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Case Report

Report of a Case with Small Bowel Obstruction by a Rare Parasite (Pentastomiasis)

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ABSTRACT

Linguatula serrata (L. serrata), an aberrant arthropod of the Pentastomida phylum, inhabits the canine respiratory system as its final host. Intermediate hosts include humans and herbivores. Humans can be infected via consumption of raw or under-cooked liver or lymph nodes of sheep, goats, and cattle. A few human cases have been reported from Iran. Here, we have reported a case of pentastomiasis infection in a 55-year-old man who presented with fever, abdominal pain, abdominal distension, and weight loss. Small bowel transit revealed partial obstruction in the small intestine. Abdominal CT scan showed increased small bowel wall thickness. Both laparoscopy and biopsy findings favored parasitic granuloma from a pentastomiasis infection. He was administered praziquantel and albendazole. After two weeks of therapy the patient recovered.

Keywords: Linguatula serrata; Pentastomiasis; Abdominal pain

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INTRODUCTION

Linguatula serrata (L. serrata), of the Pentastomida phylum, is an aberrant arthropod, also known as the tongue worm. Adult males measure 18–20 mm while females measure 80–130 mm in length(1,2). The parasite inhabits the canine respiratory system as its final host(3,4). Transmission to intermediate hosts occurs through nasal secretions. Intermediate hosts include humans and other mammals, but are often herbivores such as cattle, goats, sheep and other ruminants that have ingested plants contaminated with parasite eggs(3). Larva reaches the mesenteric lymph nodes (MLN), liver, and lungs, among others, in which it develops into an infective nymph after six to nine moltings. It usually lies in a small cyst surrounded by a viscid turbid fluid. The final host becomes infected by eating the infected viscera(5).

Humans can be infected via consumption of raw or under-cooked liver or lymph nodes of sheep, goats and cattle,(6) either by ingestion of the nymphs of L. serrata (adult stage) resulting in nasopharyngeal linguatulosis or Halzoun syndrome (including pharyngitis, salivation, dysphagia, and coughing) (2,7) or remain asymptomatic in case of visceral linguatulosis(7).

Human cases of Linguatula have been reported from Africa, Southeast Asia, the Middle East and

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America(6, 8, 9). To date, there have been few reports on human pentastomiasis, (10) which is a rare condition in Iran. Here, we discuss a case of pentastomiasis infection in a 55-year-old man. The patient presented with abdominal pain, and abdominal obstructive signs and symptoms, in addition to pathologic and imaging features, and clinical manifestations. Furthermore, an accompanying mini-review of reports from the Middle East will be discussed.

CSAE REPORT

A 55-year-old rancher, and current resident of Zanjan, Iran since birth, was admitted to our hospital with complaints of abdominal pain and weight loss. He had suffered from paroxysmal epigastric pain since 3 years prior, which gradually worsened, and was aggravated by food consumption. The pain did not radiate, nor was it altered by changes in position. He had a 15 kilogram weight loss during a 5-month period, and due to his cibophobia, had lost his appetite. There was no significant past medical history. His habits consisted of using opioids and smoking for 20 pack-years.

At the time of admission he had a body temperature of 39.5ºC. He was quite pale, but not icteric. Upon physical examination the patient had abdominal distension, though no obvious (rebound) tenderness was detected. The liver was palpable, with blunt edges. Laboratory results showed anemia (hemoglobin: 10.5 g/dL, MCV: 87.4 fL) and malnutrition (serum albumin: 1.6g/dl). Blood platelets (376×103/µL) were normal, whereas the erythrocyte sedimentation rate (ESR: 31 mm/hr), C-reactive protein (86.7 mg/dL), and eosinophil counts (4%) were elevated. The patient had neutropenia, with a white blood cell count of 3.9×103/µL. Liver function tests (AST: 10 U/L, ALT: 20 U/L) were normal. His serum iron (30 µg/dL) was below normal. In addition, the blood culture was negative for any microbial growth.

