



Case report

Anisakiasis in a Canadian patient with incarcerated epigastric hernia

Roy Hajjar^a, Arpita Chakravarti^b, Haifaa Malaekah^c, Frank Schwenter^a, Claude Lemieux^b, Antonio Maietta^d, Herawaty Sebjang^{a,*}

^a Digestive Surgery Service, Centre hospitalier de l'Université de Montréal (CHUM), Montréal, Québec, Canada

^b Department of Medical Microbiology and Infectious Diseases, Centre hospitalier de l'Université de Montréal (CHUM), Montréal, Québec, Canada

^c King Abdullah bin Abdulaziz University Hospital, Princess Nourah bint Abdulrahman University, Riyadh, Saudi Arabia

^d Department of Pathology, Centre hospitalier de l'Université de Montréal (CHUM), Montréal, Québec, Canada

ARTICLE INFO

Article history:

Received 12 September 2019

Received in revised form 8 February 2020

Accepted 8 February 2020

Keywords:

Anisakis

Anisakiasis

Incarcerated hernia

ABSTRACT

Anisakiasis is human zoonotic parasitic infection caused by a nematode parasite called Anisakis. This infection is usually reported in Asian countries where consumption of raw seafood is common. Very few cases have been reported in North America. We present the case of a female Canadian patient with an Anisakis larvae in an incarcerated ventral hernia. Cases of Anisakis infections are exceedingly rare in western countries, with very few previous reports describing extra-gastrointestinal cases. Diagnosis is often difficult since the symptoms of anisakiasis are not pathognomonic. As the larvae cannot survive in the body, conservative treatment might be effective in intestinal anisakiasis and surgery is usually performed when complications are encountered. Preventive measures are crucial and include educating the public about the risks of raw fish consumption and the importance of visually inspecting consumed fish and freezing it before ingestion to kill the larvae and prevent the infection.

© 2020 The Authors. Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Anisakiasis is a human zoonotic parasitic disease caused by Anisakis, a nematode parasite [1,2]. This disease develops in human beings after inadvertent ingestion of raw, undercooked, pickled, smoked, salted sea food infected by the third stage larvae [1]. Most of the reported cases are from Asia where consumption of raw seafood is a common practice. Anisakiasis usually presents as a gastric or an intestinal infection, with extra-gastrointestinal disease being exceptionally rare [2]. Anisakis larvae found incidentally in incarcerated ventral hernias is an exceedingly rare event.

Case presentation

We present the case of a 45-year-old female patient who was evaluated in general surgery at the emergency room for an incarcerated epigastric ventral hernia. Her past medical history was unremarkable. The patient reported having a small midline epigastric hernia (1–2 cm) for 10 years. Her hernia was always reducible, non-tender and never altered her quality of life before.

Four days before presenting to the hospital, her hernia increased in size, became irreducible and the associated pain increased. The patient did not have any nausea or vomiting, and her bowel movements were normal, making a bowel obstruction diagnosis less likely. Physical examination revealed a non-reducible epigastric 2 × 2 cm mass. The overlying skin was normal. Abdominal radiographs were unremarkable. Her total white blood count was normal (7 200 cells/ μ L) but her relative eosinophil count was slightly elevated (4.4 %) with an absolute count of 320 cells/ μ L.

The patient underwent an emergent surgical repair of her symptomatic hernia. The hernia sac was opened and revealed the presence of omental adipose tissue. The size of the hernial defect was approximately 1.5 cm and was primarily closed. The postoperative evolution was uneventful.

Histopathological examination of the resected omental specimen consisted of fibroadipose tissue showing an important infiltration with inflammatory cells, predominantly eosinophils [Fig. 1]. In the middle of the infiltration, a parasitic organism was noted, with features of Anisakis species third-stage larvae: polymyarian type of musculature, esophagus lined by a columnar epithelium, a thin external cuticle, and Y-shaped lateral epidermal cords. Retrospectively, the patient reported ingesting raw fish in the days prior to the beginning of her symptoms and her visit to the emergency. Except for the hernia-associated pain, the patient did not present any gastrointestinal symptoms suspicious of a parasitic

* Corresponding author at: CHUM, Service de chirurgie Digestive, 1000 rue Saint-Denis, Pavillon Édouard-Asselin, Montréal, Québec, H2X 0A9, Canada.

E-mail address: Herawaty.sebjang.chum@ssss.gouv.qc.ca (H. Sebjang).

