

## CASE REPORT

**Unusual case of pentastomiasis mimicking liver tumor**

Marcel Autran C Machado,\* Fabio F Makdissi,\* Leonardo F Canedo,\* Rodrigo B Martino,\* Fábio Crescentini,\* Pedro P Chieffi,<sup>†</sup> Telesforo Bacchella\* and Marcel C C Machado\*

\*Department of Surgery, University of São Paulo, and <sup>†</sup>São Paulo Institute of Tropical Medicine (LIM 06), São Paulo, Brazil

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**Correspondence**

Marcel Autran C Machado, MD, Al. Casa  
Branca 438 #101-01408-001, São Paulo, Brazil.  
Email: dr@drmarcel.com.br

**Abstract**

Pentastomiasis is a rare zoonotic disease. Almost all recorded cases of human pentastomiasis had been incidental findings at autopsy. We report an unusual case of human pentastomiasis mimicking liver tumor successfully treated by liver resection. This clinical presentation is uncommon and it was probably caused by a pentastomid that exited its cyst and migrated to the liver causing an infarct that was mistaken as a primary liver tumor. Diagnosis could not be made before the surgery. This is the first reported case of human pentastomiasis in Brazil.

**Introduction**

Pentastomiasis is a zoonotic disease of humans. Pentastomids are endoparasites of reptiles, birds and mammals. Within this group there are two important families to humans: Porocephalidae and Linguatulidae.

Pentastomiasis manifests as visceral or nasopharyngeal disease in humans. Visceral pentastomiasis occurs when a human acts as an intermediate host after ingesting infective eggs of *Armillifer armillatus* or *Linguatula serrata*. Infection in humans may occur when large numbers of parasites are present or a larval worm occurs in a vital location. Nasopharyngeal pentastomiasis occurs after ingestion of uncooked tissue of an intermediate host, such as sheep or goat, which contains encysted nymphs of *Linguatula serrata*. Pentastomiasis is common in Africa and Asia. There are very few reports of pentastomiasis in North America. In Brazil, there are two reports of pentastomiasis in rodents and no reported human case of pentastomiasis.<sup>1,2</sup> A report of an unusual case of liver pentastomiasis mimicking liver tumor treated by liver segmentectomy is presented.

**Case report**

A 53-year-old man from Mato Grosso, Brazil, was referred to our department because of a liver tumor found during routine abdominal ultrasound examination. He was a fisherman and was used to swimming in lakes and rivers. He had never been outside Brazil.

The patient presented with no gastrointestinal symptoms and had no medical history of alcohol intake, hepatitis or blood transfusions. Physical examination was unremarkable. Blood tests, serological evaluation for hepatitis and tumor markers (CEA, CA19-9, alfafetoprotein) were normal. Upper gastrointestinal endoscopy and colonoscopy were also normal.

Abdominal computed tomography revealed a 3 cm tumor with low attenuation in segment VI of the liver (Fig. 1) with a mild peripheral enhancement during the contrast infusion. The late phase showed a heterogeneous lesion. Magnetic resonance imaging disclosed a hypointense tumor on T1 and hyperintense image on T2. During the paramagnetic contrast study there was a mild enhancement of the liver lesion. As primary malignancy could not be ruled out, surgery was indicated.

A white, soft, well-defined rounded lesion measuring 2.5 cm in diameter was found in segment VI of the liver during operation, with peripheral endurance and cystic central area. Intraoperative ultrasound showed a cystic lesion with irregular echo texture surrounded by highly echogenic tissue. Resection of segment VI of the liver was carried out. During the opening of the surgical specimen, a white tubular worm measuring 2 cm in length was noted inside (Fig. 2). Pathological analysis of the surgical specimen revealed necrosis of the liver parenchyma. The recovered larva was sent to the São Paulo Institute of Tropical Medicine for further analysis. Parasitological study showed hooklets and pseudosegmentation pathognomic for pentastomiasis (Fig. 3). The final diagnosis was consistent with a third stage nymph of Pentastomid, probably belonging to genus *Linguatula*, with typical sclerotized openings (Fig. 4).

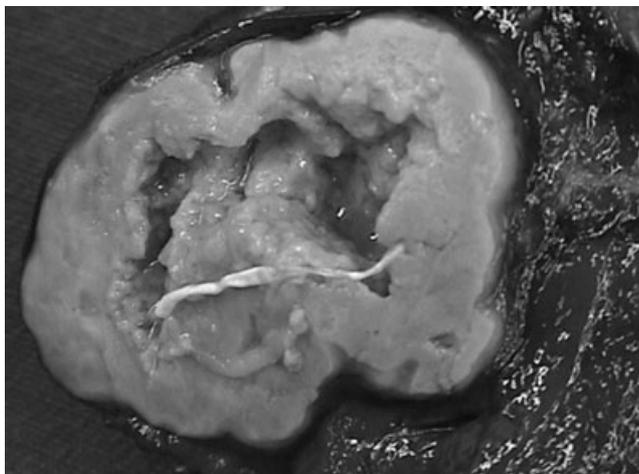
**Discussion**

Human infestation by pentastomid has been reported as early as 1847 by Pruner.<sup>3</sup> Almost all recorded cases among humans have been incidental findings at autopsy. Human pentastomiasis is uncommonly reported, except in the African continent where infestation is fairly common.

