Epithelial Inclusion Cyst of the Cecum

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Epithelial inclusion cyst of the cecum (EICC) is a rare lesion. It is considered a sequestrated cyst, either of congenital or acquired origin. As a congenital lesion, it is related to misplaced ectodermal elements at the time of neural groove closure or when ectoderm and entoderm coalesce. As an acquired cyst, it may follow implantation of cutaneous tissue after appendectomy, laparoscopic surgery or needling.

A 66-year-old man presented with acute right lower quadrant abdominal pain, which was interpreted atypical for acute appendicitis. A suberosal/muscular cystic mass in the cecum without luminal communication but accompanied by tiny pockets of free air and acute inflammatory changes in the right colonic gutter was noted on computed tomographic scan with contrast. On exploration, no obvious perforation of any viscus but a tracking of purulent material along the right pericecal gutter was found. With a palpation finding of a soft mass in the cecal wall and low suspicion for malignancy, only a short segment ileocecectomy was performed. The patient had uneventful postoperative follow-up.

The lesion microscopically was lined by stratified squamous epithelium, contained ample keratinous material, and associated with a granular cell tumor wall in the subepithelial soft tissue and surrounding smooth muscle. Special studies (periodic acid-Schiff and immunostaining with neuron specific enolase, S-100, lyzosymes, desmin and vimentin) affirmed a reactive histiocytic nature of the granular cells.

The differential diagnosis of EICC includes dermoid cyst and cystic teratoma that usually demonstrate other skin structures and embryonic tissue elements, respectively, which were absent in our case. A literature review is also presented.

INTRODUCTION

Epithelial inclusion or epidermoid cyst (EIC) lined by well defined stratified squamous epithelium enlarges by cellular proliferation and by desqua-
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A CASE TO REMEMBER

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Figure 1. The CT scan with contrast exhibits a distinct, well circumscribed subserosal muscular cystic lesion in the cecum (arrow), showing no communication with the cecal lumen.

histogenesis is controversial (1–5). An instance of ruptured epithelial inclusion cyst of the cecum (EICC) presenting clinically as an acute abdomen is reported.

CASE REPORT

A 66-year-old, diabetic and hypertensive male, was admitted complaining of an acute right lower abdominal pain accompanied by high-grade fever with chills, diaphoresis and nausea without vomiting. There was no history of prior surgical operation. Physical examination of the abdomen demonstrated right lower quadrant tenderness with localized involuntary guarding and fullness. Obturator and Rovsing maneuvers were negative. Laboratory findings were not contributory. Abdominal computed tomography scan with contrast revealed a hypodense circumscribed mass (Figure 1) juxtaposed to the cecum with an undistended appendix along with tiny air bubbles dispersed periceally. Subsequent exploratory laparotomy disclosed an ill-defined collapsed cavitary structure incorporated to the lateral wall of the cecum. The cyst had apparently ruptured concealed by fatty tissue and thick fibrinous exudative reaction extending into the adjacent cecal and appendiceal serosal surfaces. A short segment terminal ileo-cecectomy was performed.

On pathologic examination, the cystic cecal mass measured $2.5 \times 1 \times 1$ centimeters and was adherent to the anti-mesenteric side of the cecum. On cut-section, it was unilocular, containing a fair amount of white gray sebaceous material and appeared to be subserosal/muscular in location, having no communication with the intact intestinal mucosa (Figure 2).

Microscopically, the cyst contained lamellated keratin adherent to a lining of well-formed, mature, stratified squamous cell epithelium with a prominent granular cell layer, not continuous with the normal intestinal mucosal epithelium (Figure 3). No dermal adnexal structure was seen. There was also thick tumorous proliferation of granular cells in the subepithelial soft tissue and surrounding smooth muscle (Figure 4), staining brightly with periodic acid schiff (PAS) and reacting positively with neuron-specific enolase, S-100, lysozymes, desmin and vimentin immunostains, affirming a reactive histiocytic nature. A concomitant acute fibrinous pericecal peritonitis and mesoappendicitis was present.

