

## ORAL CYSTICERCOSIS: CASE REPORT AND REVIEW OF THE LITERATURE

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### SUMMARY

Cysticercosis is a condition that occurs when man is infested by the larvae of *Taenia solium*, acting as an intermediate host instead of definitive. Oral cysticercosis is a rare event, and it represents a difficulty in clinical diagnosis. A case of oral cysticercosis in a 23-year-old white female who presented a painless swelling in the dorsal portion of the tongue is reported. An excisional biopsy was performed and histopathological examination revealed a cystic cavity containing the tapeworm.

**KEYWORDS:** Oral cysticercosis; *Taenia solium*; *Cysticercus cellulosae*; Taeniasis.

### INTRODUCTION

Platyhelminthes have a life cycle characterized by two stages; first as a larva, and then as an adult worm, besides an egg phase. Each of these phases requires a different host.

The adult *Taenia solium*, or pork tapeworm, lives in the small intestine of man, its definitive host. Infestation by *Taenia solium* is common in areas where pig breeding is not controlled and sanitation is inadequate. Latin America, Southern Africa, India, Southeast Asia and Eastern Europe are the most frequent locations of occurrence<sup>17</sup>.

The parasitic infestation by the pork tapeworm larval stage, the *Cysticercus cellulosae*, is called cysticercosis, and results from the ingestion of tapeworm eggs through contaminated food and water or dirty hands. Highest incidence countries are Brazil, Chile, Ecuador, Mexico, South Africa, East Africa and India<sup>17</sup>.

The most frequent sites of cysticercosis occurrence are subcutaneous layers, brain, muscles, heart, liver, lungs, and peritoneum<sup>4,33</sup>.

Although the exact incidence is still unknown, oral cysticercosis is considered a rare event and a precise clinical diagnosis is not usually established<sup>16</sup>. We present a case of cysticercosis on the tongue of a Brazilian female, as well as a review of cases reported in the literature.

### CASE REPORT

A 23-year-old white female presented with a swelling on the tongue. During anamnesis, the patient reported that the lesion was painless

and had been present for about three years, showing a slow rate of growth. Past medical history was noncontributory.

On physical examination, the patient had a submucous nodule on the left border of the tongue. The nodule was soft and painful under palpation, measuring about 15 mm, and was lined by normal mucosa (Fig. 1). No palpable lymph nodes were present in the head and neck region.

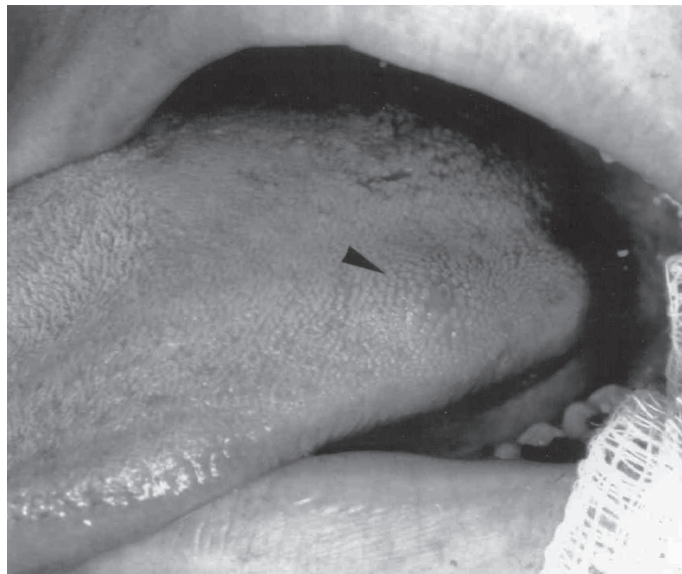


Fig. 1 - Submucous nodule on the left dorsal aspect of the tongue (arrow).

