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Case Report General Surgery

UNCORRECTED PROOF

Intraperitoneal "golden yellow" in a pediatric patient with Burkitt lymphoma: Xanthogranulomatous appendicitis

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ABSTRACT

Xanthogranulomatous inflammation is a rare chronic inflammatory reaction. Appendiceal involvement in the pediatric age group is extremely rare. We present a case of xanthogranulomatous appendicitis (XGA) that was detected incidentally during the excision of a residual intraabdominal mass in an 8-year-old male patient who was treated for Burkitt lymphoma. An 8-year-old male patient who had been diagnosed with Burkitt lymphoma underwent abdominal computerized tomography for evaluation after chemotherapy. An approximately 2.5 cm mass in the right lower quadrant of the abdomen was detected, and laparoscopic excision of the mass was planned. During the operation, it was noticed that the appendix (adjacent to the mass) was golden yellow in color and abnormal in appearance, so a synchronous appendectomy was performed. The pathology result of the mass was compatible with Burkitt lymphoma. Microscopic examination of the appendix revealed that the columnar surface epithelium had eroded and been replaced by fibrin and cell debris. Inflammatory cell infiltration rich in foamy histiocytes as well as lymphocytes and sparse neutrophils that form destructive aggregates was observed in all appendiceal layers. The final diagnosis of the appendectomy specimen was compatible with XGA. In very few XGA cases, the appendix is described as bright yellow or golden yellow. The diagnosis is usually made by the pathological examination after surgery. Though the diagnosis was made postoperatively in our case, there is now, for the first time in the literature, a view of the golden yellow color of XGA taken from an intraoperative video clip.

Keywords: Appendicitis; inflammation; pediatrics; xanthogranulomatous.

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Xanthogranulomatous inflammation (XGI) is a rare chronic inflammatory reaction described by the infiltration of affected tissue by lipid-laden foamy macrophages [1]. It is frequently detected in the kidney and gallbladder, and appendix involvement is extremely rare [1,2]. Although XGI's pathogenesis is not fully defined, it occurs as a result of a chronic, long-lasting inflammation. When seen in the appendix, it is considered an uncommon healing form of acute appendicitis [3]. Different etiological mechanisms have been described for the occurrence of XGI in different organs, with the conclusion that xanthogranulomatous pyelonephritis, cholecystitis, and appendicitis occur due to obstructive reasons [1]. The literature contains little information about xanthogranulomatous appendicitis (XGA). The preoperative diagnostic evaluation of these patients is still unclear, and the diagnosis of XGI is typically made after surgical intervention [2]. Although some patients present with acute appendicitis symptoms, clinical pre-

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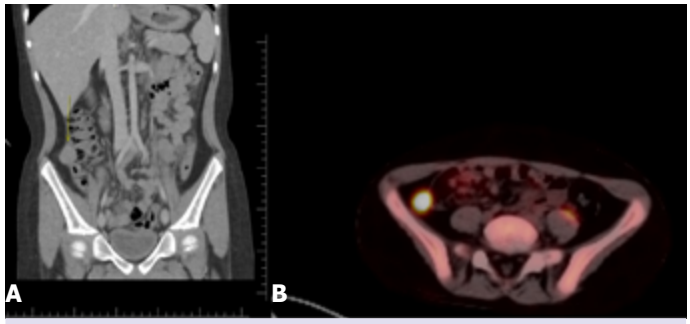

Figure 1. **(A)** A mass compatible with residual lymphoma localized in the ileocecal region (white arrow). **(B)** Radioactive activity involvement in the residual mass (PET CT).

sentation sometimes suggests an intraabdominal mass and mucocele [2, 4]. In this report, we present a case of XGA that was detected incidentally during excision of a residual intraabdominal mass in an 8-year-old male patient as he was treated for Burkitt lymphoma.

