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Case Report

Caecal dermoid cyst presenting as an appendicular mucocele on abdominal imaging

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ARTICLE INFO

Article history:

Received 31 May 2019

Revised 31 July 2019

Accepted 2 August 2019

Available online 6 September 2019

Keywords:

Mature teratoma

Cecum

Ileocecal

Dermoid cyst

Appendicular mucocele

ABSTRACT

Background: Dermoid cysts are benign tumors rarely arising from the cecum. Also, including this report, 10 cases have been described in the literature, to our knowledge. Dermoid cysts are benign tumors rarely found in the cecum. A literature research has found only 9 other cases.

Case: We describe the case of a healthy 44-year-old man with no surgical history, in whom a well-defined cystic-mass was incidentally found on abdominal computed tomography, presenting as an appendicular mucocele. An hemicolectomy was performed and pathological analysis revealed a dermoid cyst of the cecum.

This paper describes the case of a healthy 44-year-old male with no prior surgical history, with a well-defined cystic mass found incidentally on abdominal computed tomography, thought to be an appendicular mucocele. After hemicolectomy and pathological analysis, this was revealed to be a dermoid cyst of the cecum.

Conclusion: Although dermoid cysts are a rare tumor of the cecum, they should be considered in the differential diagnosis of a nontender palpable mass in right lower quadrant (RLQ) or cecal mass on imaging.

Although rarely found in the cecum, a dermoid cyst should be included in the differential diagnosis of nontender palpable masses of the RLQ, or if revealed by imagery.

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Introduction

Dermoid cysts, also known as mature teratoma, are benign tumors with elements of all 3 germinal layers [1]. Although the exact origin remains unclear, the most probable hypothesis is a failure of embryologic fusion. They commonly arise from the ovary, less frequently the testis, mediastinum, sacro-

coccygeal area, and central nervous system. Nevertheless, other locations were found, as orbit, stomach, peritoneum, subdural. Mostly, patients were asymptomatic and abdominal computed tomography (CT) showed greasy substance (93%) and calcification (56%) [2].

We report the case of a healthy 44-year-old man who developed a dermoid cyst in the cecum.

Acknowledgment: Dr. Nouredine Bouzourene. He contributes the gross and microscopic examinations.

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<https://doi.org/10.1016/j.radcr.2019.08.002>

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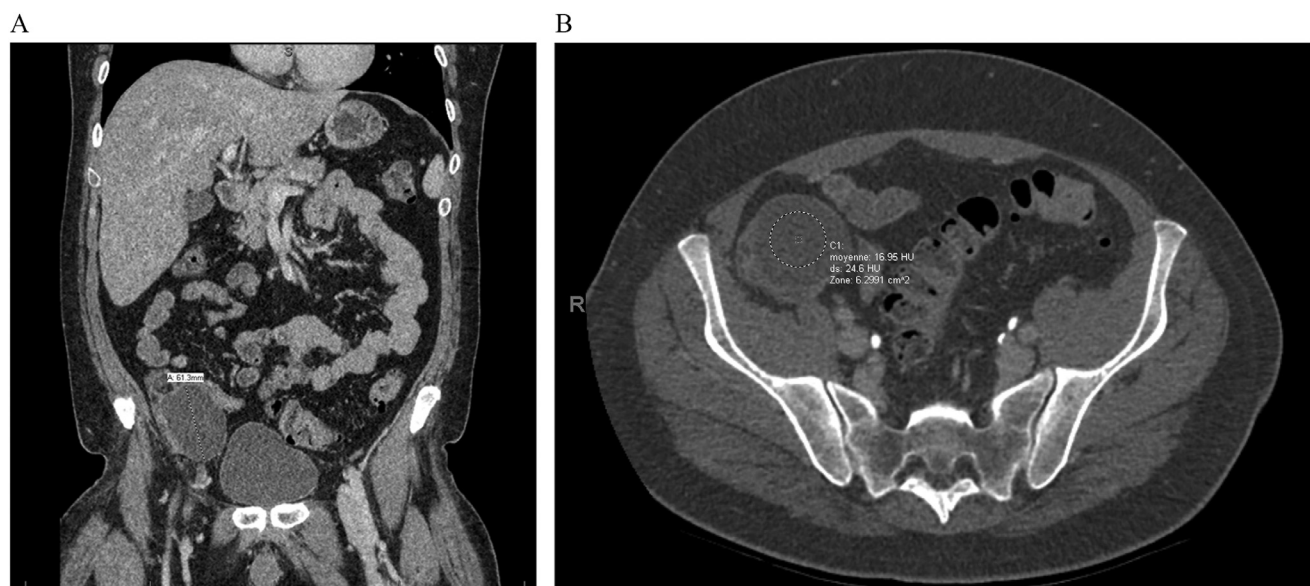


