Unusual cause of small intestine obstruction in a child

Small intestine anisakiasis: report of a case

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Abstract

Introduction: Anisakiasis is caused by human infection by the anisakis larvae, a marine nematode found in undercooked or raw fish. Infection with the parasite *Anisakis simplex* is common in Japan and northern European countries. With the increased popularity of eating sushi and raw fish infection with anisakis is expected to rise.

Case presentation: We present the case of a 14-year-old boy who had eaten sushi 3 days before the onset of symptoms and had small bowel obstruction caused by enteric anisakiasis. To the best of our knowledge this is the first reported case of intestinal anisakiasis presenting as a bowel obstruction in a child.

Conclusion: Enteric anisakiasis is very rare, and its diagnosis is usually made after laparotomy. Nevertheless, when signs of acute abdomen develop after the ingestion of raw fish, such as sushi or sashimi, the possibility of enteric anisakiasis should be considered.

Keywords

Anisakiasis, enteric anisakiasis, small intestine, intestinal obstruction, ileus, children, sushi

Introduction

Anisakiasis is a human parasitic infection of the gastrointestinal tract caused by the consumption of raw or undercooked seafood containing larvae of the nematode Anisakis simplex, which has a life cycle involving fish and marine mammals. They are infective to humans and cause anisakiasis.¹⁻⁴ Humans are only accidental hosts to this parasite. Anisakis poses a risk to human health in two ways: through intestinal infection with worms caused by eating of underprocessed fish, and through allergic reactions to chemicals left by the worms in fish flesh.²⁻⁶ Anisakiasis usually occurs in the stomach, and can be easily diagnosed by digestive tract endoscopy.⁷ On the other hand, enteric anisakiasis is very rare, with only few cases reported in literature, and its definitive diagnosis is difficult and typically made only after laparotomy performed as the treatment of acute abdomen or an intestinal obstruction.³⁻⁶ Tissue damage is due to invasion of the gut wall and development of eosinophilic granuloma, ulcer, or perforation.^{3-6,8} Within hours after ingestion of infective larvae, severe abdominal pain, nausea, and vomiting may occur. Occasionally, the larvae are regurgitated. If the larvae pass into the

bowel, a severe eosinophilic granulomatous response may also occur 1 to 2 weeks following infection, causing symptoms mimicking Crohn's disease.^{4–6} Infection with the parasite *A. simplex* is common in Japan and northern European countries.³ With the increased popularity of eating sushi and raw fish infection with anisakis is expected to rise.

We present the case of a 14-year-old boy who had small bowel obstruction caused by enteric anisakiasis and to the best of our knowlage this is the first reported case of intestinal anisakiasis presenting as a bowel obstruction in a child, and first reported case in Croatia.

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Case report

Medical history

A 14-year-old boy was presented to our pediatric surgery emergency department because of sudden onset of diffuse abdominal pain, nausea and vomiting that begun the day before. The patient had no past medical history. Parents did not mention that the boy had eaten raw fish when the case history was taken.

Clinical features

The patient was conscious, dehydrated, his blood pressure was 120/80 mmHg, pulse 80 beats/min, and body temperature 37.3° C. On physical examination, the patient had direct and rebound tenderness on the whole abdomen, especially in right lower part, accompanied with abdominal distension. The white blood cell count was 12.27×10^{9} /L (normal range; $4.4-11.6 \times 10^{9}$ /L), the proportion of eosinophils in white blood cell differential count was elevated (13.4%) and C reactive protein level was 28.1 mg/L (normal range; 0-5 mg/L). All other laboratory examinations showed normal values. Plain radiography of the abdomen showed ileus – many loops of small intestine and air-fluid levels.

Differential diagnosis

The differential diagnosis includes appendicitis, Meckels diverticulitis, mesenteric lymphadentis, Crohn's disease, eosinophilic gastroenteritis, and other parasitic infections.

Operative findings

An emergency laparotomy was performed, which revealed inflammation and edematous changes of the terminal ileum (Figure 1) and enlarged mesenteric lymph nodes. No stenosis or perforation was found in affected part of small bowel. Appendectomy and extirpation of enlarged lymph node were performed.

Pathologic examination

Histopathological examination revealed the edema and infiltration of eosinophils in muscular layer and serosa of the appendiceal wall (Figure 2). Hence, the cause of small bowel obstruction was suspicious to be enteric parasitosis.

