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Case Report

Bilharzioma of Meckel's Diverticulum: A Case Report of Unusual Location

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Abstract: Bilharzioma is pseudotumor caused by Schistosoma infestation that is localized most common in large bowel. Different localizations of Bilharzioma such as breast, lung, spinal cord and female genitalia have been reported. In this unusual case report, for a 12-year-old boy, atypical location of bilharzioma was found after operation for a presumed appendicitis. It was localized at the tip of Meckel's diverticulum (Figure 1). The diagnostic histological findings of the biopsy specimens are shown in Figure 2 and figure 3. The patient was from an endemic area of bilharziasis in Dhamar governorate. He was suffering of chronic right iliac fossa pain identical to appendicitis. The pain was intermittent and associated with irregular tenesmus, diarrhea and low grade fever. There was no history of previous treatment for bilharziasis. At discharge this patient was treated with Praziquantel and at follow-up one month later there was complete recovery. This case report shed the light on the importance of histopathologic analysis of Meckel's diverticulum as it may be mistaken for an inflammatory or malignant disease.

Keywords: Bilharzioma, Meckel's Diverticulum, Boy

1. Introduction

Schistosomiasis or bilharziasis is an infection caused by trematode flatworms of any species of the genus Schistosoma [1]. Bilharzioma is the mass resulted from trematode worms' infection. So, bilharziomas are thought to be caused by a reaction of the host to the eggs produced by worms in a single site [2]. Al Ghorab MM [3] defined bilharzioma as a localized mass of fibrous and inflammatory tissue, which contains numerous eggs frequently involving the serosa and mesentery [3]. Bilharzia ulcerations or tumors have been described in the bladder, in the intestine and even in the skin [4]. In addition to these sites, several studies reported other localizations including breast, lung, spinal cord and female genitalia [5-9]. To our knowledge; localization at the tip of Meckel's diverticulum was not reported yet. This unusual case of

bilharzioma that was localized at the tip of Meckel's diverticle in a 12-year-old patient, from an endemic area of bilharziasis in Dhamar governorate, was presented with chronic right iliac fossa pain identical to appendicitis. A Meckel's diverticulum is a vestigial remnant of the omphalomesenteric (vitellointestinal) duct that is found 45-90 cm proximal to the ileocecal valve. It is the most common malformation of the gastrointestinal tract and is present in approximately 2% of the population [10].

2. A Case Report

A 12-year-old boy was presented to emergency department in Al-Wahdah University hospital in Mabar, Dhamar governorate on 28th of April 2008 complaining of intermittent pain in right iliac fossa associated with irregular tenesmus,

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diarrhea and low grade fever. There was history of nausea but no vomiting. There was no constipation but sometimes there was history of tenesmus with mucus discharge with no macroscopic per rectum bleeding. No history of previous treatment for bilharziasis. The patient was first examined by the surgeon and then was referred to pediatrician for consultation.

By physical examination of the abdomen, there was no distention, symmetrical abdominal contour and free hernial orifices. By palpation, the abdomen was soft no guarding or rigidity, but mild to moderate tenderness in right iliac fossa was found. So, an early acute appendicitis was suspected as provisional diagnosis, therefore the patient was returned back to the surgeon. Abdominal ultrasound was done that reported edematous appendicitis. Laboratory investigations were normal except white blood cells that were 10,200/mm³. So, the patient was prepared for appendectomy that was performed in the same day of presentation after short preparation with intravenous fluids and antibiotics prophylaxis.

The intra-operative finding was surprising. The appendix was not inflamed and no signs of intestinal obstruction. This finding necessitated surgeon to explore for Meckel's diverticulum.

A mass measuring about 4×5 cm was found at the tip of Meckel's diverticulum (Figure 1).

Resection of the diverticle was done with end to end anastomosis of small bowel.

Biopsy was sent to histopathological analysis. The result of biopsy specimens was a bilharzioma (Figures 2, 3).

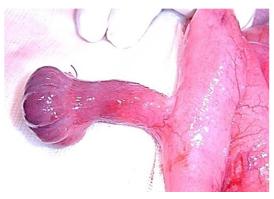


Figure 1. Meckel's diverticulum with bilharzioma at its tip.

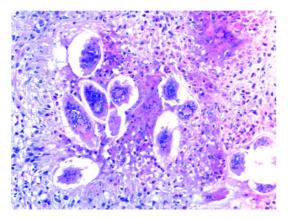


Figure 2. Collection of distorted bilharzial ova surrounded by inflammatory cells that are predominated by intact and degenerated eosinophils.

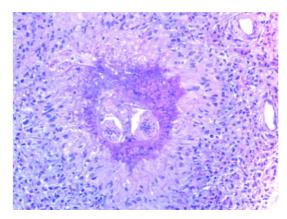


Figure 3. Small collection of distorted bilharzial ova and degenerated eosinophils surrounded by granuloma consist of histiocytes, lymphocytes and hyaline fibrous tissues.

Postoperative hospitalization was uneventful and smooth. The patient was discharged home on 6th postoperative day. Appropriate dose of Praziquantel was given on discharge. At follow-up one month later there was complete recovery.

3. Discussion

Schistosomiasis or bilharziasis is an infection caused by trematode flatworms of any species of the genus schistosoma [1]. It is responsible for 200,000 deaths annually with 200 million people affected globally [11]. Schistosoma mansoni, haematobium and japonicum are the commonest cause of this infection [1]. The earliest case of human Schistosomiasis was described over 5000 years ago in an Egyptian adolescent mummy [12]. The life cycle of this parasite involves an intermediate host (snail) that carries the miracidium. The miracidium matures into a sporocyst and is released into water as cercaria, which penetrate the human skin and becomes a schistosome. In schistosoma mansoni, the schistosome migrates to the portal blood and then matures into adult worms, which release numerous eggs that penetrate tissues. The eggs elicit a granulomatous reaction with abundant eosinophils [1]. When the infection results in formation of a mass, this is referred to as a "Bilharzioma". Bilharziomas are thought to be caused by a reaction of the host to the eggs produced by one or more pairs of worms in a single site. The term "Bilharzial polyp" was first coined in 1856 in Egypt after a German naturalist, Theodor Maximillian Bilharz. Later, in 1950, Michael Gelfand working in Zimbabwe suggested the term "Bilharzioma" a term, that continues to be used worldwide [2]. The commonest location for bilharzioma is rectosigmoid [13]. Bilharziomas have been described also in the bladder, in the intestine and even on the skin [4]. Also, they have been reported in other sites including breast, lung, intestine and the genital and perineal areas [5-8, 13, 14]. Atypical bilharzioma localized at the tip of Meckel's diverticle, as in this case, is extremely rare and was diagnosed only by histological examination. Reviewing the literature there was no similar case found. Bilharziomas are usually asymptomatic until they cause obstruction and are often picked up incidentally [13]. Gastrointestinal schistosomiasis may manifest as hepatic or

intestinal disease. Hepatic involvement is more common and results in periportal fibrosis leading to portal hypertension. The clinical spectrum of intestinal schistosomiasis is extremely broad and may be mistaken for inflammatory or malignant disease [15]. So, the bilharzioma is to be considered when the surgeon encounters a mass of any part of gastrointestinal tract.

4. Conclusion

We reported unusual case of atypical localization of bilharzioma at the tip of Meckel's diverticulum that had non-specific presentation and was treated by surgical excision and Praziquantel. The finding shed the light on the importance of histopathological analysis of the resected Meckel's diverticulum as it may be mistaken for inflammatory or malignant disease.

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