

Encysted *Tenia solium* larva of oral cavity: Case report with review of literature

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Abstract

Cysticercosis is caused by the larvae of the pig tapeworm, *Tenia solium*. Oral cysticercosis is a rare event and is often a diagnostic challenge to the clinician. We report a 12-year-old girl who presented with a single, painless, nodule on the lower lip that was diagnosed as cysticercosis. Current literature on the clinical presentations, investigations, and treatment of the condition has been reviewed in this article. We have also proposed a set of criteria for the diagnosis of oral cysticercosis.

Keywords: Cysticercosis cellulosae, cyclo-zoonosis, oral cysticercosis, *Tenia solium*

Introduction

Kuchenmaister in 1855 established that human cysticercosis is caused by the larval stage (cysticercosis cellulosae) of the pork tapeworm, *Tenia solium* (*T. solium*).^[1] The etiopathogenesis is related to the ingestion of food, vegetables or water contaminated with *T. solium* eggs, inadequately cooked pork containing cysticerci or very rarely reinfection by the transport of eggs from the bowel into the stomach by retroperistalsis in a person with intestinal teniasis.^[2] It is a “biological marker” of the social and economic development of a community, being endemic in countries of Latin America, Asia, and Africa.^[3] Brain, eyes, muscle, and heart are the most commonly affected sites.^[4] Oral cysticercosis, a rare entity, is a diagnostic challenge and is poorly documented in the English literature.^[5] We report a case of cysticercosis involving the lip in a child and also review the existing literature on the condition.

Case Report

A 12-year-old girl visited our OPD with a nodular swelling on the lower lip. She first noticed it 2 months ago. The swelling was painless, remained unchanged in size, and caused a little difficulty while eating and talking. Her family history and

previous medical and dental history were noncontributory. She belonged to the lower economic strata and had a vegetarian diet. On examination there was a solitary, spherical, well-defined swelling in the lower labial mucosa present on the left side of the midline which measured approximately 1.5 × 1.5 cm [Figure 1]. The mucosa over the swelling was normal. On palpation it was not tender, but was tense and nonfluctuant. Differential diagnoses of mucocele, lipoma, and fibroma were considered. A fine-needle aspiration (FNA) was performed. The aspirate was a clear fluid but the report was not conclusive. Hence the lesion was excised under local anesthesia which was a smooth well-encapsulated mass [Figure 2]. Hematoxylline and eosin (H/E) stained sections showed a cystic mass containing the cysticercus cellulosae surrounded by dense fibrous capsule infiltrated with inflammatory cells, mainly lymphocytes and plasma cells. The inner aspect of the capsule was a double-layered membrane in which larval form of *T. solium* were seen. Larva showed the presence of suckers and caudal to it duct-like invagination segment lined by homogenous membrane. No areas of dystrophic calcifications were present in the tissue specimen [Figures 3a and b]. The final diagnosis of cysticercosis of the lip was made. Considering the financial constraints of the patient a computed tomogram (CT)/magnetic resonance imaging (MRI) were not advised. She was referred to the pediatrician who prescribed tab albendazole 400 mg bid for 15 days and recalled. The patient however was lost to follow-up.

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Review of Literature

Life cycle of *T. solium*

A hermaphroditic cestode belonging to the phylum platyhelminthes, has a complex two host life cycles. Human beings are the only definitive host and harbor the adult tapeworm in the small intestine (teniasis) on ingesting raw or inadequately cooked pork infected with their cysticerci. The larvae evaginate from the cyst in the small intestine. The head (scolex) attaches to the mucosa through its four suckers and thereafter begin to form segments (proglottids). Two months after infection the gravid proglottids detach from the distal end

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Figure 1: Intraoral photograph of the lesion



