Rare Case of Abdominal Tuberculosis Presenting With Abdominal Cocoon in a Pediatric Patient

Erum Sarwar[1], Romeeza Zahid [2] , Huda Sarwar [3] , Hamza Suhail1[4] , Maaz Suhail Rana[5]

[1,4] Children's Hospital Faisalabad

[2 ,3] Institute of Public Health, Lahore

[5] Punjab Rangers Teaching Hospital, Lahore

Abstract

Objective: To highlight an uncommon presentation of abdominal tuberculosis (TB) in children, with the aim of improving early detection and diagnosis, particularly in high TB burden countries like Pakistan.

Method: A clinical case of a young malnourished girl from Pakistan presenting with nonspecific abdominal symptoms was examined. Due to diagnostic challenges in pediatric TB and limitations of microbiological confirmation, imaging techniques were utilized. A Computerised Tomography (CT) scan was performed to assess the abdominal condition.

Results: The CT scan revealed an abdominal cocoon (AC), a rare form of encapsulating peritoneal sclerosis, which is an extremely uncommon presentation of abdominal TB. Unlike most cases of AC that are diagnosed intraoperatively, this case was identified radiologically, enabling earlier medical intervention.

Conclusion: This case underscores the diagnostic difficulties of pediatric TB, especially its extrapulmonary forms like abdominal TB. The identification of an abdominal cocoon through radiological imaging emphasizes the importance of considering rare manifestations in children with nonspecific abdominal complaints. Early imaging can aid in timely diagnosis and treatment, which is crucial in TB-endemic regions such as Pakistan.

Introduction

Globally, approximately 650 children die due to tuberculosis every day. WHO reported in 2017 that one million children, aged less than 15, were diagnosed with TB annually and 239,000 died from this infection[1]. It was estimated that 55% of children with TB go unreported to national programs and 96% of deaths in these cases occur due to inaccessibility to treatment[3]. In Pakistan, 8% to 20% of all child mortalities are TB-related.[4] In addition to this, diagnosing the abdominal variant of TB in adolescents is quite difficult due to its nonspecific presenting complaints and other challenges let alone diagnosing an abdominal cocoon. The Abdominal Cocoon is also known as Sclerosing Encapsulating Peritonitis (SEP). A distinctive hallmark of this condition is the presence of a dense fibrotic membrane enveloping variable segments of both the small and large intestines within a protective cocoon-like structure. It could be primary (idiopathic) or secondary (to TB) in nature. It can manifest at age but its prevalence is notably higher among adolescent females.[5] The abdominal cocoon represents an uncommon underlying factor contributing to intestinal obstruction. No definite diagnostic test is available to rule out/in Abdominal Cocoon secondary to TB. Commonly, these patients present in surgical emergencies and are diagnosed on laparotomy. Usual treatment includes surgical resection of the affected bowel and Anti-tuberculous drugs. Abdominal tuberculosis has a propensity to affect nearly every organ in the abdominopelvic region, however, the distal region of the ileum is the most common site of involvement among various gastrointestinal tuberculosis.[1,6,7]

This case report aims to bring attention to rare features of abdominal Tb leading to intestinal obstruction in children, that is abdominal cocoon, so that timely diagnosis and treatment could be given to curb child morbidity and mortality.

Case report:

We present a unique case of an 11-year-old girl from Faisalabad referred from the surgery department to the radiology department. She had no history of TB contact, weight loss, night sweats or chronic cough. Her presenting complaints were recurrent abdominal pain for 3.5 months, which was sometimes associated with vomiting and constipation. Her laboratory and clinical examination findings were insignificant. Her history depicted three ultrasounds done from other facilities. The first scan showed hepatomegaly. Moderate free fluid in the abdomen, and an adnexal hemorrhagic cyst. The second scan illustrated no free fluid and the resolution of the adnexal cyst. The third scan exhibited dilated fluid-filled small bowel loops and no adnexal cysts. The patient was then admitted to our facility. Her x-ray abdomen was done in erect and supine positions. The patient did not settle so Computerized tomography (CT) abdomen with IV contrast was advised. The differential diagnosis made included: encapsulating peritoneal sclerosis secondary to abdominal tuberculosis, congenital peritoneal encapsulation, internal hernia, peritoneal carcinomatosis, and mesothelioma. Her surgery was planned and preoperative findings showed reactionary fluid, 1x1 cm perforation in the ileum proximal to the ileocecal junction, and caseating tubercle over gut loops.