CASE REPORT

An 8-year-old male patient diagnosed with Burkitt lymphoma underwent abdominal computerized tomography (CT) for evaluation after chemotherapy. An approximately 2.5 cm mass was detected in the right lower quadrant of the abdomen. Comparison with the patient's previous tomography results showed that many pathological lymph nodes in the abdomen were regressed, so the currently visualized mass was thought to be a residual mass (Fig. 1A). The standardized uptake value of the mass was detected as 24.9 using positron emission computed tomography (PET-CT) (Fig. 1B). Excision of the mass was recommended for definitive diagnosis as well as further treatment planning, so laparoscopic excision of the mass was planned. The patient's preoperative vital values were normal, and a mass was palpated in the right lower quadrant of the abdomen on physical examination (leucocyte: 3.7×10^9/L; platelets: 233,000/mL; neutrophil percentage: 36.6%; C-reactive protein: 1 mg/dL). During the operation, the appendix (adjacent to the mass) was noticed as golden yellow in color and abnormal in appearance (Fig. 2). A synchronous appendectomy was performed after obtaining written permission from the patient's parents. The patient had no postoperative complications and was discharged on the second postoperative day. The pathology result of the mass was compatible with Burkitt lymphoma, and microscopic examination of the appendix revealed that the columnar surface epithelium had eroded and been replaced with fibrin and cell debris. Inflammatory cell infiltration rich in

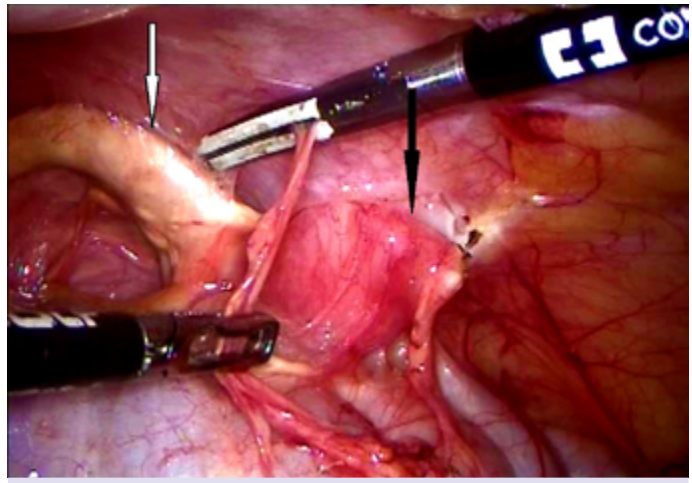

Figure 2. Residual mass in the ileocecal region (black arrow) and an appendix with a golden hue (white arrow).

foamy histiocytes as well as lymphocytes and sparse neutrophils that form destructive aggregates was observed in all appendiceal layers. The final diagnosis of the appendectomy specimen was compatible with XGA (Fig. 3).

DISCUSSION

Appendectomy for acute appendicitis remains one of the most common surgical operations, and pathological examinations often reveal the presence of an inflammatory process associated with neutrophil leucocyte infiltration in muscularis propria. The incidence of XGI, an extremely rare type of inflammation, in the appendix has been reported to be between 0.25% and 0.64% [5, 6]. No cases of XGA were reported in various large-scale studies such as Charfi et al. (24,697 appendectomy patients), Uylas et al. (2,076 patients), and Kepil et al. (1,154 patients) [7–9]. Guo et al. [10] reported higher incidence of XGA after interval appendectomies compared with normal appendectomies. XGA, which is more commonly observed in the adult age group, is very rare in the pediatric age group. Our patient, an 8-year-old male, is one of very few pediatric examples reported in the literature.

XGA is a disease that progresses with different clinical presentations. In a review, Akbulut et al. [2] reported that typical clinical appendicitis symptoms were seen in 57.9% of patients. Ito et al. [4] reported the rate as 88.9%. Interval appendectomy is more common in this patient group, and the final diagnosis of XGA is usually made by postoperative pathological examination. In patients without acute appendicitis, the diagnosis of XGA is usually made after surgery due to additional pathologies located