Fig 1 – (A) Dermoid cyst in the cecum of a 44-year-old man: the computed tomography pointed a well-defined cystic mass 6.1 × 4.9 × 4.9 cm. (B) The image was compatible with a mucinous lesion.

Case report

A fit 44-year-old man was referred to the general surgeon, by his urologist, after the incidental finding of a well-delimited appendicular mass on an abdominal CT, performed to investigate a 2-month history of macroscopic hematuria due to bladder varicose veins. He had no abdominal complaints and a normal clinical abdominal examination. He had no history of weight loss. There was no surgical history and no family history for cancer.

The CT (Fig. 1A) showed a well-defined cystic mass 6.1 × 4.9 × 4.9 cm, showing fluid content with no greasy substance or calcification, thin-walled, in contact with the appendix and developing on the bottom of the ileocaecal valve. The appendix is neither dilated nor filled. No lymphadenopathy was noted as well as no fluid collections within and around the peritoneal cavity. The radiological impression was that this mass was a mucinous lesion. Since it could be a mucocele, we proposed of a right hemicolectomy because of the risk of pseudomyxoma peritonei.

During exploratory laparoscopy, the topography perfectly matched with the imaging, revealing a mass which seems contiguous with the last intestinal loop and the caecal low-lying, having an extrinsic appearance. Instead of an ileocecal resection, given the risk associated with a second surgery, though it could be a pseudomyxoma peritonei, an oncology hemicolectomy was performed. In the postoperative course, the patient presented a paralytic ileus that was solved after 5 days of conservative treatment, and *Clostridium difficile* colitis for which he received adapted antibiotherapy.

Gross examination on pathological finding showed that the specimen consisted of a 19 cm length of right colon with an attached 8.5 cm portion of terminal ileum and appendix. A mass was attached to the cecum cylindrical in shape/form and

measuring 6.3 cm in length and 4 cm in diameter with smooth outer surface (Fig. 2A). Sectioning revealed a uniloculated cyst with a variable wall thickness from 0.1 to 0.5 cm and containing tan to brown, cheesy material (Fig. 2B). This material flaked away in layers. No luminal communication was found between this cyst and the cecum. The cyst had a smooth inner lining. No hair or other structures were found in the cyst contents.

Microscopic examination revealed that the cyst was lined by keratinizing stratified squamous epithelium with a granular layer (Fig. 3A). This was surrounded by a wall of fibrous tissue and smooth muscle continuous with the muscularis propria of the cecum.

The squamous epithelium was partially abraded with histiocytic inflammatory reaction. Sebaceous glands were very focally identified within the cyst wall (Fig. 3B). Mesodermal- and endodermal-derived tissues were absent. No immature elements were found. Sections from the colon, terminal ileum, and appendix were unremarkable. Dermoid cyst of the cecum was diagnosed.

Discussion

Mature teratoma might be the result of embryologic sequestration of germ cells while migrating in the path from entoderm of the yolk sac of the gonads via the dorsal mesentery. They might also be a mass derived from totipotential embryonic rests in the left genital ridge before rotation of the gut, during embryogenesis [3,4].

