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Case Report

A Pediatric Case of Granulomatous Appendicitis Operated Due to Recurrent Abdominal Pain

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Abstract

Granulomatous appendicitis (GA) is granulomatous inflammation of the appendix wall. It is generally idiopathic; however, it may also be associated with many diseases such as Crohn's disease, parasitic infections, tuberculosis, or foreign bodies. An 11-year-old male patient, with a 3-month history of abdominal pain and bilious vomiting, had right lower quadrant abdominal tenderness. His white blood cell count was 8.6 x10^3/μL. An abdomen ultrasound was considered to show plastron appendicitis and an appendectomy was performed. Microscopically, thickening of the appendix wall with edema, fibrosis and lymphoid infiltration was observed. The patient was evaluated as idiopathic GA since no disease was detected that caused GA. When the appendix has a firm consistency and is difficult to separate from the surrounding tissues, GA should be considered before malignancy, particularly in the pediatric age group. An appendectomy should be performed before deciding on radical surgery.

Keywords: Bilious vomiting, carcinoid tumor, granulomatous appendicitis, pediatric, recurrent abdominal pain

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Granulomatous appendicitis (GA) is a granulomatous inflammation of the appendix wall. The causes of GA include fungal infections, mycobacterium tuberculosis, blastomycosis, Histoplasma capsulatum, schistosomiasis, actinomycosis, toxoplasma, brucella, candidiasis, foreign bodies, and Crohn's disease (CD).[1] Patients diagnosed with GA usually apply with complaints of abdominal pain and are operated on based on a pre-diagnosis of acute appendicitis.[2] When all age groups are taken into consideration, the prevalence of GA is 1-2%.[3] In the pediatric age group, GA is less common, and its prevalence is 0.13-1.04%.[4,5] Since the appendix wall is thick and has a firm consistency in GA, it can be clinically and radiologically confused with malignancy.[2] The clinical course of GA cases and the treatment approach during the surgery vary considerably. Pathological diagnosis is the basis of the surgical procedure, but it is not always possible to establish a pathological diagnosis during the surgical procedure. In such cases which are urgent and apply outside of working hours, surgeons intervene in them based on their experience since they cannot reach the pathological diagnosis.[6]

We present a case of GA, the prevalence of which is lower in childhood and which can be confused with malignancy macroscopically, to underline that the treatment should be

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re-adjusted depending on pathological findings, by pointing out the macroscopic appearance of GA during the surgery.

Case Report

An 11-year-old male patient, with a 3-month history of abdominal pain and bilious vomiting, had right lower quadrant abdominal tenderness. In laboratory examinations, the white blood cell count was 8.6 x 10^3/μL (reference range of 4-10 x 10^3) and C-reactive protein was 31.03 mg/L (reference range of 0-5 mg/L). His other blood counts and biochemical tests before the surgery were normal. Abdominal ultrasonography revealed that the diameter of the appendix was 9 mm, the appendix wall was edematous, the surrounding fatty tissue was edematous and formed a sheath around the appendix, and plastron appendicitis was considered. In the examination of direct radiological imaging, no characteristic feature was observed.

Our patient underwent an emergency operation with a pre-diagnosis of acute appendicitis. The appendix, which was 6-7 cm in diameter and 7-8 cm in length, was firm and hyperemic. The appendix was located behind the bladder and was partially organized with the bladder wall. The bladder and appendix were carefully dissected and the bladder wall's integrity was preserved. The mesoappendix was noticeably short. The meso was separated. An appendectomy was performed. A carcinoid tumor or mucosal/granulomatous appendicitis was considered during the surgery. The patient was discharged along with medical advice on the third postoperative day.

The image of the appendix during surgery is shown in Figure 1. The macroscopic examination of the surgical material indicated that the appendix was highly enlarged and its wall was thickened (Fig. 2). Microscopically, the appendix wall was thickened with edema and fibrosis and lymphoid infiltration progressing to the serosa was observed. Lymphoid infiltration formed lymphoid follicles in some areas, and the presence of sparse granuloma structures was striking (Fig. 3). In addition, acute inflammation was observed around the lumen. Mycobacterium tuberculosis bacillus was not detected with the histochemical Ehrlich Ziehl Neelsen stain and no fungal hyphae or spore structure was observed with the periodic acid-Schiff stain. The case was reported pathologically as GA.

The patient had no symptoms or clinical findings associated with tuberculosis, sarcoidosis, or CD. A skin test was

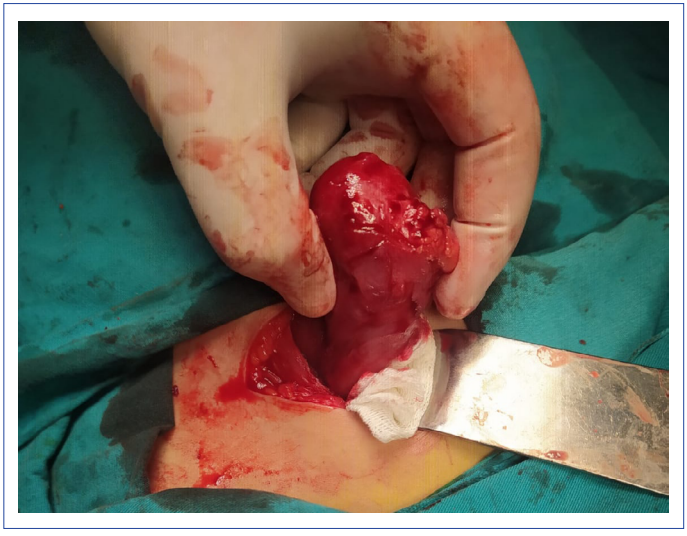

Figure 1. The during surgery view of granulomatous appendicitis.

