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Case Report

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Appendicular tuberculosis mimicking a tumor in an adolescent: A case report

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Abstract

Appendicular tuberculosis is rare, particularly in adolescents, and also rarely involves the gastrointestinal tract. It may be confused with malignancies since it has no specific clinical and laboratory findings. A 17-year-old girl was admitted to the emergency department with intermittent abdominal pain and bilious vomiting and diagnosed with perforated appendicitis. During explorative laparotomy, hypertrophic appendix with irregular serosa and enlarged mesenteric lymph nodes suggested pericecal tumor and right hemicolectomy was performed. Histopathological examination revealed appendix and lymph nodes compatible with tuberculosis. The patient received anti-tuberculosis treatment and had no recurrence of the disease or gastrointestinal problems during five-year follow-up. In conclusion, gastrointestinal tuberculosis may mimic malignancies and preoperative diagnosis may be challenging.

Keywords: Abdominal pain, appendicular tuberculosis, gastrointestinal, right hemicolectomy.

Tuberculosis presents with various clinical features depending on the involvement of different organs and often mimics other diseases. Pediatric tuberculosis is most commonly found in the lungs, with 80% of cases being the most common site of disease. Extrapulmonary involvement includes lymph nodes (67%), meninges (13%), pleura (6%), miliary spread (5%), musculoskeletal system (4%), kidney and skin diseases.[1] Among extrapulmonary manifestations, abdominal tuberculosis is rare in children. Abdominal involvement is found in approximately 1 to 3% of all tuberculosis cases and in 12% of extrapulmonary diseases in children <15 years of age.[2] The ileocecal region is the most common site of gastrointestinal tuberculosis.[3] The appendix is located in close proximity to the ileocecal region and the incidence of appendicular tuberculosis (ATB) among all appendectomies performed in adult series is reported to vary between 0.1 and 3.0%.[4] The mechanism of appendicular involvement in tuberculosis is unclear, clinical presentation is variable, and management is still controversial. Most cases of ATB have been reported in adults.

In this article, we present a case of an adolescent patient with a preliminary diagnosis of perforated appendicitis who underwent aggressive surgery for considering a pericecal tumor and was diagnosed with ATB.

CASE REPORT

A 17-year-old female patient presented to the emergency department after intermittent abdominal pain that worsened for a month. The patient had no chronic disease and was previously healthy. At the time of admission, she had complaints of bilious vomiting and loss of appetite, and her body temperature, heart rate, and respiration were within normal ranges. On her physical examination, there was generalized tenderness, as well as defense and rebound in the right lower quadrant. Laboratory tests revealed leukocytosis (white blood cell [WBC] count: 19.750 10^3/mL), shift to the left (neutrophils 88%), and elevated C-reactive protein (CRP, 29.7 mg/L). Routine biochemical and urine analyses were within normal limits. There was no abnormality in direct abdominal

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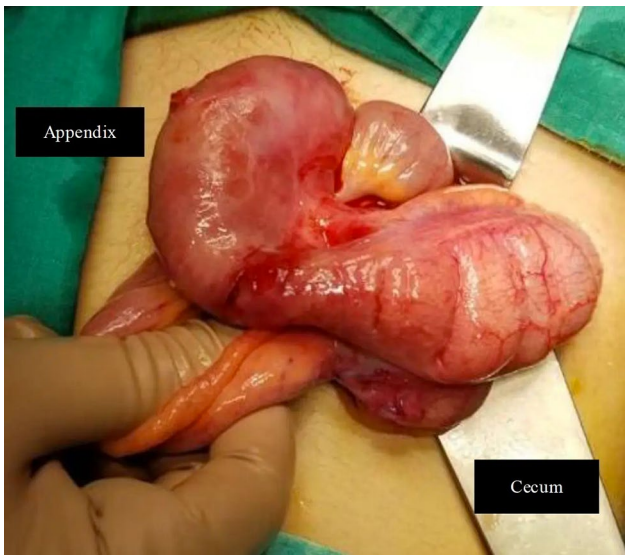

Figure 1. Intraoperative view of the appendix and cecum. The surface of appendix appears tumor-like.