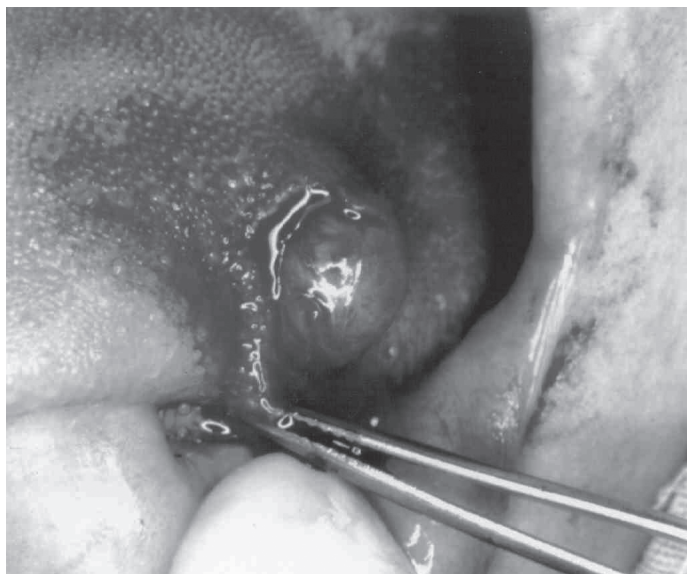
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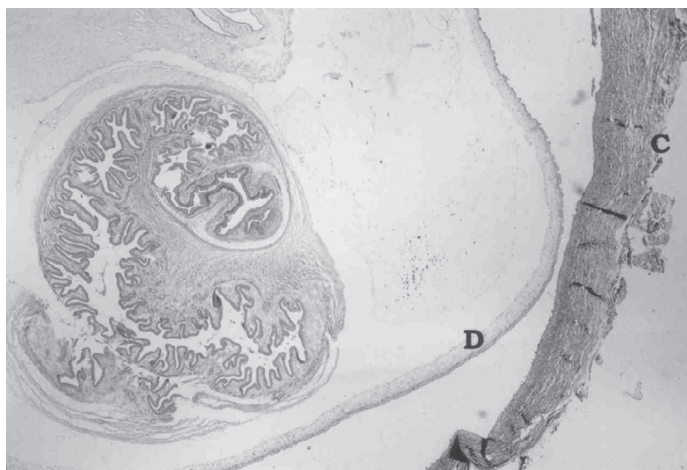
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Under a clinical diagnosis of a benign neoplasm of neural origin, an excisional biopsy was performed under local anesthesia. During the surgical procedure, the lesion could be seen as encapsulated (Fig. 2).

Microscopic examination revealed a capsule of fibrous connective tissue surrounding a cystic cavity, which contained the *Cysticercus cellulosae*, the *Taenia solium* larval form (Fig. 3). The capsule showed intense inflammatory infiltrate, consisting mainly of lymphocytes and plasma cells. Groups of eosinophils were seen throughout the connective tissue. The larva was composed of a scolex, where a sucker could be identified, and a duct-like invaginated segment - the caudal end. Both larva and cystic structure were lined by a homogeneous eosinophilic membrane (Fig. 4 and 5). No areas of dystrophic calcification were present in the specimen. Based on these findings, a diagnosis of cysticercosis was made.



**Fig. 2** - Trans-operative aspect of the cysticerci, revealing an encapsulated lesion on the submucosa.

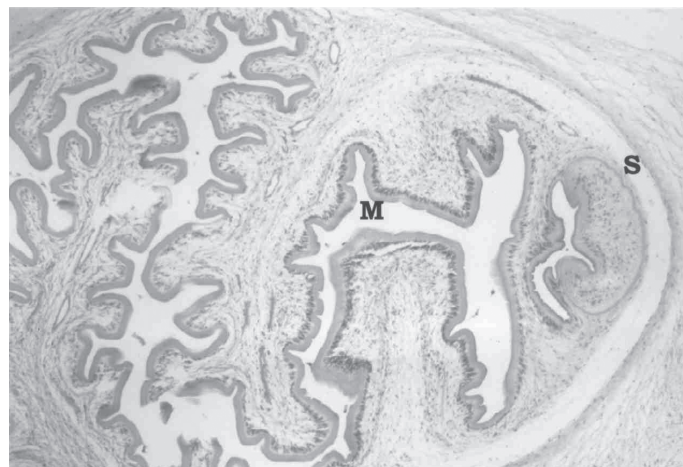


**Fig. 3** - Histopathological aspect of the cysticerci. The larva within the cyst cavity surrounded by the double layered membrane (D) and connective tissue capsule (C). Haematoxylin and eosin x 3.2.