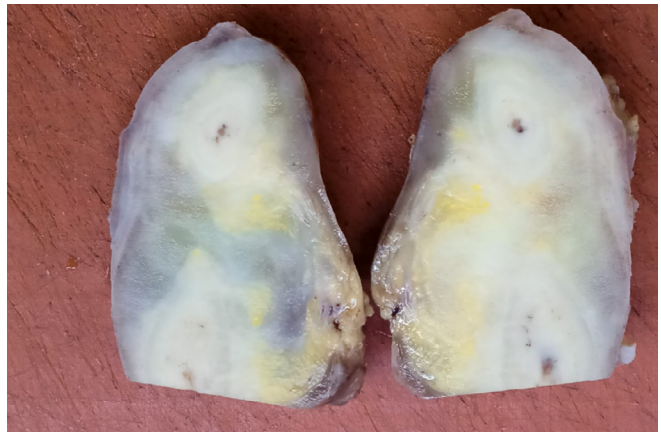

Figure 2. The macroscopic view of granulomatous appendicitis: A diffused and noticeable thickening of the appendix wall is noticed in the cross-section of the appendix.

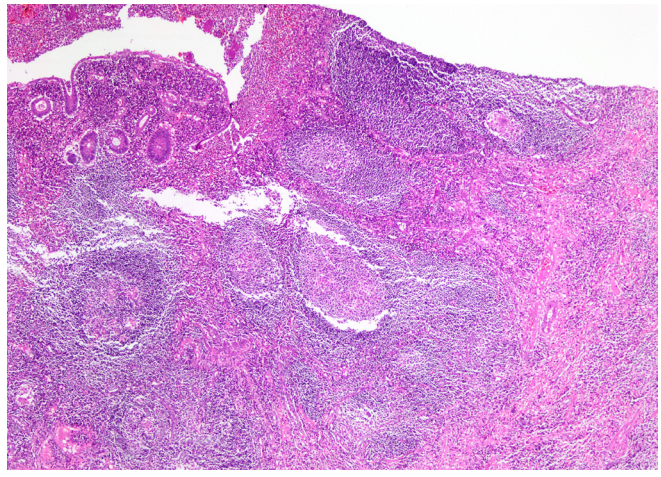

Figure 3. The microscopic view of granulomatous appendicitis: Acute inflammation rich in leukocytes with polymorphous nuclei was observed in and around the appendix lumen, as well as chronic inflammation developing in lymphoid follicles in the appendix wall, thickening with fibrosis and edema, and a small number of granulomatous structures composed of epithelioid histiocytes and giant cells. HEx50.

performed to exclude tuberculosis and was evaluated as negative. Serum angiotensin-converting enzyme was normal in terms of sarcoidosis.

In the fifth month after the surgery, our patient was not diagnosed with any disorder that causes GA and was evaluated as idiopathic GA.

Discussion

Idiopathic (primary) GA has a low prevalence and is a diagnosis of exclusion.[7] Secondary GA is associated with many infectious and non-infectious causes. GA is seen in various conditions such as CD, Yersinia infection, tuberculosis, sarcoidosis, foreign body reaction, fecal-related obstructions, diverticulitis, mucosal, and tumors.[8] Before establishing a diagnosis of idiopathic GA, the cases should be investigated in terms of the causes of GA.

GA has been used to be associated with CD, upon the increase in cases. It is now understood that there are pathologies other than CD.[9] Pathologies that cause GA in patients must be studied. In many GA cases, the patient's treatment is started with an appendectomy, but the patient's follow-up, examination, and treatment continue until the final diagnosis is established.

When the appendix is characterized by a macroscopically firm consistency, a thick wall, and an increased blood supply during the surgery, it is remarkably similar to a carcinoid tumor.[2] In the literature, there are cases of the adult age group who have undergone right hemicolectomy+omentectomy+appendectomy according to the macroscopic examination.[10] When surgeons know macroscopy in GA cases before pathological examination, they can make decisions during surgery, accordingly; therefore, macroscopy becomes increasingly more important.[2] Surgeons should take microscopic diagnoses into account in order to prevent radical and unnecessary procedures.

The prevalence of GA was reported to be 1.04 % in an African-centered series of 1150 cases in the pediatric age group.[5] The etiology of GA in five of the patients in this series was unexplained. No malignancy was reported for any patient. The literature includes mostly idiopathic cases in etiology of GA.[11] No additional diagnosis was established for our patient upon his examinations. We also consider GA as idiopathic. In both the case report and the literature, the appendix has a firm consistency and is attached to the surrounding tissues during the surgery of GA cases.[5]

Identification of the etiology of granulomatous inflammation after appendectomy is the most important parameter in determining the treatment course for GA. The etiology of granulomatous inflammation must be investigated, and the patient should be closely monitored for a long time after surgery. In cases that were macroscopically thought to have GA, the cecum and ileum should be thoroughly examined, as in all appendectomies, and the findings should be recorded in the surgical notes.

Conclusion

When the appendix has a firm consistency, is attached to the surrounding tissues, and is difficult to separate from the surrounding tissues during surgery, particularly in the pediatric age group, GA should be considered. An appendectomy should be performed before deciding on radical surgery such as hemicolectomy or ileostomy and the patient should be re-evaluated with the pathological diagnosis.

Disclosures

Informed consent: Written informed consent was obtained from the patient's parent.

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