X-ray. Abdominal ultrasonography showed a blind-ending, non-compressible, approximately 4-cm long, 12-mm in diameter at the thickest point, and markedly edematous blind-ending bowel in the right lower quadrant of the abdomen. With a preliminary diagnosis of perforated appendicitis, an operation was planned and she was taken to the operating room.

In the laparotomy performed with the right paramedian incision, approximately 100 mL of reactional serous fluid was aspirated. During exploration, the appendix surrounded by omentum was severely hypertrophied (8×4 cm), inflamed, edematous and its serosa was irregular (Figure 1). The base of the appendix and the cecum wall were also thickened and inflamed, and extremely hypertrophied lymph nodes were observed in the cecum mesentery toward the duodenum. The terminal ileum and ascending colon appeared normal. The appearance suggested a tumor, frozen-section analysis could not be performed intraoperatively, and it was decided to perform aggressive surgery considering that a biopsy and second look would affect the staging. A right hemicolectomy was performed from the ileum 10 cm proximal to the ileocecal valve to ¼ proximal to the ascending colon and transverse colon, including the mesenteric hypertrophic lymph nodes and cecum, followed by an ileocolic anastomosis. Postoperative course was uneventful. Histopathological examination revealed granuloma foci in the

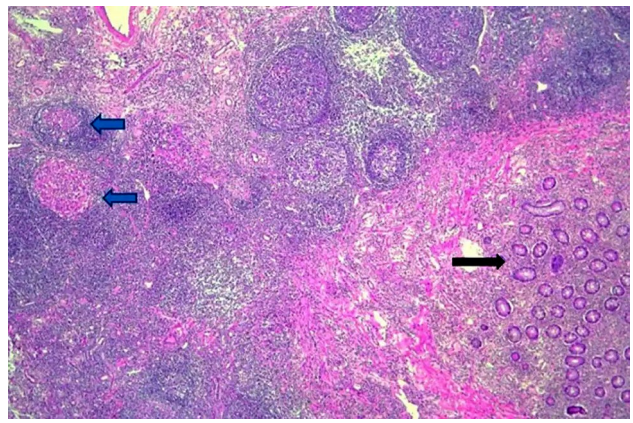

Figure 2. Granuloma areas in the appendix wall (blue arrow), appendix mucosa (black arrow) (H&E, ×100).

appendix wall, a granuloma structure consisting of multinucleated giant cells with epithelioid histiocytes, and granulomas in the mesenteric lymph nodes, compatible with tuberculosis (Figure 2). After the diagnosis of tuberculosis was made by histopathological examinations, no other focus was detected in the whole-body scans, and the primary source was considered to be pericecal and appendix. The patient was referred to the Department of Pediatric Infection Diseases and quadruple anti-tuberculosis treatment was initiated after a whole-body scan. There was no disease recurrence or gastrointestinal problem during the five-year follow-up. A written informed consent was obtained from the parents of the patient.

DISCUSSION

The gastrointestinal system is the sixth most frequently affected site in cases of extrapulmonary tuberculosis.[5] The most affected areas are the ileocecal region, followed by involvement of the jejunum and colon.[3] It has been suggested that, as a result of involvement of this region, spasms and hypermotility may occur in the early period followed by thickening of the ileocecal valve and ulcers, stenosis, and adhesions may occur in the advanced stages.[6,7] It has been also proposed that tuberculosis is a great mimicker and should be included in the differential diagnosis.[8] Indeed, abdominal tuberculosis presents with abdominal pain, diarrhea, fever, and weight loss, making it difficult to distinguish from inflammatory bowel disease and gastrointestinal malignancies.[9] In our case, we consider that tuberculosis involvement

was primarily in the ileocecal region, and that abdominal pain developed due to proximal hypermotility and distension in the intestinal wall as a result of thickening and occlusion by spasm of the ileocecal valve. The presented case was initially presented with abdominal pain and bilious vomiting as perforated appendicitis.