Dermoid cysts are a type of mature teratoma characterized by the predominance of ectodermal derivation, usually unilocated. They are known to be congenital as described above, but also as a consequence of squamous metaplasia of

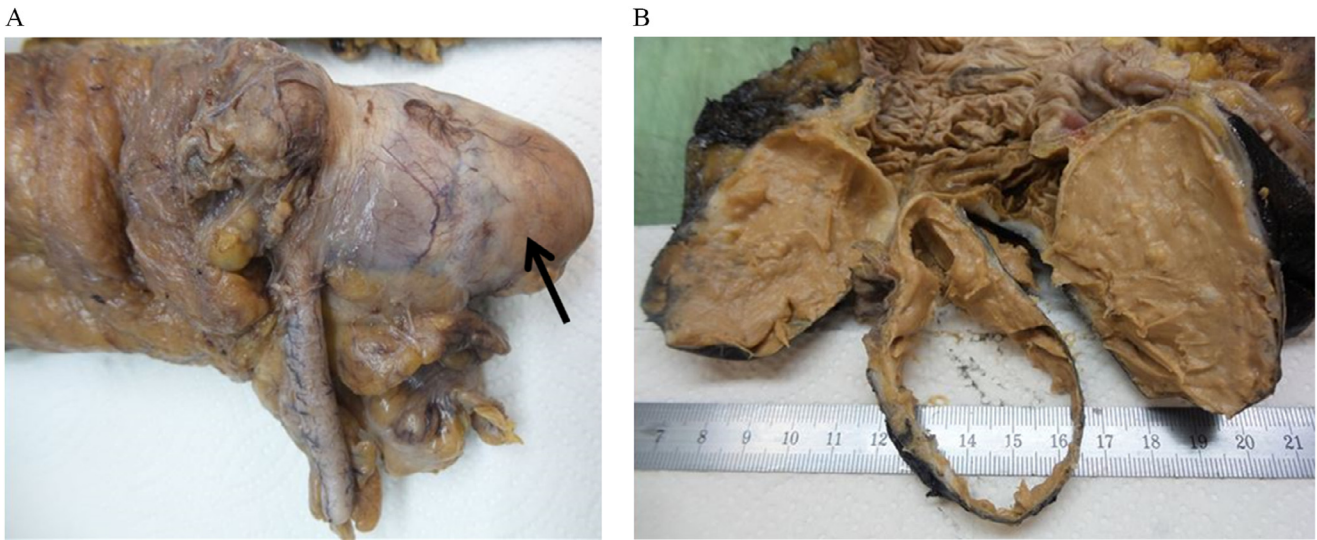


Fig 2 - (A) The black arrow identifies the dermoid cyst arising from cecum and measuring 6.3 cm in length with 4 cm in diameter. (B) The uniloculated cyst showed a variable wall thickness with tan to brown, cheesy material cyst contents.