Figure 2: A smooth well-encapsulated mass removed after excision

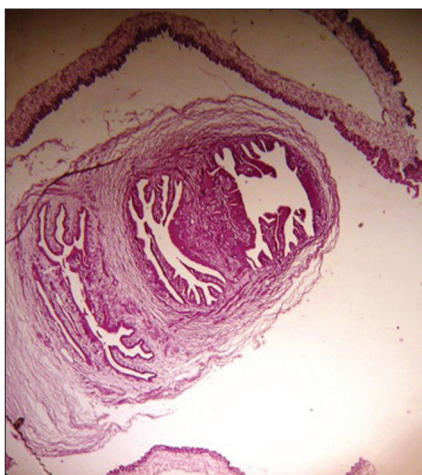


Figure 3: Photomicrograph of the tissue section showing the dense fibrous capsule infiltrated with inflammatory cells. Inner aspect of the capsule shows a delicate double-layered membrane in which larval forms of *Taenia solium* can be seen. (Low-power view: H&E $\times 40$)

and are excreted in the feces. When these eggs are ingested by humans and pigs (the intermediate host), the embryos (oncospheres) are released due to the action of gastric acid and intestinal fluids and penetrate the bowel mucosa. Subsequently they enter the blood stream to reach various tissues to develop into larval vesicles or cysticerci thereby producing human and porcine cysticercosis, respectively. In humans the cysticerci can remain viable for as long as 10 years evading immune recognition during which time it evolves through three stages: stage of involution, a granular stage, and the final stage of calcification following a granulomatous reaction. It is the dead calcified larva that produces the various symptoms of the disease depending on the anatomic location of its encystment.^[1,4,6]

Spectrum of clinical presentations

Clinical manifestations of cysticercosis are varied and depend on (i) the location, growth, size, and the number of cysticerci, (ii) stage of cyst degeneration and presence of calcification,

and (iii) the extent of host inflammatory response or scarring.^[6] Neurocysticercosis (NCC) is the most common cause for epilepsy in developing countries and is associated with substantial morbidity, manifesting as headache, seizures, hydrocephalus, meningitis, focal neurological deficit, psychological disorders, dementia or spinal cysts.^[3] Persistent antigenic stimulation seen in NCC may increase the frequency of aberrations in the chromosomes 7 and 14 which may play an important role in the oncogenesis in such patients.^[4] Ophthalmic cysticercosis may present as proptosis, diplopia, and loss of vision.^[1] Muscular cysticercosis is a casual finding and has been reported in the orofacial region as a painless swelling involving masseter and the mylohyoid.^[6,7] Three distinct types of clinical presentations are described in the muscular form. The myalgic type where there is severe local pain in the muscle due to the acute inflammation resulting from the death of the larva and leakage of fluid from the cyst. The second is the mass-like, pseudotumour or abscess-like type in which there is intermittent leakage of cystic fluid that elicits a chronic inflammatory response with collection of the fluid around the cyst which presents as a mass. Rarely, the third, pseudohypertrophic, type occurs when the cyst retracts, its capsule thickens and the scolex calcifies.^[6] Only two cases, both from India, of extracranial cysticercosis of the parotid gland have been reported in the literature which presented with features mimicking acute parotitis.^[8,9]

Oral cysticercosis is a rare event. A thorough search in the PUBMED indexed English literature reported 69 cases till date [Table 1]. Asymptomatic cystic swelling or nodule is the only evidence of the disease. Any region of the oral cavity can be involved but a review of published cases suggests that tongue is a site of predilection in the oral cavity with 32 of the 69 cases (46.37%) [Table 1] reported, followed by labial and buccal mucosa. Some authors, however, postulate that the muscular activity and high metabolic rate may prevent lodging of the cysticerci in the tongue.^[4] Only one case of cysticercosis involving the gingival tissue has been documented

till date which is also from India.^[2] The cystic nodules may persist or rupture and heal uneventfully.^[15] Oral cysticercosis is a diagnostic challenge to a clinician as the condition mimics other oral nodular lesions like mucoceles, fibroma, granular cell tumor, benign salivary gland tumor. The high intraluminal pressure in oral cysticercosis helps to differentiate it from lipoma and hemangioma.