During his admission, the patient had right lower quadrant tenderness and bloody stool. Considering the possibility of Crohn's disease, anti-saccharomyces cerevisiae antibodies (IgA and IgG) were negative. His stool specimen revealed 3+ occult blood, without any fatty drops or trophozoite of the parasite. Ultrasound of the abdomen and pelvis, in addition to a colonoscopy and abdominal vessel angiography were performed on the day following admission, all of which were normal. A small bowel transit was subsequently performed, which revealed partial obstruction in the small intestine (Figure 1). Abdominal CT scan showed increased small bowel wall thickness. Both laparoscopy and biopsy were performed. The laparoscopy showed a thickness of the small intestine (ileum) wall and its meso, without any evident obstruction. In addition, multiple masses were seen in the anti-mesenteric area. Based on a suspected primary diagnosis of lymphoma, he was referred to a surgeon and underwent segmental intestinal resection. Histopathologic examination revealed parenchymal tubercles that contained several parasitic larvae of pentastomids, numerous inflammatory cell infiltrations, and a granulomatous reaction with surrounding fibrotic bundles (Figures 2,3). The parasite diagnosis of pentastomid was performed based on morphologic features and confirmed by a professional parasitologist. At this point, it was revealed that the patient had consumed the liver of a lamb (kebab) with poor hygiene several months prior to his admission. Following the diagnosis, he was given praziquantel (0.5 g, tid) and albendazole (0.75 g, bid). After 14 days of therapy the fever had disappeared and abdominal pain was diminished. The patient made an uneventful recovery after 2 weeks of therapy and he was discharged from the hospital.

DISCUSSION

Human pentastomiasis is a rare parasitic zoonosis caused by pentastomids. Most reported human cases have been incidentally found in autopsies, except for cases in the African region. There are 10 known pathogenic pentastomid parasites in which 2 families, Linguatulidae and Procephalidae, consist of species of the genus Linguatula and Armillifer, both zoonotic parasites that are important in veterinary and human medicine(1). L. serrata is the most common cause of pentastomiasis in humans, accounting for 90% of infections (11,12). The prevalence of L. serrata in animals has been reported in various areas such as Lebanon, Turkey, Sudan, and Iran; carnivores such as dogs and foxes are the definitive hosts of this parasite. (Table 1) A high prevalence rate (62.2%) of L. serrata in stray dogs in Shahre-Kord, Iran has been reported (2,13-15). There are few articles reporting human infections with L. serrata. Close contact between animals and the intermediate host (i.e., humans) plays a pivotal role in transmission of L. serrata in the above mentioned areas. Human involvement is reported from developing countries, particularly due to the lack of food and water sanitation in these regions(7). In addition, our patient had a history of consuming raw lamb liver. According to the literature there are

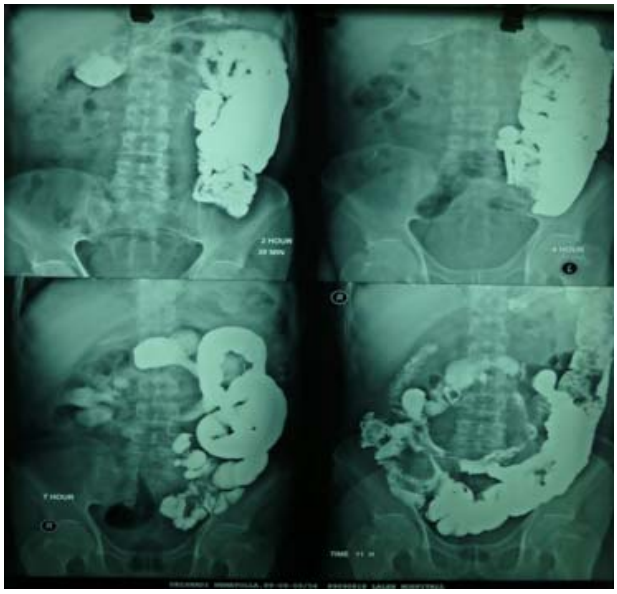

Figure 1: Small bowel transit showing intestinal obstruction.

