Dermoid Cyst (Mature Cystic Teratoma) of the Cecum
Histologic and Cytologic Features With Review of the Literature

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The gastrointestinal tract is an unusual site for teratomas to occur. Only 4 prior cases of teratomas involving the cecum have appeared in the English literature, the latest published in 1977. To the best of our knowledge, this is the first report to describe fine-needle aspiration cytologic findings in addition to histologic features of a mature cystic teratoma involving the gastrointestinal tract. A 30-year-old man presented with right upper quadrant abdominal pain and a palpable abdominal mass. Radiographic studies identified a mass lesion inferior to the liver and in close association with the ascending colon. Fine-needle aspiration biopsy showed scant keratinous material and anucleated squamous cells. Right colectomy revealed a cystic mass in the wall of the cecum that contained keratinous material and was lined by stratified granular squamous epithelium with sebaceous glands. Clinical and pathologic features with review of the literature are presented, and the differential diagnosis for pericolic cystic masses is discussed in detail.

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The bowel is one of the unusual sites where teratomas are known to occur. To the best of our knowledge, only 4 prior case reports of teratomas involving the cecum have appeared in the English literature, the latest reported in 1977.1–4 This article reports a case seen more recently at our institution. Fine-needle aspiration biopsy and histologic features with review of the literature are presented, and the differential diagnosis for pericolic cystic masses is discussed in detail.

REPORT OF A CASE

A 30-year-old man had been suffering from intermittent right upper quadrant abdominal pain during the last several years presented with worsening pain that led him to seek medical care. On physical examination a palpable, nontender abdominal mass was identified in the patient’s right upper quadrant. Prior upper gastrointestinal barium studies performed a month earlier were normal. His past medical history was significant for occasional indigestion, gas, bloating, and diarrhea. His family history was noncontributory.

An ultrasound revealed a prominent mass inferior to the right lobe of the liver, measuring 7 × 6.34 cm, with a heterogeneous echogenic pattern and no internal echoes. The mass was separate from the right kidney. Computed tomographic scan demonstrated a nonenhancing, well-marginated mass separate from the liver with an imperceptible wall (Figure 1). It had a density similar to water and a close association with the ascending colon. The surrounding organs and tissues appeared normal, although the mass was compressing the lumen of nearby contrast-filled loops of small bowel. Additional masses, lymphadenopathy, evidence of inflammation, and abnormal fluid collections within and around the peritoneal cavity were absent.

Fine-needle aspiration biopsy was attempted, but it was not possible to aspirate fluid after multiple passes. Small flakes of tissue present in the needle were rinsed in formalin and examined as a cell block. It showed scant layered keratin material and anucleated squamous cells, which were thought to be contaminants from the skin and were reported as “nondiagnostic.”

An exploratory laparotomy identified an 8 × 6 × 5-cm mass intimately adherent to the cecum on its mesenteric border. It was not possible to dissect the mass from the cecum without risking perforation and contamination of the peritoneal cavity; therefore, the mass was resected along with the cecum, appendix, and a portion of terminal ileum. Side-to-side anastomosis of the terminal ileum and colon was then performed.

On intraoperative consultation, an 8-cm uniloculated cyst was identified with a smooth inner surface and thin wall. The cyst was filled with off-white, cheesy material (Figure 2). A touch preparation of the cyst contents revealed anucleated and benign nucleated squamous cells consistent with a benign epidermoid cyst.

The patient had an uneventful postoperative course and recovery, and was discharged to his home.

PATHOLOGIC FINDINGS

Gross Examination

The specimen consisted of a 4-cm length of cecum with an attached 3.7-cm portion of terminal ileum and appendix. A mass was readily identified firmly attached to the cecum. Sectioning revealed a uniloculated cyst measuring 8 cm with a thin uniform wall (0.1 cm thick) and containing tan to white, cheesy material (Figure 2). This material flaked away in layers. No luminal communication was found between this cyst and the cecum. The cyst had a smooth lining. No hair or other structures were found in the cyst contents.

Microscopic Examination

The cyst was lined by keratinizing stratified squamous epithelium with a granular layer (Figure 3). This was surrounded by a wall of fibrous tissue and smooth muscle continuous with the muscularis propria of the cecum. Sebaceous glands were also identified within the cyst wall (Figure 4). Mesodermal- and endodermal-derived tissues were absent. No immature elements or atypia were found. The overlying colonic mucosa was unremarkable, as were sections from the appendix and terminal ileum. Dermoid cyst/cystic mature teratoma of the cecum was diagnosed.

COMMENT

Teratomas are neoplasms comprised of cell types representative of more than 1 germ cell layer, usually all 3.
Mature Cystic Teratoma of Cecum—Schuetz & Elsheikh

They may occur at any age. Four histologic variants of teratoma are described: (1) mature teratoma, (2) immature teratoma, (3) teratoma with malignant transformation, and (4) monodermal teratoma. Mature teratoma is a benign neoplasm that is usually cystic. Structures derived from ectoderm, mesoderm, and endoderm are commonly represented. Dermoid cysts are a special form of mature teratoma in which there is predominately an ectodermal derivation. They are characteristically uniloculated cysts lined by skin, complete with special structures such as sebaceous glands, hair follicles, and teeth, and are filled with off-white, cheesy, sebaceous material. Immature teratomas are rare malignant neoplasms that demonstrate incomplete differentiation, evidenced by a fetal histologic appearance with or without obvious atypia. They have a solid or a predominately solid structure. Teratomas with malignant transformation show clear evidence of malignancy in a derivative of 1 or more germ cell components, usually consisting of carcinoma or sarcoma. Monodermal teratoma is quite rare and is characterized by highly specialized stroma. The most common forms of this variant are struma ovarii and carcinoid in the ovary.

