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Xanthogranulomatous Appendicitis in a child: Report of a case and review of the literature

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Xanthogranulomatous inflammation is a well-described inflammatory process, which may involve any organ but is most frequently encountered in the gall bladder and the kidney. There are rare reports of Xanthogranulomatous Appendicitis (XA) in the adult population, but only one brief mention of such a diagnosis in a child. In this report, we describe the case of an 11-year-old boy who presented with clinical signs and symptoms of acute appendicitis necessitating appendectomy. Upon microscopic examination, the appendix showed the typical features of XA. To the best of our knowledge, this is the first well-described case XA in a noninterval appendix in a child. We also reviewed the limited medical literature on the subject.

Case Report: The patient is an 11-year-old boy who presented with a 1-day history of abdominal pain and emesis but no fever. His past medical history was unremarkable. In particular, there was no history of hemorrhagic problems or systemic disease. The family history was also unremarkable. On physical examination, he had persistent and well-localized right lower quadrant and right flank tenderness at the expected location of his appendix (McBurney's point). The laboratory findings were unremarkable. WBC was 4.9 K/microliter with the following differentials: 49% segmented neutrophils, 41% lymphocytes, 9% monocytes, and 1% eosinophils. There was no evidence of anemia or thrombocytopenia (hemoglobin = 13 g/dL, hematocrit = 36%, and platelet count = 291 K/microliter). A computed tomography scan (CT-Scan) of the abdomen showed an enlarged appendix without inflammation; however, ultrasound images showed a fluid-filled appendix with a diameter within the upper ranges of normal. A subsequent physical examination revealed an increase in abdominal pain and tenderness, and, consequently, the patient underwent a laparoscopic appendectomy. Gross evaluation showed a pink-tan appendix, measuring 8.3 cm in length and 1 cm in diameter. The serosal surface was unremarkable and cut surface demonstrated no fecalith. Microscopically, hematoxylin-eosin-stained sections of the tip of the appendix revealed numerous lipid-laden xanthoma cells in the mucosa which were surrounded by lymphocytes and plasma cells admixed with multiple multinucleated giant cells containing cholesterol clefts. The rest of the mucosa showed patchy mild neutrophilic infiltration. The postoperative clinical course was unremarkable. The patient was discharged home the following day and had an unremarkable physical examination on a follow-up visit three weeks later.

Discussion: Xanthogranulomatous inflammation is a rare form of chronic inflammation, manifested by the presence of lipid-laden macrophages admixed with lymphocytes, plasma cells, neutrophils, and often multinucleated giant cells with or without cholesterol clefts. It was initially described in the kidney by Osterlind in 1944. It has also been reported in other organs, such as gall bladder, prostate, epididymis, ovary, urinary bladder, kidney, appendix, and the exact etiology of xanthogranulomatous inflammation is uncertain.

Conclusion: Xanthogranulomatous Appendicitis (XA) is a rare clinical entity, particularly in the pediatric population. It may be encountered in specimens resected in the acute phase, but is much more common in interval appendectomy specimens. As observed in the present case and in previous reports, most patients with XA present with a picture of acute or sub-acute abdominal pain, and occasionally with a mass lesion.

Biography

Sura Al-Rawabdeh a senior specialist in Pediatric pathology is currently working in the royal medical services in Jordan. She got fellowship training in Pediatric Pathology. In nationwide children's hospital Columbus Ohio USA

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