Investigations

Diagnosis of human cysticercosis is impaired by its polymorphic clinical presentations. Excisional biopsy, sections stained with H&E, is the only confirmatory diagnostic procedure for the condition which demonstrates the presence of the larva in the tissue or the scolex in the cystic lesion. However, other investigations must be considered to detect the disease in the diverse tissues that may be affected, including the oral cavity.

FNA

It reveals the larval fragments which can be identified by its lightly stained outer wavy membrane and multiple tiny ovoid nuclei in the fibrillary stroma. This has been demonstrated in 45-100% of the aspirates, especially when the aspirated material shows a speck of pearly white content which is actually the larva in an acute or chronic inflammatory background.^[4] Routine laboratory findings show marked eosinophilia and raised immunoglobulin E (Ig E). Parasitological examination of *T. solium* eggs in the stool samples can be advised for the patient as well as the family members and domestic workers.^[15]

Serology

A wide range of serological assays also aid in the diagnosis and epidemiological studies of cysticercosis. Immunodiagnosis can be achieved in the serum, CSF, and saliva either by enzyme linked immune sorbent assay (ELISA) or enzyme linked immune electrotransfer blot (EITB).^[17] However, use of unfractionated antigens is associated with high rates of false-positive and false-negative results.^[3] It is also important to note that calcified nodules are not antigenic and hence cannot be diagnosed.^[4] Recently a new diagnostic tool, *T. solium* cyst fluid antigen-based lymphocyte transformation test (LTT), has shown specificity and sensitivity greater than ELISA and EITB.^[3]

Imaging

Conventional radiographs are useful for detection of calcifications in the muscles.^[15] Computed tomography (CT) and magnetic resonance imaging (MRI) have greatly improved the accuracy of diagnosis of cysticercosis, NCC in particular. The vesicular stage of the cyst is seen on the CT as a hypodense area containing a hyperintense small scolex along with a nonenhancing or mildly enhancing cyst wall. CT depicts the colloidal vesicular stage as a ring enhancing cystic lesion with hyperintense fluid content and surrounding edema. In the granular nodular stage when the cyst retracts to form a nodule, the CT shows an enhancing nodule with mild surrounding edema. The final stage can be seen as single or multiple calcified nodules. MRI is considered best for the detection of

degenerating and innocuous (viable) cysticerci. It also helps in visualizing the perilesional edema very well. Hence, although MRI allows better detection of the active parasites, it can miss the calcified lesions, especially when the gradient echo sequence is not used; in which case a CT must be advised.^[3] The advent of high-resolution ultrasound has greatly augmented the role of sonology in diagnosing muscular cysticercosis. It gives four different appearances that are pathognomonic and aid in diagnosing with great confidence. It may appear as a cysticercus cyst with an inflammatory mass around it, as an irregular cyst with very minimal fluid on one side, due to the leakage of fluid or as a large irregular collection of exudative fluid within the muscle, with the cyst containing the scolex situated eccentrically within the collection. The fourth appearance on ultrasound is that of calcified cysticercosis, wherein multiple elliptical calcifications in soft tissue similar to the millet seed-shaped calcifications seen in plain radiography.^[6]

No objective diagnostic criteria was found in the English literature for diagnosing oral cysticercosis unlike the one established for NCC.^[1] We propose a set of criteria which can help the clinician to diagnosis a case of oral cysticercosis with reasonable certainty [Table 2].