Excision of the fibrous sac was sent for histopathology. Adhenolysis and ileostomy were done. Whereas, no adnexal cysts were spotted during surgery. Histopathology showed epithelioid granulomas with caseous necrosis, which are used by clinicians as a strong argument to establish the diagnosis of TB. M.TB culture was not performed.

Post-operatively the patient was kept in pediatric intensive care unit (PICU) for the period of 5–7 days with intensive monitoring. I.V. fluids, blood and plasma were given as per requirements. She tolerated oral feed on 5th post-operative day, sutures were removed on 8th post-operative day and she were discharged. Oral ATT was started in consultation with the pediatric medical team. No post-operative complications were reported and the patient had a smooth recovery. Periodic follow up was satisfactory. Patient gained weight and no recurrence of TB was seen.

Fig 1: CT images of the patient

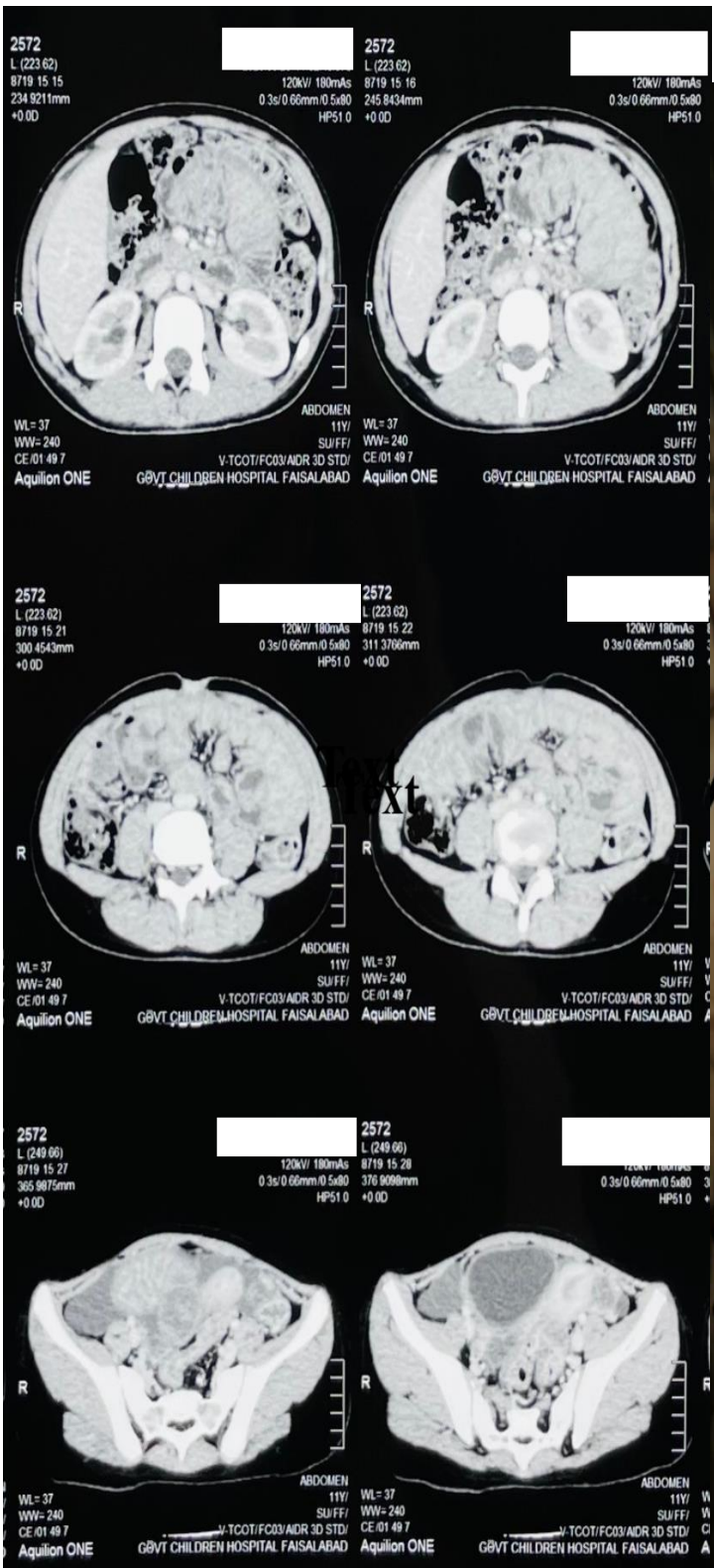
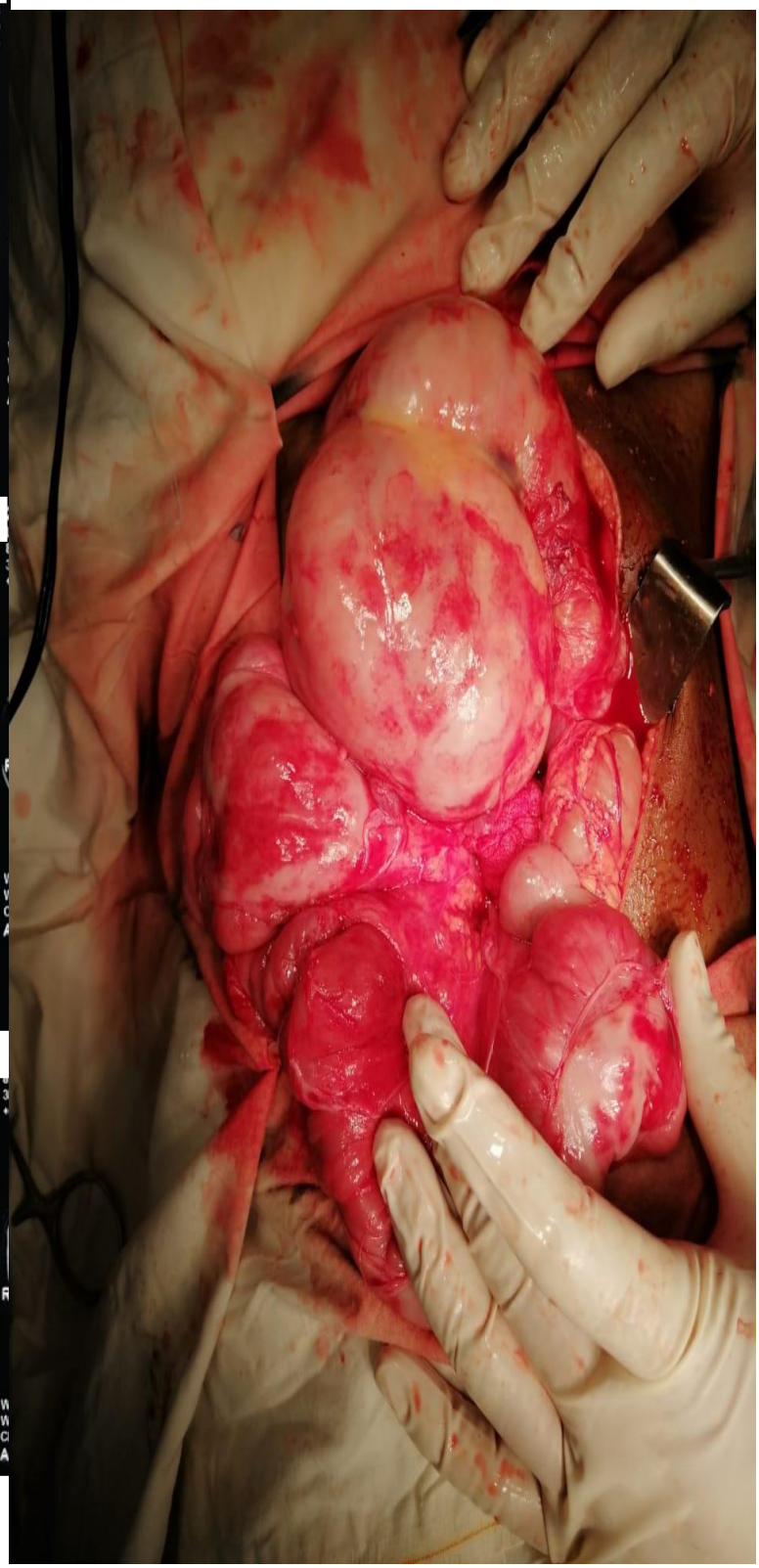