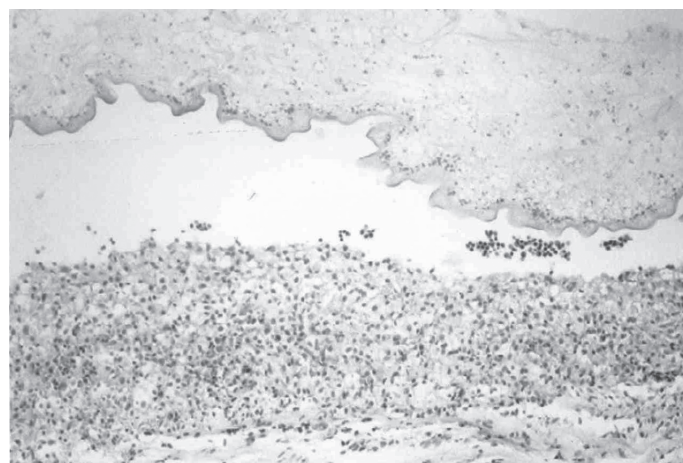
After diagnosis, complete blood count and stool examination were performed, and resulted normal. To eliminate the possibility of neurocysticercosis, a computerized tomography of the head and an electroencephalogram were performed and no other parasitosis focus was found. The patient was scheduled for regular check-ups but was subsequently lost to follow-up.

## REVIEW

The tapeworm is composed of the scolex (head) and proglottids (caudal end). Each proglottid contains from 40,000 to 60,000 eggs, which are released through faeces. The pig becomes infected by ingesting the eggs from the ground contaminated by human faeces and develops the larval form. By consuming inadequately cooked infected pork, man acquires the larva, which becomes, inside the small intestine of the host, an adult worm, the *Taenia* sp<sup>4,26</sup>. Humans can have taeniasis for long periods and contaminate environment continuously<sup>4</sup>.



**Fig. 4** - The duct-like invagination, which composes the caudal end of the larva, lined by the homogeneous membrane (M). Scolex (S) at the cephalic end. Haematoxylin and eosin x 12.5.



**Fig. 5** - Fibrous connective tissue membrane showing intense inflammatory infiltrate, and the double layered membrane. Haematoxylin and eosin x 25.

When man ingests the eggs, he becomes the intermediate host, a role usually played by the pig, and a larval form will develop. The ingestion of *Taenia solium* eggs happens through ingestion of fecally contaminated vegetables, food or water, as well as self-contamination, by reflux of the proglottid from the intestine into the stomach or contaminated hands<sup>22,26,30</sup>. The eggs develop into oncospheres that penetrate intestine wall and - via lymphatic or vascular circulation - reach a destination, where larvae develop and become the cysticerci or "bladder worm", a fluid-filled cyst<sup>22,32</sup>.

Once a person becomes the host of *Cysticercus cellulosae*, cysticercosis can develop in various organs and tissues<sup>22</sup>. The most serious involvement is that of the central nervous system, followed by ocular involvement, usually the only ones which are symptomatic<sup>4,8</sup>. Generalized symptoms include headache, fever and myalgia. Patients with neurocysticercosis can present with several signs and symptoms, being the most frequent seizures, increased intracranial pressure, obstructive hydrocephalus, meningitis, and mental disorders<sup>11,32</sup>.