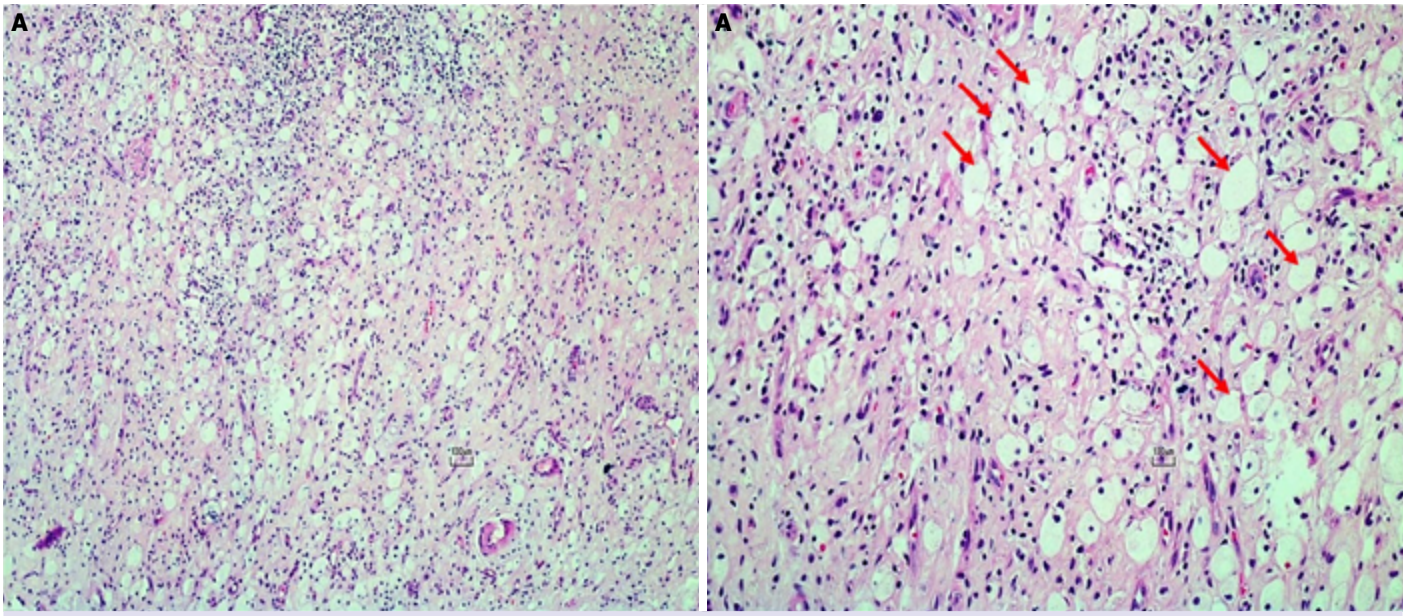

FIGURE 3. Inflammatory reaction composed of prominent xanthoma type foamy histiocytes (arrows) admixed with lymphocytes and sparse neutrophils (A: H&E, 200x, B: H&E, 400x).

in the pericecal region [4, 11, 12]. Our patient also did not present with clinical symptoms of appendicitis. XGA was discovered incidentally during surgery for a pericecal residual lymph node. Similarly, Nam et al. [13] described a 23-year-old patient on a follow-up for Burkitt lymphoma who underwent appendectomy due to fluorodeoxyglucose (FDG) uptake on PET-CT imaging, which was considered a residual disease. In our case, no FDG uptake was observed in the appendix in PET-CT.

No pathognomonic radiological findings exist for XGA. In patients with appendicitis, classical radiological findings of acute appendicitis are typically observed [2]. Incidental cases describe findings suggesting pericecal mass, fistula, and mucocele [2, 3]. In the literature, FDG uptake in PET-CT was reported in only one case [12]. In the presented case, no findings suggestive of appendicitis were observed in abdominal CT or in PET-CT.

Conclusion

In the literature, the appendix is described as bright yellow or golden yellow in very few XGA cases [12, 14]. Because the diagnosis is usually made during the postoperative pathological examination, macroscopic and microscopic images are available in pathological case reports in the literature [15]. Similarly, although our diagnosis was made postoperatively, there is now, for the first time in the literature, a view of the golden yellow color of XGA taken from an intraoperative video clip.

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