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Xanthogranulomatous appendicitis in interval appendectomy specimens of children

Yusuf Hakan Çavuşoğlu [a,b,*], Burak Ardıçlı [b], Sema Apaydın [c], Çağatay Evrim Afşarlar [b,d], Engin Yılmaz [b]

[a] Department of Pediatric Surgery, Gazi University Faculty of Medicine, Yenimahalle, 06560 Ankara, Turkey
[b] Department of Pediatric Surgery, Dr. Sami Ulus Children's Hospital, Altındağ, 06080 Ankara, Turkey
[c] Department of Pathology, Dr. Sami Ulus Children's Hospital, Altındağ, 06080 Ankara, Turkey
[d] Department of Pediatric Surgery, Hitit University Faculty of Medicine, 19030 Çorum, Turkey

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ABSTRACT

Xanthogranulomatous inflammation is common in interval appendectomy specimens in adults, but it is unusual in children. Histopathologic specimens of interval appendectomy, within an 8-year period, were reevaluated to assess the true incidence. A computer search of the hospital database of all appendectomies was performed between January 2008 and June 2015 to identify all interval appendectomy cases. A total of 2694 patients underwent appendectomies. Of these, 13 were interval appendectomies. After pathologic evaluation, 2 (15.4%) of the specimens were reported as xanthogranulomatous appendicitis (XA). Histopathologic examination of these interval appendectomy specimens, granulomas (59%), xanthogranulomatous inflammation (36%) and Crohn-like changes (50%) were common in adults. However, XA is a particularly rare clinical entity among children. Two cases of XA were reported in children in the English literature. One was a 12-year old boy that underwent interval appendectomy 6 weeks after an episode of acute appendicitis. The other was an 11-year old boy with acute (non-interval) appendicitis, but the complete blood count was suggestive of an acute suppurative inflammation. These two cases are the 3rd and 4th cases of XA reported in children in the English literature, and both were managed by interval appendectomy. Thus, XA may be encountered in interval appendectomy specimens and association with IBD has to be ruled out.

Xanthogranulomatous inflammation (XI) is common in interval appendectomy specimens in adults [1], but it is rarely reported in children [2]. We evaluated the interval appendectomy specimen of a 12-year-old boy, who was conservatively treated because of appendicular mass 6 weeks earlier.

Additionally, we reevaluated the histopathologic specimens of interval appendectomy samples, within an 8-year period, to assess the true incidence of XA in a high-circulation pediatric surgery clinic.

1. Methods

We conducted a database search of all appendectomies performed at Dr. Sami Ulus Children's Hospital, Department of Pediatric Surgery, Ankara, Turkey from January 2008 to June 2015 to identify interval appendectomy cases. Dr. Sami Ulus Children's Hospital is a high-circulation training hospital that accepts patients from Ankara and other cities. A total of eight pediatric surgery specialists work at the pediatric surgery clinic.

For patients who were treated conservatively with intravenous antibiotics and diagnosed with an appendiceal mass, elective appendectomy was recommended. Patients who underwent interval appendectomy were included in this study. Demographic and clinical data (B.A.) and pathologic archive specimens (S.A.) were reevaluated by two of the authors.

The patients diagnosed with appendiceal masses were treated with intravenous triple antibiotic therapy (ampicillin or sulbactam/ampicillin, amikacin, and metronidazole). All patients were

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* Corresponding author. Department of Pediatric Surgery, Gazi University Faculty of Medicine, Yenimahalle 06560, Ankara, Turkey. Tel.: +90 312 2026219.
E-mail address: hakancavusoglu@hotmail.com (Y.H. Çavuşoğlu).

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Table 1
Demographic and histopathological findings of interval appendectomy patients.

No	Age	Sex	Symptom duration (days)	Expected duration (day)	Histopathological findings					
					Suppurative inflammation	Xanthogranulomatous inflammation	Lymphoid hyperplasia	Chronic transmural inflammation	Peritonitis	Other
1	11	M	7	100			+			
2	12	M	2	81			+			
3	8	F	4	66			+			
4	5	M	3	118	+		+	+	+	
5	1.5	F	8	81	+		+	+	+	
6	17	F	9	71			+	+	+	
7	11	M	7	113	+		+	+	+	
8	9	M	2	97			+		+	
9	13	F	7	50	+		+	+	+	
10	15	M	–	–			+	+	+	
11[a]	12	M	14	54	+	+ (CD68 and CD45)	+	+	+	
12	13	M	3	59	+			+	+	
13	17	M	8	85	+	Rare granulomas and focal xanthoma cells	+	+	+	Myxoid lesion

[a] Index patient.

discharged with a prescription of oral amoxicillin/clavulanic acid for 2 weeks and scheduled for interval appendectomy after 6 weeks.

