



## **Lingual Cysticercosis - A Case Report**

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### **Abstract**

Tapeworm infection in humans falls into two categories: Taeniasis and Cysticercosis. Taeniasis is an intestinal infection caused by the adult tapeworm. Larval form of *Taenia solium* causes cysticercosis due to ingestion of the eggs through faeco-oral route. The disease is common in underdeveloped countries. The prevalence of the disease in India, which is a developing country is high as large population are living below poverty line. Even though cysticercosis is reported commonly in sites like central nervous system, muscles, heart, liver, lungs, eyes, and subcutaneous tissue etc; it is rare to find this in tongue and may present as a cystic swelling. Clinically, it is difficult to diagnose as it mimics common lesions like mucocele. We present one such rare case in a 24 years old male who presented with a cystic lesion over the ventral aspect of tongue and clinically suspected to be lipoma and on histopathological examination, proved to be cysticercosis of tongue.

**Keywords:** Cysticercosis, Tongue Nodule

### **Introduction**

The word *Cysticercus* is derived from Greek words *Kystis* = cyst and *Kertos* = tail because of their

appearance. [1] In spite of abundant intermuscular tissue in oral tissues, it is an uncommon site for *cysticercus* due to high muscular activity and metabolic rate of oral tissues in humans, which act against lodgement and development of *cysticerci*. Frequently involved oral locations for cysticercosis are tongue, labial or buccal mucosa and mouth floor. [2] Histopathological findings are confirmatory and necessary before starting the treatment.

Prevalence of oral cysticercosis was 3.5% (of 769 cases) as per one study and tongue is the most commonly affected sites amongst all sites in oral cysticercosis. [3] Cysticercosis presenting as a solitary mass in the tongue or a tongue nodule is extremely rare. There are only 34 cases reported in world literature. [4,5] Cysticercosis is eradicated from most of the developed countries, but is still prevalent in Central and South America, South Asia, India, and China. [6] As per the available literature, the prevalence of oral cysticercosis is 4.1%. [7] When the implanted *Cysticercus* dies, the residual lesion undergoes calcification and then it becomes evident. Generalized symptoms include headache, fever, and myalgia. Patients with neuro cysticercosis can present with several signs

and symptoms, the most frequent being convulsions, increased intracranial pressure, obstructive hydrocephalus, meningitis. [8, 9] The most commonly involved intraoral sites are buccal mucosa, tongue, and lips. [10] Most of the cases of cysticercosis of tongue present as painless, well circumscribed soft swelling that may resemble lesions like mucocele; which is a common oral mucosal lesion originating from minor salivary glands. [11, 12]

### Case report

A 24 years old male came to the oral-maxillofacial surgery out-patient department with painless, slowly growing swelling over the ventral aspect of tongue. He was advised ultrasound investigation. The ultrasound study revealed a cystic lesion over the tongue. The differentials on clinical examination were lipoma or infective lesion. The cyst was removed surgically and sent for histopathological evaluation.



Figure 1: Image showing cystic lesion over the ventral aspect of the tongue.

[Courtesy: OMFS Department, D.Y. Patil Hospital, Nerul, Navi Mumbai]

Gross examination revealed a cystic mass measuring 1 cm in diameter. Cyst wall measuring 0.1 cm in thickness. Contents were watery. Microscopy showed a cyst wall and a parasite with irregularly shaped membranous foldings of the spiral canal. Parasite was single and

invaginated. Features were suggestive of classical cysticercosis.



Figure 2: Microscopic image of the parasite showing membranous foldings on H & E staining (40X)

### Discussion

*Taenia solium* has two host life cycle. It is a cestode that inhabits the human small intestine of those individuals who have ingested raw or inadequately cooked pork infected with their viable larvae (cysticerci). The scolex of the larva evaginates from the cyst inside the small intestine and attaches to the bowel wall. After 3 months, the adult tapeworm develops within its human definitive host, producing a condition known as taeniasis and thereafter begin forming proglottids, which are frequently detached from the worm and are excreted in the faeces. [13] Each proglottid contains 50 000 to 60 000 fertile eggs, which can remain viable for a long time in water, soil, and vegetation. Cysticercosis develops when these eggs are ingested by humans and pigs (intermediate hosts), and oncospheres are liberated by the action of gastric acid and intestinal fluids and cross the bowel wall, enter the bloodstream and lodge in various other tissues and organs where they develop into cysticerci or 'bladder worm' cysts; which are fluid filled cysts. [14-17] In humans, this disease mainly occurs due to ingestion of contaminated food or polluted drinking water, but it also may develop by faecal-oral contamination. [18] The implication of knowing the life cycle is that cysticercosis, as opposed to taeniasis, develops by the ingestion of

tapeworm 'eggs' releasing embryos (oncospheres) in the intestine. This ingestion of the eggs can occur with consumption of contaminated food or water.

Oral cysticercosis is a rare and an asymptomatic condition. The most common intraoral sites reported include tongue (42.15%), lips (26.15%) and buccal mucosa (18.9%). [19] However, increased immigration and travel-related intestinal tapeworm infestation have also resulted in spread of this disease to non-endemic areas as well. [20] This condition is predominant in the lower socioeconomic communities particularly in countries of Latin America, Asia and Africa. [6]

Differential diagnosis of lingual cysticercosis includes mucocoele, lipoma, neurofibroma. It is important to consider the diagnosis of cysticercosis in oral solitary cystic nodular lesion presenting in patients living in endemic areas.

Grossly these cysts are variably sized ranging from few millimetres to few centimetres and contain clear fluid. The cyst wall is pearly white; scolex may be visible inside the cyst as tiny white nodule. Lingual cysticercosis is rare in humans. Definitive diagnosis can only be made by demonstration of *Cysticercus cellulosae* on histopathology.

Laboratory investigations suggested include:

- 1) IgG antibodies against cysticercus [21]
- 2) IgG antibodies in CSF [21]
- 3) Experimental studies have documented the presence of several cysticercal antigens like antigen B which is a paramyosin [21-23]
- 4) Enzyme Linked Immunosorbent Assay [2]

Treatment includes surgical excision of the cystic lesion along with administration of anti-helminthics such as praziquantel and albendazole. [20] It is said that the use of systemic anti-helminthics is advised in symptomatic or disseminated cases such as those with neurocysticercosis.

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). [24]

Excision of the lesion followed by histopathological examination is the only confirmatory diagnostic modality. Other preoperative diagnostic modalities include ultrasonography, conventional radiographs (that show presence of calcifications within muscles), computed tomography and magnetic resonance imaging.

### Conclusion

It is important to consider the diagnosis of cysticercosis in oral solitary cystic nodular lesion presenting in patients living in endemic areas. Definitive diagnosis can only be made by demonstration of *Cysticercus cellulosae* on histopathology. The present case reveals the importance of the histopathologic examination and the need to include cysticercosis as a differential diagnosis in oral nodular lesions. The treatment of oral *Cysticercus cellulosae* is surgical excision. Drugs should be used in symptomatic patients and disseminated cysticercosis. [25]

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