In spite of the abundance of muscular tissue in the oral and maxillofacial region, this is not a frequent site of occurrence for cysticercosis. We have found sixty-five cases reported in the literature<sup>1-3,6-16,18-20,22-24,26-32</sup>. The age was reported in sixty-four cases and varied from three to seventy years, with a mean age of 23.7 years. The most frequently affected decade was the third (32%), followed by the fourth decade (20.6%). There was an equal distribution between genders. The most frequently involved site were the tongue (42.15%), followed by the lips (26.15%) - with the lower lip louching 64.7% of those - and the buccal mucosa (18.9%). Several reports<sup>1,24,27,28,30,32</sup> revealed multiple foci in the same patient.

Usually, the patient complaint is a swelling. Although pain is not a frequent feature, it had been reported in secondarily infected cases<sup>13</sup>. In the case here reported the patient presented pain under palpation but not spontaneous pain. It was suggested that the lesion on the tongue could interfere with movement, causing discomfort during speaking and eating<sup>27</sup>.

Differential diagnosis of oral lesion depends on the site involved. In this case, in which the aspect was of a solitary nodule on the tongue, lined by normal mucosa, the differential diagnosis would be benign neoplasm of neural tissue origin, as neurofibroma and benign schwannoma, granular cell myoblastoma, vascular neoplasm, vascular leiomyoma, fibroma, and lipoma.

Histopathological examination makes up a diagnosis of cysticercosis by the detection of a cystic space containing the *Cysticercus cellulosae*. The scolex has four suckers and a double crown of rostellar hooklets<sup>13</sup>. A duct-like invaginated segment, lined by a homogeneous anhistic membrane, composes the caudal end. The eosinophilic membrane that lines the capsule is double-layered, consisting of an outer acellular and an inner sparsely cellular layer<sup>15</sup>. After a period within three and five years the larva dies and the cyst undergoes calcification<sup>10</sup>.

Currently other diagnostic tools as radiologic imaging and serology can be used. Besides normal radiographic examination other modalities of imaging exams are very effective in the detection of cysticerci, as

computerized tomography and magnetic resonance<sup>21</sup>. SARAN *et al.*<sup>26</sup> proposed the use of fine-needle aspiration cytology, which identifies the tegument layer of the larva, to help the clinician in planning the treatment.

Immunodetection of cysticercosis can be achieved in sera, cerebrospinal fluid and saliva, by ELISA (enzyme-linked immunosorbent) assay or EITB (enzyme-linked immunoelectrotransfer blot)<sup>4</sup>, but it is important to consider that individuals living in an endemic area may have antibodies because of an exposure instead of an established infection<sup>5</sup>. SANCHEZ *et al.*<sup>25</sup>, performing EITB in patients from endemic areas with brain lesion compatible with neurocysticercosis, found a similar proportion of seropositive and seronegative groups.

Every case of oral cysticercosis should be thoroughly investigated to determine the involvement of multiple foci, since there is a high incidence of such feature<sup>11</sup>.

Drugs as praziquantel and albendazole are potent antihelminthics used in the treatment of cysticercosis<sup>22</sup>, replacing niclosamide, which was the drug of choice for the treatment of the disease for a long period<sup>7,13</sup>. Drugs should be used especially in cases where surgical treatment is risky or not possible, as in neurocysticercosis. As the patient in the present case had no other occurrence of cysticercosis or any other symptoms, no additional treatment was prescribed except the periodic follow-ups.

It is important to consider the diagnosis of cysticercosis in oral solitary nodular lesions presenting in patients living in an endemic area. This case emphasizes the role of the dentist in the detection of a disease that can have more serious involvement, as well as the importance of routine histological examination.

## RESUMO

### Cisticercose oral: relato de caso e revisão da literatura

A cisticercose é uma doença que ocorre quando o indivíduo é infectado pela larva da *Taenia solium*, atuando como hospedeiro intermediário ao invés de definitivo. A cisticercose em cavidade oral é rara e seu diagnóstico clínico é difícil. Neste trabalho, é relatado um caso de cisticercose oral em paciente de 23 anos, sexo feminino que apresentou um crescimento indolor na região de dorso de língua. Foi realizada uma biópsia excisional e o exame histopatológico revelou uma cavidade cística apresentando em seu interior a larva.

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