Both masses suggests dermoid with fistulation suspected in the second mass in view of gas fluid level. Sigmoidoscopy showed a suspected fistula opening at 10cm from anal verge with hair-like structure protruding out. CA125 was 98U/mL, and CEA level was normal. Exploratory laparotomy was performed and a fistula was found between the infected left dermoid cyst and rectum. Left salpingo oophorectomy, anterior resection of rectum and ileostomy were performed in September, 2016 accordingly. Final histology reviewed benign mature teratoma, acute and chronic inflammation. Patient recovered well, and closure of ileostomy was done 3 months later.



Fig 1: Selected axial CT image shows central pelvic mass (arrows) with internal nodule (arrowhead) and gas fluid level

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Case two demonstrated a 22 years old lady presented also with chronic diarrhoea for 3 months with a weight loss for 10kg. Her body mass index was down to 14 when she was first seen by us. CT enteroclysis on 29th March 2018 showed a thick-walled 6.2 x 7.4cm (WxAP) mass in central pelvis just anterior to the recto-sigmoid and superior to uterus. The mass contains heterogeneous dependent densities with air-fluid level as well as fat attenuating areas. Multiple tooth-like densities are noted in its posterior aspect (Figure 2). Features were suggestive of a dermoid cyst with fistulation with sigmoid colon in view of internal gas densities. Intraoperatively, a fistula tract was noted between the right ovarian cyst and rectum (Figure 3). Laparotomy, right salpingo oophorectomy, and anterior resection of rectum were performed in April, 2018, patient recovered well and final histology showed mature cystic teratoma.

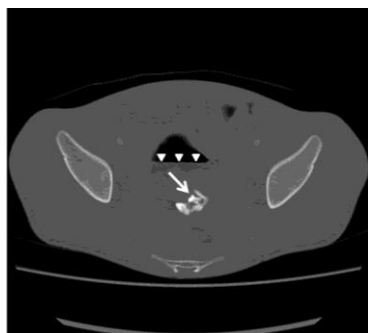


Fig 2: Selected axial CT image in different window highlights gas fluid level (arrowheads) and tooth-like calcifications (arrow)



Fig 3: Gross specimen demonstrating the dermoid cyst with teeth-like content and bowel fistulation

Discussion

Chronic diarrhoea is one of the commonest reason for referral to a gastroenterology clinic, estimated prevalence of chronic diarrhoea in a Western population are 4–5%. Most commonly encountered causes for chronic diarrhoea would be irritable bowel syndrome (IBD), bile acid diarrhea, dietary related, colonic neoplasia, inflammatory bowel disease, and drug related^[4].

Benign ovarian dermoid cysts are usually asymptomatic but occasionally complicate into ovarian torsion (16%), peritoneal rupture (1–4%), malignant transformation (1–2%) infection (1%), invasion into adjacent viscera and autoimmune hemolytic anemia (< 1%), producing variable symptoms^[5-10].

Fistula formation is a rare complication of dermoid cysts. Urinary bladder is the most frequent organ involved in fistula formation and malignant transformation is not a prerequisite for fistula formation. Shiels *et al.* reported a case of entero-dermoid cyst fistula, and postulated that a small leakage from the cyst caused a dense adhesion between the cyst and the bowel. Circulatory disturbance in the wall of these structures with resultant necrosis and inflammatory response were probably responsible for the fistula formation^[11].

As in the presented cases, computed tomography (CT) remains the modality of choice for complete evaluation of these lesions. Diagnosis can be straightforward at CT scan with the recognition of fat attenuation and calcifications within an adnexal mass. CT scan can also help to delineate the mass in relation to the adjoining viscera and to identify various associated complications^[11, 12].

Kizaki had reviewed 17 cases of ovarian mature cystic teratoma with fistulation. Of the 17 cases, malignant transformation was the cause of fistulation in 4 of the cases, whereas the remaining was due to inflammatory process. The mean age for the four cases of malignant transformation was 66, while the mean age for the benign group was 35. For those with bowel fistulation, the most common presentations were diarrhea, rectal bleeding, abdominal pain and weight loss^[13].

The lesson to learn in our two cases was that, in a young lady with image finding of a dermoid cyst presenting with gastrointestinal symptoms, one has to bear in mind the remote possibility of ovarian cyst complication leading to the gastrointestinal manifestation.

Conflict of interest

Authors declare that there is no conflict of interest.

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