Tuberculosis of the appendix, first recognized by Corbin as early as 1837, may present as either a primary or secondary condition. Primary cases are rare, with an incidence ranging from 0.1 to 0.6%.[10] The mechanism of appendicular involvement in tuberculosis has not been fully elucidated yet, its clinical presentation is variable, and management is controversial. Some authors have proposed that tuberculosis can spread to the gastrointestinal tract by several routes, including ingestion of the bacilli, which invade the mucosal tubular glands and submucosal lymphoid tissue, hematogenous spread or reactivation from a primary pulmonary focus, and direct spread via the lymphatics from infected lymph nodes and adjacent infected structures.[1,11] Since the appendix is also an organ of the ileocecal region, it is thought that it can be affected by the same mechanisms.

A typical clinical presentation of ATB has not been described in the literature. Singh et al.[11] reported that patients might present with mild disease with moderate intermittent right iliac fossa pain, chronic disease accompanied by vomiting and diarrhea, or with signs of acute obstructive appendicitis, or as an incidentally discovered as latent disease. Akbulut et al.,[4] in their study examining 155 cases of ATB, showed that the most common complaints were right lower quadrant pain (38%) and recurrence (30%), generalized abdominal pain (12%), intestinal obstruction (6%), and other gastrointestinal system complaints. In our case, the main complaint was intermittent abdominal pain, and there were complaints of bilious vomiting and loss of appetite.

Furthermore, there is no specific laboratory test or radiological image to confirm the ATB. Therefore, the diagnosis can only be confirmed by histopathological examination of the appendix. In the literature, it has been suggested that diagnosis is usually made by colonoscopy/biopsy in suspicious cases; however, there is no study in which preoperative diagnosis is made and anti-tuberculosis treatment is initiated; and appendix removal is almost performed routinely for histopathological examination and additional surgeries are also performed.[8] Singh et al.[11] reported in their study that they performed right hemicolectomy (65%) in 11 patients with ileocecal appendix involvement and appendectomy (35%) in six patients with periappendicitis and the authors concluded surgery as the treatment of choice for ATB, as medical treatment alone could not control recurrent attacks of inflammation. Akbulut et al.[4] also found that 151 of 155 cases underwent one or a combination of the following surgical procedures: appendectomy (79%), hemicolectomy (15%), ileocecal resection (3%), and gynecologic surgery (3%). Four cases were diagnosed and treated without appendectomy. However, in recent years, ATB has been diagnosed more frequently with appendectomy alone or even intra-abdominal fluid sampling alone.[12-19] Over the last 14 years, three cases underwent right hemicolectomy due to suspicion of malignancy.[4,9] In our case, during surgery, the appendix was highly hypertrophied and its surface had a nodular appearance, as well as the appendix root and cecum wall were very thickened, and there were numerous enlarged lymph nodes in the mesentery, which initially suggested malignancy and a right hemicolectomy was performed.

In clinical practice, confirmatory diagnosis is made by mycobacterial culture, positive polymerase chain reaction, and/or histology; tuberculous granulomas with or without central caseation on histopathological examination are diagnostic.[8] Since delay in treatment may lead to serious complications, early examination of histology results is critical to prevent cases from being overlooked and treated without delay.[20-22] Treatment of ATB, whether primary or secondary, is similar to that for pulmonary tuberculosis, using an empirical antibiotic regimen with rifampin, isoniazid, pyrazinamide, and ethambutol or streptomycin.

In conclusion, preoperative diagnosis of gastrointestinal tuberculosis involving the appendix is challenging due to the lack of specific clinical signs and confirmatory laboratory tests, and aggressive surgery may be necessary to manage the condition, as it can often be mistaken for malignancy. In countries with a high incidence of tuberculosis, ATB should be kept in mind in patients with active or previous pulmonary disease, in patients with non-specific gastrointestinal symptoms, in those with enlarged

lymph nodes in the cecum on preoperative imaging, and in cases who have a hypertrophied appendix with irregular surface detected during surgery even in children.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Concept, data collection, writing the article, references: H.O.; Design, control, critical review: M.K.; Analysis, literature review: H.O., M.K.

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