Outcome, prognosis and follow-up

After the operation and histopathological examination, we took a detailed history of the diet, and the parents remembered that the boy had eaten sushi, 3 days before surgery. Elevated levels of specific IgE (256.1 kU/L) to *A. simplex* and total IgE (410.2 kU/L) were found and the diagnosis of enteric anisakiasis was confirmed. The patient was treated with symptomatic therapy and discharged on the eighth day after surgery following an uneventful course of recovery. The patient was



Figure 1. Intraoperative findings. Inflammation and edematous changes of the terminal ileum.

followed-up for 15 months and no recurrence of disease was found.

Discussion

Sushi and Japanese cuisine are quite popular, even in Mediterranean countries outside Asia. As a result of increased popularity of eating sushi and raw fish the number of reported cases of anisakiasis in United States and more recently in Spain and other Mediterranean countries is increasing.^{3,9,10} This is the first reported case of anisakidosis in Croatia. This is also the first reported case in a child; all previous published reports were in adults.^{3-6,9,10} The disease of anisakiasis is caused by the third stage larvae of the Anisakis spp, mainly A. simplex, after the ingestion of raw or insufficiently cooked fish. The adult anisakis lives in the stomach of marine mammals such as whales and dolphins. Crustaceans are the first intermediary hosts. The second intermediary hosts include various species of fish and some cuttlefish. Humans are only accidentally contaminated. When ingested by humans, the anisakis larvae migrate to the digestive tract and cause various symptoms.^{1,3,5,7-10} Anisakis larva is susceptible to high temperatures, being killed in 1 s at 60°C and in 15 min at 50°C, hence cooking fish will effectively prevent anisakiasis.^{3,7} Anisakiasis is classified as either the luminal or the invasive form, according to the presence of bowel wall invasion by snisakis larvae.^{3,5} The luminal form does not cause major clinical symptoms, but the invasive form can.^{5,7} The invasive form is subdivided into gastric and intestinal types, according to the penetration site. Anisakiasis is caused by two main mechanisms: allergic reactions and direct tissue damage. The former ranges from isolated urticaria and angioedema to life-threatening anaphylactic shock associated with gastrointestinal symptoms. Allergic reactions can occur after the primary infection of anisakis with exposure to allergens in the food. Direct tissue damage is due to parasite invasion of the gut wall, development of eosinophilic granuloma, or perforation.^{3,5,7} Cases of gastric anisakiasis are more common than those of enteric anisakiasis and enteric anisakiasis is rarely reported. Gastric anisakiasis accounted for 95.6%, enteric anisakiasis for 4.1%, and other sites for 0.3%.3 In all reported cases of enteric anisakiasis resection of involved intestine has been performed due to bowel stenosis or perforation.^{3–6,9,10} In our case there was no perforation or significant obstruction and resection was not performed. Cases of intestinal intussusceptions due to anisakiasis have also been reported.¹¹ While gastric anisakiasis develops within 1 day after intake of anisakis nematodes, the onset of enteric anisakiasis is delayed from 1 to 5 days, making it difficult to ascertain ingestion of raw fish. Moreover, a definitive diagnosis of enteric anisakiasis is done only after confirmation of the presence of anisakis, usually by surgical resection, while gastric anisakiasis is immediately confirmed and treated endoscopically. Increased eosinophil levels are

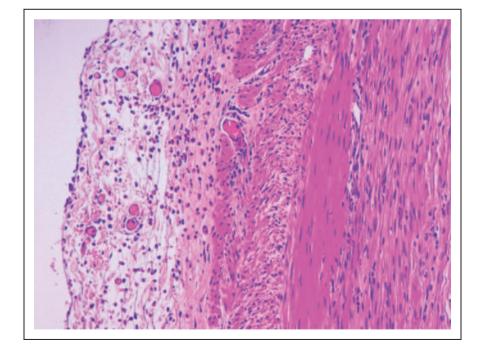


Figure 2. Histopathologic findings. Infiltration of eosinophils in muscular layer and serosa of the appendiceal wall. Haematoxylin and eosin staining, conventional light microscopy, magnification $\times 200$.

observed in less than half of the patients with anisakiasis, and when the eosinophil levels are increased, they tend to be normal on the first day in hospital and gradually become enhanced later.^{2,4,8} In our case eosinophil level was elevated at admission. The most important and useful tool in diagnosing enteric anisakiasis is obtaining an accurate history of the present illness and determining whether the patient has eaten raw fish before the onset of symptoms. Evidence of small intestinal enlargement and focal thickening of the intestinal wall seen on ultrasonography and computed tomography can be helpful, but it is difficult to make a definitive diagnosis based on these findings alone. Usually, a diagnosis of enteric anisakiasis is made after the operation.^{3–7} Although it is possible to make a diagnosis using the immune reaction, time is required to measure the antianisakis antibody titer, and the titer may not necessarily rise in the early stage after onset.² Since anisakis larvae die in the course of time, enteric anisakiasis is generally alleviated by conservative treatment. There is no drug treatment for anisakiasis although there are few case reports reporting successful treatment with albendazole alone.¹⁰ However, symptoms usually resolve in 1 to 2 weeks when the larvae die. Corticosteroids have been recommended by some authors, but only for management of the inflammatory reaction caused by the worms. The worms cannot survive in the human body and they die eventually.¹⁰ Most patients make a full recovery.