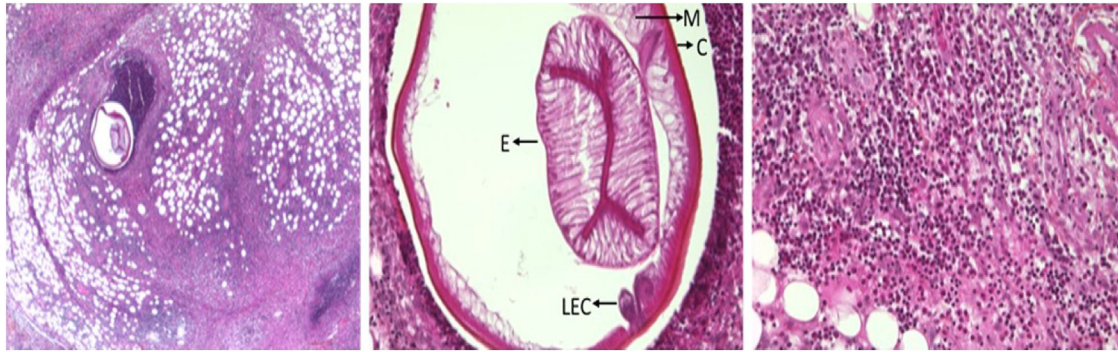


Fig. 1. Images show cross section of the hernia sac with mixed inflammatory infiltrate rich in eosinophils. An *Anisakis* larva is seen in the hernial sac, after hematoxylin and eosin staining. The nematode cross section shows an external cuticle (C) polymyarian type of musculature (several muscle cells) (M) and the lateral epidermal cords (LEC). The esophagus (E) is lined by columnar epithelial cells.

infection. She is being followed 4 years later at the surgical outpatient clinic with no gastrointestinal symptoms nor signs of hernia recurrence.

Discussion

Anisakiasis is a disease caused by the larvae of the nematode genus *Anisakis* [3]. Patients are mainly infected with *Anisakis simplex* [2]. *Pseudoterranova decipiens*, *A. physeteris*, *A. pregreffii* and *Contracaecum* species are other anisakids known to cause man infection. Marine mammals such as whales and dolphins are the definitive hosts of *Anisakis* species and adults reside within the stomach of their hosts, laying eggs that are excreted in the feces [1]. Subsequently, second-stage larvae hatch and are eaten by small crustaceans (krill) and develop into third stage larvae. These are consumed by marine fish, eel and squid and the third-stage larvae migrate into viscera and peritoneal cavity. When these intermediate hosts are consumed by marine mammals, the larvae grow into adult worms completing the lifecycle. Human beings are incidental hosts and the larvae cannot grow or replicate inside the human host.

Four major clinical types of human infection result from anisakiasis: gastric, intestinal, extra-gastrointestinal and allergic. Abdominal pain has been reported to appear 12–24 h and 5–7 days after fish ingestion with gastric and intestinal anisakiasis respectively [4]. Cases of colonic anisakiasis have also been reported but seem to be rather incidentally diagnosed cases [2]. Gastrointestinal anisakiasis accounts for more than 99 % of the cases, with non-gastrointestinal infections being exceptionally rare [1]. Anisakiasis could be associated with intestinal bleeding, obstruction or perforation. In addition to gastrointestinal symptoms, anisakiasis may sometimes cause allergic symptoms, such as urticaria, angioedema and even anaphylaxis [2,5–7].

Diagnosis is often difficult, with endoscopy being an important diagnostic modality. Since the symptoms of anisakiasis are not pathognomonic, gastric anisakiasis is often misdiagnosed as peptic ulcer, stomach tumor or stomach polyps, while intestinal anisakiasis is often misdiagnosed as appendicitis, ileus or peritonitis. Increased levels of anti-*Anisakis* specific IgE or *Anisakis* IgA/IgG may sometimes be helpful but are not always the best suited strategy in early diagnosis when the patient is symptomatic, especially since their sensitivity and specificity is not perfect, and that many asymptomatic patients ingesting raw fish might carry antibodies as well [1,2].

No definitive therapy is known for anisakiasis. Though albendazole has been shown to be effective in some reports, medical treatment is not consistently effective. The role of anti-helminthic drugs is debated. Emergent endoscopic examination and worm extraction has been suggested for gastric disease,

usually with spontaneous resolution of symptoms [2,5]. As the larvae cannot survive in the body, conservative treatment might be effective in intestinal anisakiasis and surgery is usually performed when complications are encountered or if there is clinical deterioration. While the majority of described cases have been reported in Asian countries, namely Korea and Japan, the growing popularity of raw fish in the western diet probably explains the increasingly reported cases in Europe, particularly in Italy with an association with consumption of marinated anchovies [8]. Very few cases have been reported in North America [2,8].