The patient had an uneventful post-operative recovery.

COMMENTS

Epithelial inclusion cyst and dermoid cyst are considered to be sequestration or implantation cyst and not as neoplastic growth, separating from the benign cystic teratomas predominantly observed in the ovary (1). It could be acquired following implantation of epider-
Embryonic cecum relative to the closing midline neural groove and other epithelial fusion lines.

Equally remarkable are rare intestinal dermoid cysts (other than those seen in the rectum), occurring mostly in the ileocecal region (7–10). Andiran (5) has proposed that this cutaneous anlage inclusions might have taken origin while the cecum as the last part of the gut re-enters the abdominal cavity in the process of intrauterine rotation.

Pan and associates (2) have discussed some possible complications of EICC comparable to the relatively more common mesenteric cysts, including rupture, hemorrhage, necrosis, torsion, volvulus, intussusception, obstruction, peritonitis, and malignant change. The epithelial inclusion cyst in the appendix has a concomitant acutely inflamed appendix and clinically presents as such. On the other hand, our case was complicated by spontaneous rupture, resulting in a

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Figure 3. The cyst is lined by a mature stratified squamous epithelium and contains lamellated keratin (Hematoxylin eosin stain, ×100).

Figure 4. The incidental granular cell tumor in the wall of the EICC is shown (Hematoxylin eosin stain, ×100, and neuron-specific enolase immunostaining, ×100).
localized peritonitis that clinically simulated a perforated acute appendicitis or appendiceal abscess. This interesting manifestation should be considered in the differential diagnosis. ■

References

Table 1
Clinicopathological characteristics of epithelial inclusion cysts of the cecum and appendix

<table>
<thead>
<tr>
<th>Reference</th>
<th>Age (years)</th>
<th>Sex</th>
<th>Clinical Presentation</th>
<th>Location</th>
<th>Size (Cent.)</th>
<th>Previous Abdominal Operation</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pan et al (2)</td>
<td>22</td>
<td>F</td>
<td>right lower abdominal mass</td>
<td>cecum, anterior wall, intramural*</td>
<td>10×7×4</td>
<td>appendectomy 12 years prior</td>
<td>right hemicolectomy</td>
</tr>
<tr>
<td>Candreviotis (3)</td>
<td>27</td>
<td>F</td>
<td>chronic right lower abdominal pain</td>
<td>cecum, medial wall, muscular</td>
<td>8</td>
<td>exploration of appendix</td>
<td>distal ileo-cecetomy</td>
</tr>
<tr>
<td>Pear and Wolff (4)</td>
<td>71</td>
<td>M</td>
<td>rectal bleeding, loose stool</td>
<td>cecum, medial wall, submucosa</td>
<td>6×3.5×3</td>
<td>appendectomy 16 years prior</td>
<td>cecectomy</td>
</tr>
<tr>
<td>Andiran et al (5)</td>
<td>8</td>
<td>F</td>
<td>periumbilical abdominal pain for 2 days</td>
<td>cecum, antimesenteric, subserosa</td>
<td>3×3×2</td>
<td>none</td>
<td>local resection appendectomy</td>
</tr>
<tr>
<td>Piserchia and Davey (6)</td>
<td>10</td>
<td>M</td>
<td>periumbilical pain localizing to the right iliac fossa over the preceding 6 months, fever, nausea, anorexia</td>
<td>tip of appendix</td>
<td>4×3×3</td>
<td>none</td>
<td>appendectomy</td>
</tr>
<tr>
<td>Current case</td>
<td>66</td>
<td>M</td>
<td>acute right lower abdominal pain, fever, chills, nausea</td>
<td>cecum, antimesenteric, subserosa/muscular</td>
<td>2.5×1×1</td>
<td>none</td>
<td>distal ileo-cecetomy</td>
</tr>
</tbody>
</table>

F – female; M – male
* specific layer not specified