Treatment

Cysticercosis outside the nervous system is a benign condition and usually does not warrant-specific treatment. NCC,

Table 1: Details of oral cysticercosis cases reported in the literature

Site involved	No. of cases reported internationally	No. of cases reported in India	Total no. of cases reported
Labial mucosa	11 [‡] [4,10-12]	07 [‡] [2,5,13,14]	18
Buccal mucosa	06 [‡] [4,15,16]	08 ^[2,5,14]	14
Tongue	18 [‡] [4,15,17-22]	14 [‡] [2,13,14,23-29]	32
Parotid gland	None	02 ^[8,9]	02
Masticatory muscles	None	02 ^[6,7]	02
Gingiva	None	01 ^[2]	01
Total no. of cases			69

[‡][4,13,15] are cases where patient reported with multiple intraoral swellings

Table 2: Proposed criteria for diagnosis of oral cysticercosis

A compatible clinical presentation
FNA aspirated material showing a speck of pearly white content
Histopathologic demonstration of the parasite on biopsy
Cystic lesions with scolex on imaging (muscular cysticercosis)
Positive epidemiological factors like household contact, endemic region or travel to or from an endemic area

however, due to its morbidity and mortality requires effective intervention.^[1] Existing treatment regimens are largely based only on nonrandomized trials and investigators' observation. Treatment includes medical management with larvicidal drugs, corticosteroids to decrease or prevent inflammation, surgical procedures for the removal of the cyst, and supportive measures. Until the late 1970s the only treatment available for NCC was surgery for cyst excision or steroids to reduce inflammation. Antihelminthic praziquantel, priorly used in porcine cysticercosis, was the first drug tried in human cysticercosis. Later albendazole became a cheaper and more effective alternative. The currently accepted regimens are either 8 days of albendazole (15 mg/kg/day with a maximum of 400 mg bid) with simultaneous administration of steroids or 15 days of praziquantel (50 mg/kg/day in three divided doses). Both the drugs have documented drug interactions with antiepileptics like dilantin and carbamazine. Oxfendazole, a veterinary benzimidazole, is very effective (95%) in killing the porcine cysts when administered as a single dose of 30 mg/kg. However this drug is yet to be tested in human beings. *T. solium* oncospherical antigen vaccine has been tried with partial protection of the pigs.^[1]

Corticosteroids, dexamethasone or prednisone is used to suppress and/or prevent edema and intracranial hypertension that usually occurs 2--5 days after the start of antiparasitic therapy. It is administered as 10--16 mg/day in divided doses and tapered over 1--3 months depending on the MRI findings. Seizures in these patients respond well to first-line antiepileptic drugs, but usually complete withdrawal of these drugs may not be possible.^[1]

Surgical modality of treatment in cysticercosis has dramatically reduced over the past two decades due to the favorable outcomes of antiparasitic drugs. Need for surgery arises only to approach cysts that impinge on critical structures in order to prevent serious complications and to decrease the duration of corticosteroid use.^[1]

Oral cysticerci, being usually localized and superficial lesions, are easy to excise and have good prognosis. Simple surgical excision is often all that is required to ensure complete removal of the lesion without any postoperative complications in such cases.^[4] Lesions involving the muscles like the masseter may be treated conservatively with antiparasitic therapy.^[6] Unlike the NCC or orbital cysticercosis which are severe in their clinical presentations, the oral cysticercosis are usually well tolerated. However, it is important to exclude the presence of the parasite in other sites through a detailed case study and systematic investigations.

Conclusion

Cysticercosis is a global public health problem. Its prevalence is high in less developed and developing countries. Systematic population-based studies are lacking in India to

estimate the disease burden of cysticercosis in general and oral cysticercosis in particular. General improvement in the sanitary conditions, adequate treatment of human sewage or feces, and mass education about personal hygiene (hand washing) can help in the prevention and possible eradication of human cysticercosis. Abattoirs should block the sale and consumption of infected pork through regular inspections of slaughterhouses. Oral cysticercosis should be considered in the differential diagnosis of intraoral solitary swellings especially in endemic areas. In endemic countries a lesion can be first referred for a FNA which is cost-effective, quick, and reliable. Expensive investigations, like immunological tests and CT, can be undertaken only in the absence of a definitive pathological diagnosis.

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