Fig 2: Intra-operative view of the abdominal cocoon



Discussion

The abdominal cocoon is quite an uncommon reason for intestinal obstruction. Xia J et al. 2018 stated that the Abdominal cocoon (AC) has an erratic spread. It can occur at any age and is not specific to a particular gender. Its diagnosis is also quite difficult to make due to the non-specificity of signs and symptoms. While several hypotheses have been suggested, the exact cause and development of (AC) remain elusive. Some theories connect AC to young girls in tropical regions, suggesting factors like retrograde menstruation, viral infections, peritonitis via fallopian tubes, and immune-mediated tissue damage. Population, location, and gender could play a role in AC's aetiology. However, recent cases challenge these theories, as AC affects adults (male and female), premenopausal women, children, and those in temperate regions. Generally, it is diagnosed on exploratory laparotomy, whereas, technological advancement has enabled the preoperative AC diagnosis, for instance, CT scans and contrast studies. The histologic findings of excised membranes typically reveal dense fibrous tissue composed of collagen and infiltration of some inflammatory cells. The Chinese literature reviewed depicted 81 cases, among whom the average age was 35.5 years with the range of 6 years to 78 years. All cases were found to have greyish-white fibrous membranes in the shape of a cocoon.[8] AC can be classified into three categories based on the degree of encapsulation by the surrounding membrane, the encapsulation can vary from partial encapsulation of the intestine to encapsulation of entire intestine along with intraperitoneal organs. [9]

Finding abdominal cocoon-related bowel obstruction in children due to abdominal tuberculosis is rare. Singal R. et al. 2017 reported that 17 patients were admitted to the emergency ward, displaying symptoms indicative of acute intestinal obstruction from a rural area of India. The cohort exhibited an average age of 15.3 years, with an age range spanning from 9 to 16 years. Common clinical presentation consisted of abdominal pain, bilious vomiting, constipation, and abdominal distention. Out of these 17 patients under investigation, four individuals had previously initiated treatment for pulmonary tuberculosis, with treatment durations spanning from 3 weeks to 4 months, albeit with irregular adherence. Furthermore, a history of contact with tuberculosis was ascertained in 3 out of the 17 patients, accounting for 17.6% of the total cohort.

Because of these unusual and rare symptoms/examination findings/imaging, it becomes very difficult to diagnose this medical issue in children and treat it. Isolating bacilli by culture is even more problematic due to malnutrition and poor hygiene. The absence of a single definitive diagnostic test for tuberculous abdominal cocoon underscores the high significance of employing imaging modalities in the diagnostic process.[5]

Rastogi R reported a case of thirty years old male with issues of vomiting, abdominal pain for a few days, and a family history of pulmonary tuberculosis. His clinical exam depicted mild abdominal distension. On abdominal radiography, the small bowel was mildly dilated in the mid-abdominal area and ultrasound revealed small intestine clustering in the same region along with some enlarging of mesenteric lymph nodes. On the chest X-ray, the upper right lobe of the lung exhibited fibro-calcifications. Furthermore, CT abdomen depicted clusters of intestinal loops with thick membranous sac encapsulating them, stomach and duodenal dilation, and lymphadenopathy. The diagnosis of abdominal cocoon secondary to TB was confirmed on exploratory laparotomy. CT proved to be quite a useful tool in making the definitive diagnosis and wanting prompt intervention.[10]

These case scenarios differed from our case as they depicted that abdominal cocoon can occur in any age group and in any gender. Besides advancements in technology, its diagnosis is still a herculean task for physicians. Therefore, it needs extensive research to establish a proper understanding of the cryptic nature of this disease and to clarify the exact aetiology.