Conclusion

Enteric anisakiasis is very rare, and its diagnosis is usually made only after laparotomy. Nevertheless, when signs of acute abdomen develop after the ingestion of raw fish, such as sushi or sashimi, the possibility of enteric anisakiasis should be considered.

Learning points

- 1. Anisakiasis is caused by the third stage larvae of the *Anisakis* spp, mainly *A. simplex*, after the ingestion of raw or insufficiently cooked fish.
- 2. Anisakiasis pose a risk to human health in two ways: through intestinal infection with worms caused by eating of underprocessed fish, and through allergic reactions to chemicals left by the worms in fish flesh
- Cases of gastric anisakiasis are more common than are those of enteric anisakiasis and enteric anisakiasis is rarely reported.
- 4. The diagnosis is usually made after surgery, the most important and useful tool in diagnosing

enteric anisakiasis is obtaining an accurate history of the present illness and determining whether the patient has eaten raw fish before the onset of symptoms.

5. When signs of acute abdomen develop after the ingestion of raw fish, such as sushi or sashimi, the possibility of enteric anisakiasis should be considered.

Declaration of conflicting interests

None declared.

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Authors' contributions

The work presented here was carried out in collaboration among all authors. Ivo Jurić and Zenon Pogorelić performed operation. Ivana Mrklić performed pathohistological analysis and report. Ivana Mrklić and Zenon Pogorelić defined the research theme, performed literature review and wrote the paper. Zenon Pogorelić, Ivo Jurić, and Ranka Despot have been involved in drafting the manuscript and revising it critically for important intellectual content. Ivo Jurić and Ranka Despot have given final approval of the version to be published. All authors have contributed to, seen and approved the manuscript.

References

- Baldwin RE, Rew MB, Johansson ML, et al. Population structure of three species of Anisakis nematodes recovered from Pacific sardines (Sardinops sagax) distributed throughout the California current system. *J Parasitol* 2011; 97: 545–554.
- Daschner A, Rodero M and Cuéllar C. Low immunoglobulin E response in gastroallergic anisakiasis could be associated with impaired expulsion of larvae. J Investig Allergol Clin Immunol 2011; 21: 330–331.
- Sasaki T, Fukumori D, Matsumoto H, et al. Small bowel obstruction caused by anisakiasis of the small intestine: report of a case. *Surg Today* 2003; 33: 123–125.
- Ishida M, Harada A, Egawa S, et al. Three successive cases of enteric anisakiasis. *Dig Surg* 2007; 24: 228–231.
- Kang DB, Oh JT, Park WC, et al. Small bowel obstruction caused by acute invasive enteric anisakiasis. *Korean J Gastroenterol* 2010; 56: 192–195.
- Pellegrini M, Occhini R, Tordini G, et al. Acute abdomen due to small bowel anisakiasis. *Dig Liver Dis* 2005; 37: 65–67.
- Hwang D, Park SI, Pack SC, et al. A case of duodenal anisakiasis with duodenal ulcer. *Chonnam Med J* 2012; 48: 73–75.
- 8. Domínguez-Ortega J, Martínez-Alonso JC, Alonso-Llamazares A, et al. Measurement of serum levels of eosinophil cationic protein in the diagnosis of acute

gastrointestinal anisakiasis. Clin Microbiol Infect 2003; 9: 453–457.

- Repiso Ortega A, Alcántara Torres M, González de Frutos C, et al. Gastrointestinal anisakiasis. Study of a series of 25 patients. *Gastroenterol Hepatol* 2003; 26: 341–346.
- Moore DA, Girdwood RW and Chiodini PL. Treatment of anisakiasis with albendazole. *Lancet* 2002; 360(9326): 54.
- 11. Chikamori F, Kuniyoshi N and Takase Y. Intussusception due to intestinal anisakiasis: a case report. *Abdom Imaging* 2004; 29: 39–41.