2. Results

Of the, 2694 appendectomies conducted between 2008 and 2015, 13 were interval appendectomies and were included in this study (Table 1). The male to female ratio was 2:1. The mean patient age was 11.1 ± 4.4 years (range: 1.5–17). The mean white blood cell count and *C*-reactive protein were 13686 ± 4145/mm^3 (range: 6100–21 300) and 126.1 ± 88.9 mg/L (range: 24–334), respectively. Hospitalization and intravenous triple antibiotic drug duration were 8.16 ± 3.21 days (range: 4–14). All patients were given oral antibiotics for 2 weeks after discharge. The mean waiting time before surgery was 81.3 ± 22.4 days (range: 50–118). A drain was inserted in two patients, one preoperatively and one intraoperatively. Appendectomies were performed by open ($n = 10$) or laparoscopic surgery ($n = 3$).

After pathologic evaluation, 2 (15.4%) of the specimens were reported as XA. Case #11 was our index patient. In the histological examination, all layers of the appendix, especially the serosa, revealed focal lipid-laden histiocyte (xanthoma cells) collections processing non-caseous granuloma without cholesterol clefts (Fig. 1). They were mixed with multinucleated giant cells, plasma cells, lymphocytes, and neutrophils. No parasites, foreign bodies, or obstructing lesions (fecalith, mucocele, or tumor) were found; and specific stains for acid-fast bacillus and fungi were negative. In Case #13, there were focal xanthoma cell groups and rare giant cells in the serosa, but no granuloma or myxoid lesions were observed (Fig. 2). There were microabscesses in the appendix layer. The histopathological findings of all patients have been summarized in Table 1. Patients were discharged on a mean of 2.9 ± 2.4 (range: 1–9) postoperative days after interval appendectomies.

Two XA patients were reevaluated whether their diagnosis was associated with inflammatory bowel disease (IBD), and found no

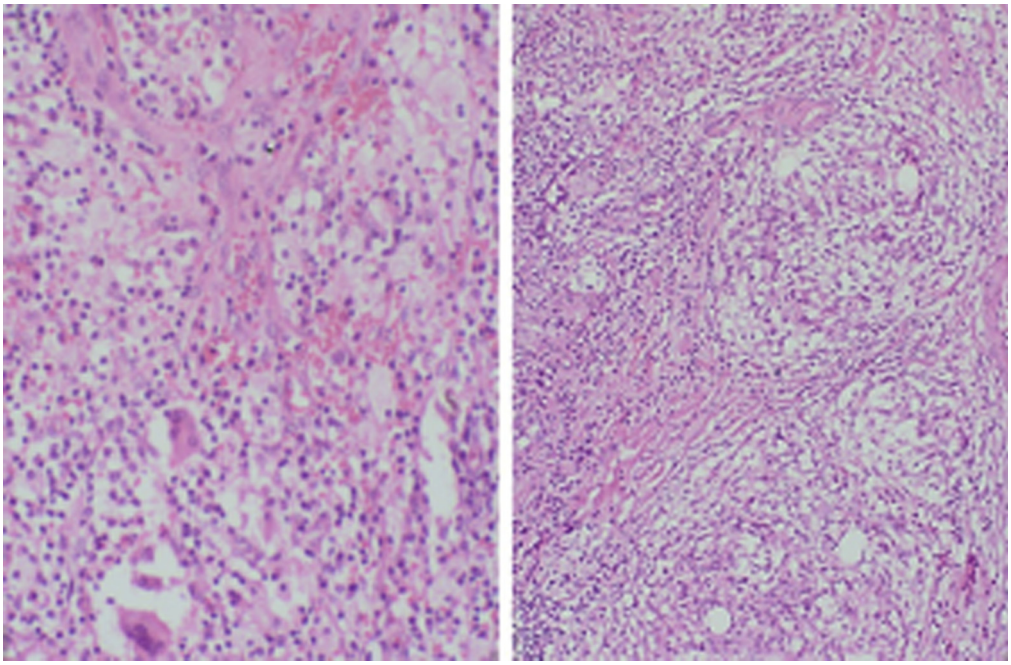

Fig. 1. Case #11- Hematoxylin and eosin-stained sections. Multinucleated giant cells and xanthoma cells with pale, abundant cytoplasm (left) and granulomas (right).