Mature teratomas have been commonly reported in the ovaries, testes, and mediastinum, and are less commonly reported to involve various midline locations, including the sacrococcygeal area. Teratomas are rarely found in the neuraxis, spermatic cord, or gastrointestinal tract, including the floor of the mouth, rectum, sigmoid colon, appendix, and terminal ileum. A review of reported cases of gastrointestinal teratomas indicates a higher frequency in females and a greater number of cases localized to hindgut, including the terminal ileum, colon, appendix, and rectum. All 19 cases of teratoma reported in hindgut derivatives were mature, although 1 had a focus of adenocarcinoma.

Our literature review revealed only 4 cases of teratoma involving the cecum in the English literature. Patients with mature teratoma of the cecum had a wide age range at the time of presentation (1–53 years) and a mean age of 25 years. There seems to be no association between mature teratoma in the gastrointestinal tract and developmental anomalies of the spine, sacrum, and urogenital tract. This finding is in contrast to the reported associations with duplications of the gut, including enteric duplication cysts.

The pathogenesis of mature teratomas in the cecum is
unknown, although embryologic theories have been proposed to explain their origin. During embryogenesis, germ cells migrate in a path from the entoderm of the yolk sac to the gonads via the dorsal mesentery of the hindgut, and rests of totipotential cells may theoretically become sequestered along this path. This process may explain the more common occurrence of teratomas in the gonads and presacral regions, in addition to those arising along the distal gastrointestinal tract. Other authors have proposed that such teratomas may originate from totipotential embryonic rests in the left genital ridge. Cells from these rests are theoretically implanted in the cecum before rotation of the gut during embryogenesis.

To the best of our knowledge, fine-needle aspiration biopsy of a mature cystic teratoma of the gut has not been previously reported in the English literature. Previous reports of fine-needle aspiration of dermoid cysts from other sites have demonstrated various cytologic findings, including nucleated and anucleated squamous cells, hair, keratinous debris, cholesterol, calcifications (calcium deposits), and inflammatory cells. Fine-needle aspiration in our case demonstrated anucleated squamous cells and keratinous debris.

Mature teratomas of the cecum have had variable presentations, including rectal bleeding, abdominal pain with or without nausea (present case), intestinal obstruction, and nontender palpable right-sided abdominal masses. Clinically, the differential diagnosis of intra-abdominal masses near the cecum is broad. Radiographic studies can be of some value in narrowing this differential. Detailed radiographic appearances of mature teratomas in a pericolic location have not been described, although radiographic detection of calcifications in a reported case of midcolonic teratoma by plain abdominal radiography and ultrasound was helpful in considering the diagnosis of teratoma. One author suggests that if calcifications are found in a colonic tumor, the diagnosis of primary teratoma should be considered. The differential diagnosis of well-margined pericolic cystic lesions on computed tomographic scan includes mesenteric cysts (including cystic lymphangioma and mesothelial cyst), lymphatic cyst, appendiceal mucocoele, nonpancreatic pseudocyst, enteric duplication, epidermoid cyst, and cystic teratoma (Table). Due to the close proximity of intramural cysts in the bowel to surrounding structures such as the liver, the differential diagnosis may also include hepatic cyst and choledochal cyst. Epidermoid cysts are lined by mature squamous epithelium and may resemble mature teratoma grossly and histologically. Epidermoid cysts, however, lack the specialized structures of the skin, such as hair follicles, sebaceous glands, and hair follicles, characteristic of mature teratoma. The absence of specialized structures in the wall of epidermoid cysts is the most important factor differentiating epidermoid cysts from the dermoid cyst variant of mature teratoma. Three cases of epidermoid cysts have been reported in the cecum, and they constitute a distinct entity. Two of these cases occurred years after appendectomy. Previous authors have concluded that this favors implantation of squamous epithelium from the skin as the etiology for epidermoid cysts of the cecum, rather than de novo neoplasia.

Possible reported complications associated with gastrointestinal teratomas include adenocarcinoma, ulceration, rupture and bleeding, foreign body giant cell reaction secondary to keratinous contents, regional lymphadenopathy, intestinal obstruction, failure to thrive, and anemia. All 4 previously reported cases of teratoma of the cecum, as well as our case, were benign and had no immature or malignant elements.

Treatment in the present case, as with most previously reported cases in the cecum, was local resection of the cystic mass along with a variably sized length of contiguous bowel. In 1 case report, simple enucleation of a cecal dermoid cyst was accomplished without complications.

The prognosis for mature cystic teratoma is excellent, as local resection has been curative in all reported cases arising in the colon.

References