Conclusion

This case report sheds light on the exceptional presentation of abdominal cocoon secondary to tuberculosis leading to intestinal obstruction. Abdominal cocoon, though less common, remains a significant health concern, as it can pose diagnostic challenges due to its non-specific symptoms and clinical findings. This case emphasizes the importance of considering tuberculous AC as a differential

diagnosis in cases of unexplained abdominal pain. It underscores the critical and invaluable role of imaging in the diagnostic process when faced with atypical presentations of tuberculosis and secondary AC. In resource-limited settings, increasing awareness among healthcare providers about the various manifestations of tuberculosis and AC is crucial for timely diagnosis and prompt initiation of appropriate intervention to prevent complications and reduce morbidity and mortality in children.

Ethical Consideration

Data was anonymized to protect patient privacy. Patient and parents' consent was acquired before publication.

References

1. World Health Organization. Roadmap towards ending TB in children and adolescents. Geneva Switzerland; 2018.

2. Lal SB, Bolia R, Menon JV, Venkatesh V, Bhatia A, Vaiphei K, et al. Abdominal tuberculosis in children: A real-world experience of 218 cases from an endemic region. JGH Open [Internet]. 2020;4(2):215–20. Available from: http://dx.doi.org/10.1002/jgh3.12245

3. Wong SA, Lee Meijuan D, Loh SW, Thoon KC, Tan NWH, Chong CY. Pediatric abdominal tuberculosis in Singapore: A 10-year retrospective series. Glob Pediatr Health [Internet]. 2020;7:2333794X2090395. Available from: http://dx.doi.org/10.1177/2333794x20903952

4. Fatima R, Yaqoob A, Qadeer E, Hinderaker SG, Ikram A, Sismanidis C. Measuring and addressing the childhood tuberculosis reporting gaps in Pakistan: The first ever national inventory study among children. PLoS One [Internet]. 2019;14(12):e0227186. Available from: http://dx.doi.org/10.1371/journal.pone.0227186

5. Singal R, Satyashree, Mittal A, Sharma BP, Singal S, Zaman M, et al. Tuberculous Abdominal cocoon in childern– A Single Centre Study in remote area in northern India. Med Pharm Rep [Internet]. 2017;90(2):179–84. Available from: http://dx.doi.org/10.15386/cjmed-725

6. Sartoris G, Seddon JA, Rabie H, Nel ED, Schaaf HS. Abdominal tuberculosis in children: Challenges, uncertainty, and confusion. J Pediatric Infect Dis Soc [Internet]. 2020;9(2):218–27. Available from: http://dx.doi.org/10.1093/jpids/piz093

7. Bhatta OP, Verma R, Shrestha G, Sharma D, Dahal R, Kansakar PBS. An unusual case of intestinal obstruction due to abdominal cocoon: A case report. Int J Surg Case Rep [Internet]. 2021;85(106282):106282. Available from: https://www.sciencedirect.com/science/article/pii/S2210261221007847

8. Xia J, Xie W, Chen L, Liu D. Abdominal cocoon with early postoperative small bowel obstruction: A case report and review of literature in China. Medicine (Baltimore) [Internet]. 2018;97(25):e11102. Available from: http://dx.doi.org/10.1097/md.0000000000011102

9. Sharma V, Mandavdhare HS, Rana SS, Singh H, Kumar A, Gupta R. Role of conservative management in tubercular abdominal cocoon: a case series. Infection [Internet]. 2017;45(5):601–6. Available from: http://dx.doi.org/10.1007/s15010-017-1012-5

10. Rastogi R. Abdominal cocoon secondary to tuberculosis. Saudi J Gastroenterol [Internet]. 2008;14(3):139. Available from: http://dx.doi.org/10.4103/1319-3767.41733