The presence of the parasite in the extra-gastrointestinal omental fat is unexpected and unusual. The invasive capacity of *Anisakis* spp. and, less commonly, *Pseudoterranova* spp. is nonetheless known and cases of larvae penetrating the abdominal cavity through the abdominal wall, and its association within inguinal hernias has been described [1,9]. Kawashima et al. (2019) reported a case of gastrointestinal hemorrhage due to ulcer formation caused by the invasion of the bowel wall by *Anisakis* larvae [4]. One of the cases described by Shimamura et al. (2016) showed a larvae in in the mesenteric adipose tissue with a significant inflammatory reaction [2]. In a case series, Pampiglione et al. (2002) documented extra-gastrointestinal cases of anisakiasis, namely in the omentum, spleen, mesentery and epiploic appendix [10]. In most of the cases, surgical intervention was required and the diagnosis of anisakiasis was confirmed only after the histological examination of the sample.

The larvae has been described as having possibly a predilection for penetration of the gastric greater curvature [2]. One could assume that the larvae invaded the gastric wall in our patient to reach the omentum, which in turn got incarcerated in the existing abdominal wall hernia with a local inflammatory reaction recruiting eosinophils. However, the patient did not present any signs of gastrointestinal perforation, sepsis or abdominal pain suggestive of such an event.

To summarize, the presence of *Anisakis* larvae in an incarcerated hernia in a North American patient is an exceptional event. The pathophysiology of this finding remains unclear. Preventive measures are crucial and include educating the public about the risks of raw fish consumption and the importance of visually inspecting consumed fish to detect parasites. The Food and Drug Administration recommends blast freezing of raw or semiraw food to $\leq -35^{\circ}\text{C}$ for 15 h or $\leq -20^{\circ}\text{C}$ for 7 days as this systematically kills live larvae [2]. This might potentially prevent complications of a disease that constitutes a major diagnostic challenge in Western countries.

Sources of funding

None.

Consent

Written informed consent was obtained from the patient for the publication of the present case report.

Author contribution

RH was responsible for the data collection, literature review, drafting, and critical review of the manuscript. AC was responsible for the literature review, drafting, and critical review of the manuscript. Histopathological data and figure were provided by AM. All authors were responsible for the data analysis and presentation, and the critical review of the manuscript. HS is the senior author and guarantor.

Declaration of Competing Interest

None.

References

- [1] Sasaki T, Fukumori D, Matsumoto H, Ohmori H, Yamamoto F. Small bowel obstruction caused by anisakiasis of the small intestine: report of a case. *Surg Today* 2003;33:123–5.
- [2] Shimamura Y, Muwanwella N, Chandran S, Kandel G, Marcon N. Common symptoms from an uncommon infection: gastrointestinal anisakiasis. *Can J Gastroenterol Hepatol* 2016;2016:5176502.
- [3] Toyoda H, Tanaka K. Intestinal anisakiasis treated successfully with prednisolone and olopatadine hydrochloride. *Case Rep Gastroenterol* 2016;10(1):30–5.
- [4] Kawashima K, Fujiwara T, Katakura K, Gunji N, Yokokawa A, Sakamoto A, et al. Anisakiasis in the small intestine with excessive bleeding that was difficult to diagnose endoscopically. *Intern Med* 2019;58(1):63–6.
- [5] Kang WH, Kim KS, Lee SH, Park YW. Gastric anisakiasis after eating raw salmon. *Dig Liver Dis* 2018.
- [6] Shweiki E, Rittenhouse DW, Ochoa JE, Punja VP, Zubair MH, Baliff JP. Acute small-bowel obstruction from intestinal anisakiasis after the ingestion of raw clams; documenting a new method of marine-to-human parasitic transmission. *Open Forum Infect Dis* 2014;1(2)ofu087.
- [7] Audicana MT, Kennedy MW. Anisakis simplex: from obscure infectious worm to inducer of immune hypersensitivity. *Clin Microbiol Rev* 2008;21(2):360–79 table of contents.
- [8] Guardone L, Armani A, Nucera D, Costanzo F, Mattiucci S, Bruschi F. Human anisakiasis in Italy: a retrospective epidemiological study over two decades. *Parasite* 2018;25:41.
- [9] Mitsuboshi A, Yamaguchi H, Ito Y, Mizuno T, Tokoro M, Kasai M. Extra-gastrointestinal anisakidosis caused by *Pseudoterranova azarasi* manifesting as strangulated inguinal hernia. *Parasitol Int* 2017;66(6):810–2.
- [10] Pampiglione S, Rivasi F, Criscuolo M, De Benedittis A, Gentile A, Russo S, et al. Human anisakiasis in Italy: a report of eleven new cases. *Pathol - Res Pract* 2002;198(6):429–34.