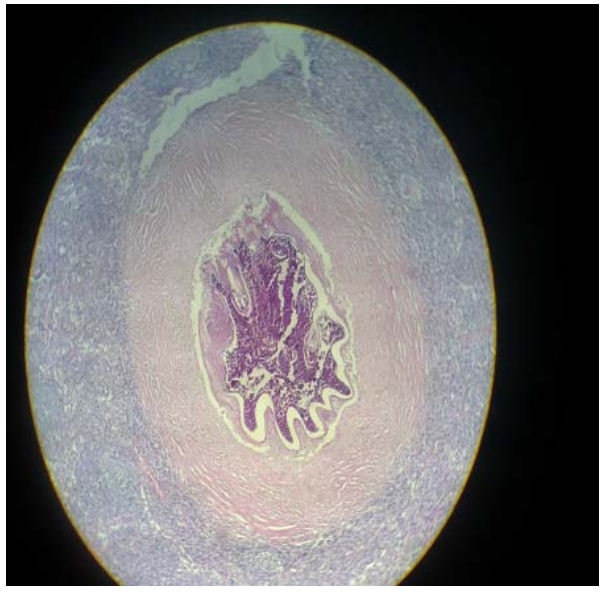

Figure 2: Microscopic examination stained by hematoxylin and eosin (H&E)
showing a parasite (pentastomid) surrounded by fibrotic bundles (100x).

Figure 3: Gross view for L. serrata located in the patient's small intestine.

only 10 reports of pentastomiasis from Iran(16,17). As mentioned in our reported case the infection usually has subclinical manifestations. Our patient presented with chronic abdominal pain for a 3-year duration. According to the literature, there are 2 types of infections in humans, nasopharyngeal pentastomiasis which presents as a severe illness with symptoms of thoracic pain and upper respiratory tract bleeding. This type is termed Halzoun syndrome which has been described previously by Schacher et al.(18) Visceral pentastomiasis, the usual and most common form of the disease, infects internal organs such as the

esophagus, stomach, ileum, colon, appendix, rectum, mesentery, gallbladder, lungs, pleura, omentum, bladder, adrenal glands, heart (pericardium), lymph nodes, and skin, however the liver is the most commonly affected site(10).

Our case was a rare form of visceral pentastomiasis. In the current report, abdominal pain with evidence of small bowel obstruction noted on CT scan, in addition to multiple intra-abdominal masses were odd presentations of a parasitic infection which caused the patient to undergo surgery due to suspicion of malignancy.

According to the literature, humans are usually tolerant to pentastomiasis and treatment is not necessary unless symptoms are present. In our case the patient was treated with medical therapy after definitive diagnosis, with subsequent symptom resolution.

To summarize, this was the eleventh report of pentastomiasis from Iran. We have presented a rare case of pentastomiasis infection with symptoms of high fever, abdominal pain, lower GI bleeding, and significant weight loss with involvement of the small intestine and intra-abdominal lymph nodes, but with normal WBC and eosinophil counts on CBC. Also, identification of the larva on the histologic examination was a rare event with academic and practical significance. Since there are a lack of documented noninvasive diagnostic methods for the detection of parasitic infections, emphasis should be placed on diagnosing these infections particularly in endemic areas. Conclusively, pentastomiasis should be considered in the differential diagnosis of odd abdominal symptoms, notably in patients with histories of consuming uncooked food in endemic areas. However, the definitive diagnosis is the pathology report.

Table 1:Characteristics of pentastomiasis case reports.

	Age (years) /gender	Symptoms	Histologic specimen	Time/region
Current case	55/male	Abdominal pain	Live parasite	2012/Tehran
Lai C, et al. (10)	3/female	Abdominal pain	Live parasite	2010/China
Anaraki Mohammadi, et al. (19)	10/male	Nasopharyngeal	Live parasite	2008/Tehran
Siavashi, et al.(20)	23/female* 43/female* 27/male*	Nasopharyngeal	Live parasite	2002/Tehran
Maleky (21)	28/female	Nasopharyngeal	Live parasite	2001/Tehran
Yeganeh Moghadam, et al.(22)	30/male	Nasopharyngeal	Live parasite	2001/Kashan
Sadjjadi, et al.(23)	30/female	Nasopharyngeal/sore throat	Parasite	1998/Shiraz

*Three members from one family.

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