Pentastomiasis is an infection caused by a member of the phylum Pentastomida. The name pentastomid was given because of



**Figure 1** Abdominal computed tomography shows a 3 cm tumor in the segment VI of the liver.



**Figure 2** Macroscopic view of the liver specimen. Note the non-caseous granuloma with a still viable Pentastomid.

the two pairs of anterior hooks, and a mouth which gives the appearance of having five openings.<sup>4</sup> The Pentastomida constitute an old and aberrant group of parasites. Within this group there are two families of medical importance to humans: porocephalidae and linguatulidae. The adult porocephalidae inhabit the respiratory tract of some reptiles, and adult linguatulidae the nasal passages of carnivorous mammals. Embryonated eggs, discharged in the feces or in the secretion from the respiratory tract, lodge on vegetation or contaminate water. Many vertebrates, including primates and humans, may serve as intermediate hosts.<sup>5</sup> Self and Kuntz<sup>6</sup> have studied the host-parasite relations and revealed that certain species have the ability to dwell in tissues of their natural definitive or secondary hosts and elicit little or no inflammatory reaction.



**Figure 3** Anterior end of a third stage nymph of Pentastomid, probably belonging to genus *Linguatula*, showing two of the four typical hooks.



**Figure 4** Lateral view of a third-stage nymph, showing typical sclerotized openings.

The fact that this is the first reported case in Brazil certainly does not mean that this is a unique patient. Mato Grosso, the Brazilian state where the patient lives, is located in the western region, well-known for holding the largest swamp (wetland, Pantanal) in the world. The Pantanal of Mato Grosso presents distinct landscape units: permanently, occasionally and periodically flooded areas. It is as big as the French territory and is a perfect ecological niche for a great number of birds, reptiles and mammals.<sup>7</sup> It is a very rustic place with unsatisfactory conditions of sanitation for a significant portion of this population. This environment was probably responsible for the infestation of our patient.

Pentastomes are cosmopolitan, with a concentration in tropical and subtropical areas. Their distribution is determined not only by the range of the natural host but also by the degree of water and food sanitation in these areas.<sup>3</sup> In humans, pentastomid infestation generally results from the consumption of water or crude vegetables contaminated by feces, or by the nasobronchial secretions of

infected snakes or carnivores, more likely by eating raw or inadequate cooked snake or lizard. Once the final diagnosis is consistent with *Linguatula* and knowing the baseline conditions of sanitation, our patient was probably infected by ingesting an infective egg, becoming an intermediate host. Another possibility was ingestion of contaminated water during swimming practice in rivers or lakes from the Mato Grosso State.

Humans are usually highly tolerant to infections with pentastomids. Most cases are asymptomatic and only discovered incidentally, as occurred with our patient. Symptoms can arise when the encysted larvae enlarge through molting and cause pressure on vital structures or when third-stage larvae perforate organs during larval migration. Enlarged larvae have caused collapse of a lung,<sup>8</sup> intestinal obstruction,<sup>9</sup> peritonitis<sup>4</sup> and glaucoma.<sup>5</sup> The liver is the most common affected organ, however, there are no previous reports of pentastomiasis mimicking liver tumor in the English literature. This presentation is quite unusual.

There are three types of pentastomiasis lesions with a human acting as secondary host. In the commonest, necrotic pentastomid granuloma, the parasite has disintegrated into amorphous debris and is often calcified but retains a characteristic C-shaped outline easily seen at radiological exams. In the second type of lesion, the larval pentastomid cyst is seen only when the infection is of recent acquisition. The least common finding is cuticle granulomas which are the remnants of a cuticle shed during molting and left behind by a migrating larva. The present case is a relatively rare condition of an excysted pentastomid causing a substantial lesion in a vital organ mimicking a liver tumor where the third-stage nymph was found inside the liver lesion. The morphology is consistent with a larva that has exited its cyst and migrated to the liver where it apparently had caused an infarct. Identification of the entire larva in the lesion is a very rare situation and has a practical function; offer precise data to ethio-pathological diagnosis.

However, the course and the outcome of this rare condition are still unknown.

The treatment of larval pentastomiasis is unnecessary except when symptoms arise from pressure in adjacent structures or diagnostic doubt. In these situations surgery is indicated for removal. Because this patient was asymptomatic, the indication for liver resection was the impossibility to distinguish this lesion from a primary malignant tumor. As a tropical disease, control of pentastomiasis would be best achieved by more effective water and food sanitation.

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