LETTER TO THE EDITOR

Benign mesothelioma of the appendix: an incidental finding in a case of sigmoid diverticular disease

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Benign multicystic mesothelioma is a well recognised but rare entity. The aim of this report is to describe a case of a small mesothelial proliferation of the peritoneum. A 58 year old postmenopausal woman presented with left sided abdominal pain and altered bowel habit. Radiological investigations (barium enema and computed tomography scan of the abdomen and pelvis) were undertaken. An operation was performed for symptomatic sigmoid diverticular disease. Unusually, the appendix was adherent to the sigmoid colon. Microscopy revealed a benign mesothelioma. The patient remains symptom free to date.

A 58 year old, female, ex-smoker was admitted via the accident and emergency department because of sudden onset left sided lower abdominal pain and altered bowel habit. She had been constipated for two weeks and reported losing about 3 kg over the past three months. Her past medical history included ischaemic heart disease, hypertension, and peripheral vascular disease.

Examination on admission revealed left iliac fossa tenderness and vaginal bleeding.

A pelvic ultrasound showed no ovarian abnormality or ascitic fluid, effectively excluding the clinically suspected possibility of an ovarian cyst.

A barium enema was performed and demonstrated "moderate diverticular disease within the sigmoid colon, but no other significant pathology". Fever after the radiological examination prompted an abdominal computed tomography scan, which revealed "a 4 × 2 × 2 cm fluid density collection in the right hemi pelvis with a thin enhancing wall and some internal septations but no significant intra-abdominal collection of fluid".

About five months after initial presentation, a left hemicolectomy and appendicectomy was undertaken. The surgical findings were "sigmoid colon adherent to the left pelvic side in association with significant thickening of the bowel wall. The appendix tip was involved in this inflammatory area."

PATHOLOGICAL FINDINGS

The appendix and sigmoid colon were sent separately.

Macroscopy

The appendix appeared unremarkable.

The segment of sigmoid colon measured 255 mm in length with a congested serosal surface.

The bowel wall was greatly thickened (up to 8 mm), with numerous apparent diverticulae present along the length of the specimen. No focal lesions were identified.

Figure 1 Benign mesothelioma of the appendix: lesion displaying the interweaving papillae comprising bland uniform cuboidal cells. Occasional psammoma bodies are seen. Original magnification, ×40.

Figure 2 Benign mesothelioma of the appendix: lesion arising from the serosal surface of the appendix with unapparent traditionally described cystic structures. Inset: Immunohistochemical positivity for the calcium binding protein calretinin. Original magnification, ×10.

Microscopy

The appendix itself was unremarkable. However, arising from the serosal surface was a lesion composed of uniform bland cuboidal cells with indistinct cell borders forming papillae and ill defined tubular structures.

Abbreviation: BMMP, benign multicystic mesothelioma of the peritoneum

Table 1 Immunohistochemical profile of the lesional cells

Positive	Negative
Calretenin	CEA-M
CK7	Ber EP4
CK5/6	CK20
	CD68

CEA-M, carcinoembryonic antigen (membrane staining); CK, cytokeratin.

Examination of the sigmoid colon confirmed diverticular disease. There was no evidence of diverticulitis, cytological atypia, or neoplasia. A lymph node sampled showed reactive changes only.

Immunohistochemistry

Table 1 shows the immunohistochemical profile of the cells making up the lesion adherent to the serosal surface of the appendiceal wall.

The findings in this case were those of a small benign mesothelioma.

DISCUSSION

Benign mesothelioma is a rare but recognised entity referred to in the literature as benign multicystic mesothelioma of the peritoneum (BMMP),¹ and is also known as peritoneal inclusion cysts.

A rare lesion, it mainly arises from the serosal surfaces of the ovary, uterus, bladder, and rectum. A single case report of an identical lesion in the pleural cavity has been reported.²

McFadden and Clement undertook a clinicopathological analysis of six cases of BMMP.³ All six cases had several common features: all occurred in women (age range, 15–51 years; median, 37) presenting with abdominal symptoms, who had invariably in the past undergone gynaecological surgery. However, all these cases were seen in association with one or both ovaries. Previous exposure to asbestos was absent in all cases.

As far as we are aware, this is the first case of an incidental mesothelioma seen in a patient in association with diverticular disease of the large intestine. A literature search (using the NCBI PubMed database) revealed only three other case

Table 2 The likelihood (probability) of a benign mesothelioma with the immunohistochemical profile found in our patient occurring by chance alone

Immunohistochemical stain	% Positivity	
Calretinin	97	Probability: 15.5%
CK5/6	80	
CK7	20	

CK, cytokeratin.

Table 3 The probability of CK5/6, CK7, and calretinin arriving at a diagnosis of benign mesothelioma

Diagnosis	CK5/6	CK7	Calretinin
Benign mesothelioma	80%	74%	84%

The probability of the above IHC profile for the entity "benign mesothelioma" is 50%.
CK, cytokeratin.

Take home messages

- We describe the case of an incidental benign mesothelioma on the serosal surface of the appendix occurring in a patient with diverticular disease who presented with sudden onset acute abdominal pain
- Immunohistochemistry using these "newer" markers should be used only as an adjunct in arriving at a final diagnosis
- Newer markers to confirm mesothelial origin may be no more robust than older ones and further studies are required to provide more precise markers

reports of BMMP associated with the appendix.^{4–6} Two of these case reports were in middle aged women. The first was in a 53 year old woman presenting with abdominal pain, where laparoscopy revealed a 7 cm retroperitoneal mass close to but not involving the caecal serosal surface.⁴ The second was a small cystic mass involving the visceral and parietal layers of the peritoneum in the appendiceal region in a 40 year old woman with clinical signs of acute appendicitis.⁵ The third report was of a 28 year old man presenting with the symptomatology of acute appendicitis, where a 25 cm cystic appendiceal mass was found.⁶ Only an estimated 17% of BMMPs occur in men.

"As far as we are aware, this is the first case of an incidental mesothelioma seen in a patient in association with diverticular disease of the large intestine"

In our case, cystic structures were not seen microscopically. The architecture was tubulopapillary and occasional laminated concretions (psammoma bodies) were seen. We suspect that the examined lesion was only part of a larger mass. Clinically, the symptoms appear to have been mostly related to the concomitant diverticular disease of the sigmoid colon, with symptoms of acute appendicitis itself being absent. It is probable that this finding was entirely incidental.

This lesion was negative for CD68, a marker mostly positive in an entity called nodular histiocytic/mesothelial hyperplasia. This entity, which comprises an admixture of mainly histiocytes and some mesothelial cells, can be mistaken for a mesothelioma or carcinoma by the unwary.

Table 2 shows the likelihood (probability) of a benign mesothelioma with the above immunohistochemical profile occurring by chance alone (DM Frisman. ImmunoQuery; www.ipox.org).

Taking a different perspective and looking at this panel of markers in arriving at a diagnosis of benign mesothelioma, table 3 shows the results obtained for a benign mesothelioma (DM Frisman. ImmunoQuery; www.ipox.org).

These results show that current markers are unsatisfactory for confirming the mesothelial nature of a neoplasm.

Whether BMMPs are reactive or neoplastic in nature remains unresolved.⁷

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