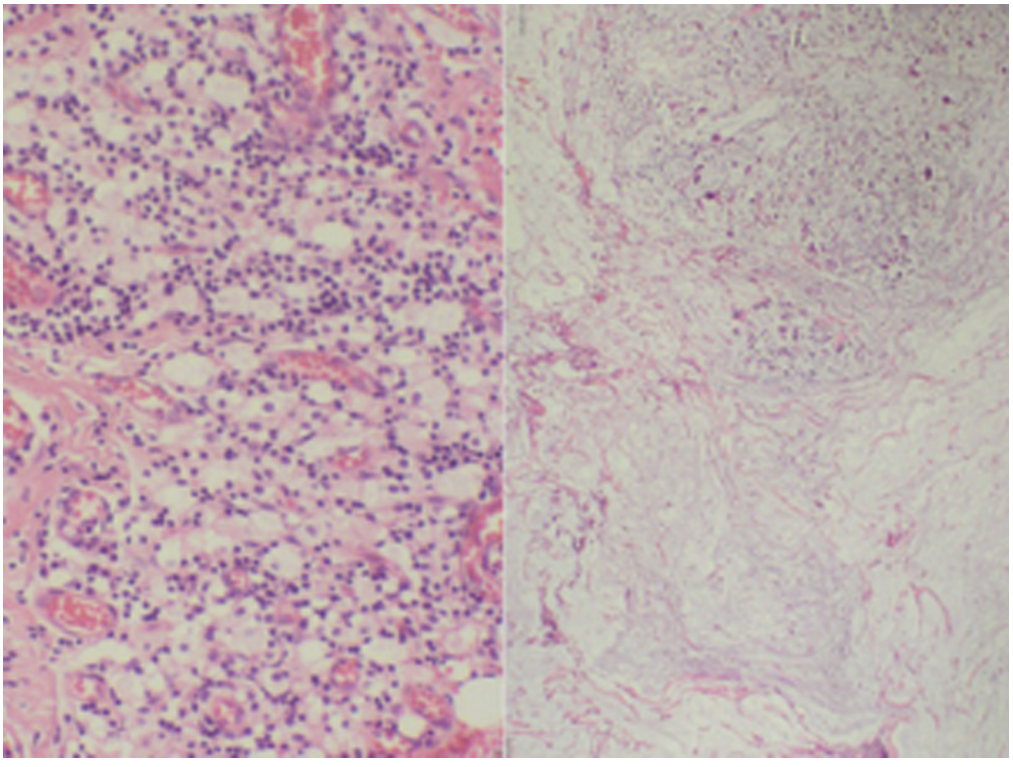

Fig. 2. Case #13- Hematoxylin and eosin-stained sections. Focal xanthoma cell groups (left) and rare giant cells in serosa (right).

remarkable signs and symptoms. Follow-up periods of these XA patients were 3 and 12 months.

3. Discussion

XI is a rare form of chronic appendiceal inflammation, manifested by the presence of lipid-laden macrophages combined with lymphocytes, plasma cells, neutrophils, and often multinucleated giant cells with or without cholesterol clefts [3]. An association has been suggested between xanthogranulomatous response with long standing appendiceal inflammation and the formation of appendiceal mass [4,5]. It's also concluded that this diagnosis might be associated with inflammatory bowel disease and without appropriate clinical history this changes may be misinterpreted as Crohn's disease [2].

Conservative treatment with IV antibiotics, followed by elective appendectomy is the recommended management for patients with appendiceal mass. However, unresponsiveness to medical treatment is an indication for immediate operation [6]. On the histopathological examination of interval appendectomy specimens, granulomas (59%), XI (36%), and Crohn's-like changes (50%) are common in adults [1]. However, XA is a particularly rare clinical entity in the pediatric population. Two cases of XA have been reported in children in the English literature [2]. One was a 12-year-old boy who had an interval appendectomy 6 weeks after an episode of acute appendicitis. The other was an 11-year-old boy with acute (non-interval) appendicitis, but the complete blood count was suggestive of an acute suppurative inflammation [2].

The main limitation of this study was that we only evaluated the cases of interval appendectomy. A control group of patients with routine acute appendicitis would have strengthened this study. The small number of interval appendectomies is another limitation.

4. Conclusion

In children, XA is rare. These two cases are the third and fourth cases of XA reported in children in the English literature, and both were managed by interval appendectomy. Thus, XA may be encountered in interval appendectomy specimens and association with IBD has to be ruled out.

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