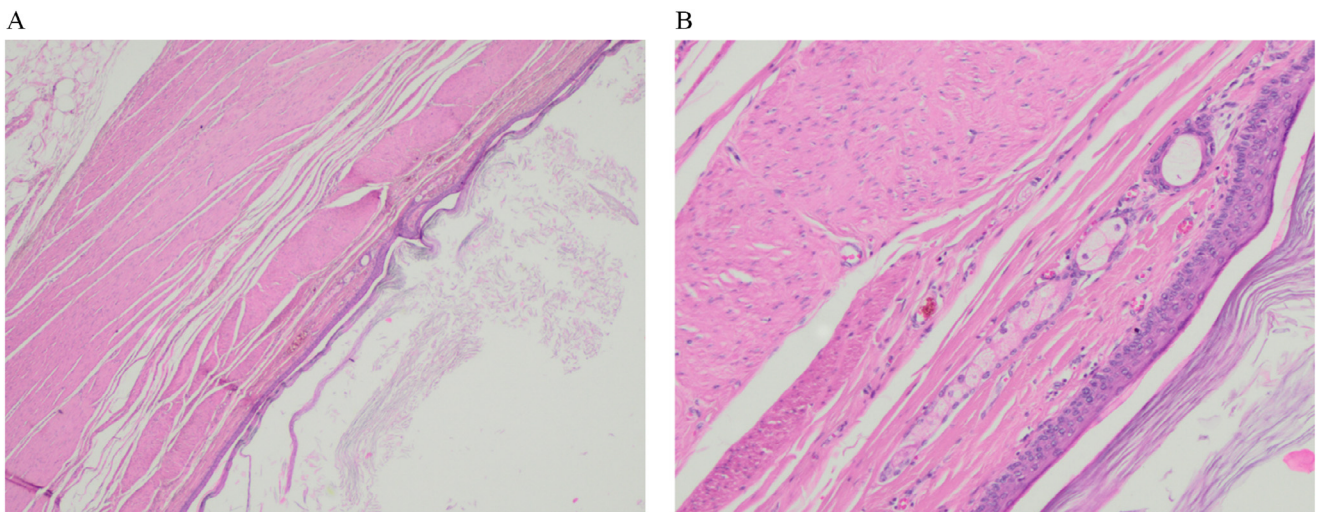


Fig 3 - (A) The cyst is lined by keratinizing stratified squamous epithelium (hematoxylin-eosin, original magnification 2x). (B) Rare sebaceous glands were identified in the wall of the cyst (hematoxylin-eosin, original magnification 10x).

enterogenous cyst and teratoma, or acquired through cell implantation following surgery [5]. They occur at any age, presenting mostly in third or fourth decade women. Commonly found in gonads, especially ovaries, they sometimes occur in middle sites, near the lines of embryologic fusion.

Surgical management is a local excision by laparotomy or laparoscopy, or a hemicolectomy due to the inability to rule out malignancy or to safely separate cyst and colon wall.

Therefore, in front of an unusual location, there is a clear need for expanding physical examination, supplemented by blood analyses and imaging if required. In our patient, further examinations were done, more precisely, a thoracic CT showed no sign of malignancy and no testicular tumor was found at ultrasound imaging.

Our literature review revealed 16 cases of dermoid cysts in the ileocecal region. Patients were from 1 to 53-year old, mean age 24.9-year old, mostly women. The first case was published in 1956. The cysts were located at the mesenteric border of the cecum, intracecal, and abutting the ascending colon, in the appendicular area [6] or adjacent to the ileocecal valve. The most common location was the cecum, with 9 cases reported.

Only those cysts were removed with hemicolectomy. Indeed, 5 patients received this treatment [6-10], while the others underwent local resection [11-14,16] with or without the appendix. Past surgical histories were positive for 2 of them [7,8].

Some authors have postulated that [3] acquired dermoid cysts may result from the implantation of squamous

epithelium from the skin following a surgical procedure. One had undergone cholecystectomy and laparoscopic sterilization [7], the other, hysterectomy and appendectomy. Thus, the theory could be supported since all the operations were involving the RLQ.

In our case, past surgical history was negative. The mass was found incidentally, which is the second to be recorded in the literature [14], to the best of our knowledge. In our patient, physical examination was normal, which suggests that without the imaging, the mass could have been discovered several years later.

As demonstrated, ultrasonography can be used to evaluate these masses, as well as abdominal CT, classically depicting greasy substance and calcification. Yet, none of the patients had a typical imaging. Interestingly, in our patient, initial diagnosis was appendicular mucocele based on CT findings, since the image was compatible with a mucinous lesion (Fig. 1B), involving therefore a more aggressive surgery. This diagnosis was likely based on the unusual topography, mature teratoma arising from the cecum being rare. Dermoid cyst resembles epidermoid cysts which are known to be confused with other intra-abdominal cystic lesions, including mesenteric cysts, lymphatic cysts, appendicular mucocele, or duplication cysts [15]. Lack of specialized structures of the skin allows the distinction between both entities.

Frequently mature teratomas are characterized by hair follicles, sweat glands, and sebaceous glands filling the cyst. The cases reported suggested that in front of a mass in the cecum, found by clinical examination, imaging or exploratory laparoscopy, dermoid cysts should be considered in the differential diagnosis. Even though those masses are still removed surgically, it could be helpful to assess the patient.

Conclusion

In conclusion, the development of a dermoid cyst in the cecum is a rare occurrence; thus, dermoid cyst are very rarely included in the differential diagnosis of caecal mass. This is the tenth case described in the literature, and for the second time, physical examination was unremarkable. From this case, we found that, although mature teratoma of the cecum are not common, the diagnosis should be considered in the presence of a nontender palpable mass in the RLQ despite an atypical presentation on imaging. Furthermore, they require surgical management.

Declaration of Competing Interest

There is no competing interests statement to mention concerning this article.

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