

Cysticercosis presenting as Neck Swelling: A Rare Case diagnosed on Ultrasound Report

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ABSTRACT

Introduction: Cysticercosis is a condition that occurs when humans are infested by the larvae of *Taenia solium*, acting as an intermediate host instead of definitive occurrence in neural and extra-neural forms. The latter commonly involves subcutaneous tissue, skeletal muscles, and eyes.

Aims and objectives: Oral cysticercosis is a rare event, and it represents great difficulty for clinical diagnosis. In present report we report a case of cysticercosis in the right submandibular region where it presented as a large, soft cystic swelling and tried to explain the clinical sign & symptom of oral cysticercosis. We emphasize on the importance of ultrasonographic and routine microscopic examinations for the diagnosis of even apparently innocuous lesions in submandibular regions.

Results: This is a very rare case of oral cysticercosis showing neck swelling extended from the lower border of the mandible to the thyroid cartilage. The diagnosis was made by sonography and confirmed by gross and microscopic examination of cysticercosis cellulosae. In ultrasonography, there was well-defined cystic lesion with hyperechoic eccentric tiny nodules.

Keywords: Cysticercosis cellulosae, Oral cysticercosis, *Taenia solium*. Taeniasis.

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INTRODUCTION

The life cycle of the flatworm is characterized by two stages: One is larva, and the other is the adult worm, apart from egg phase. Every phase requires a different host. The definitive host of adult *Taenia solium*, or pork

tapeworm is the small intestine of man. If the sanitation is not proper and pig breeding is not controlled, infestation by T. solium is common in those areas. The areas where the most frequent occurrences of T. solium are present are in Southeast Asia, Southern Africa, Latin America, India, and Eastern Europe. 1,2 Cysticercosis cellulosae is a parasitic infestation by the pork tapeworm in its larval stage. Cysticercus results from the ingestion of tapeworm eggs through contaminated water and food or unclean hands. The highest incidences in the world are in countries, such as Brazil, Chile, Ecuador, Mexico, South Africa, East Africa, and India. The most frequent sites of cysticercosis occurrences are subcutaneous layers, brain, muscles, heart, liver, lungs, and peritoneum.^{3,4} Although the exact incidence is still unknown, oral cysticercosis is considered a rare event and a precise clinical diagnosis is not usually established. The disease is often confused with other benign swellings.6

We present a case of cysticercosis in the submandibular region of an Indian female, as well as a review of cases reported in the literature. We emphasize on the importance of ultrasonographic and routine microscopic examinations for the diagnosis of even apparently innocuous lesions in submandibular regions.

CASE REPORT

A 7-year-old girl presented with a chief complaint of swelling localized at the right submandibular region (Fig. 1). This was so since 1 year and she had no history

Fig. 1: Clinical extraoral photograph of the swelling in the submandibular region on the right side

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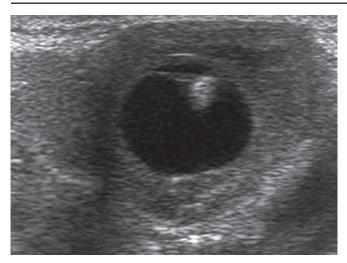


Fig. 2: Ultrasonographic image shows well-defined cystic lesion with eccentric hyperechoic tiny nodules

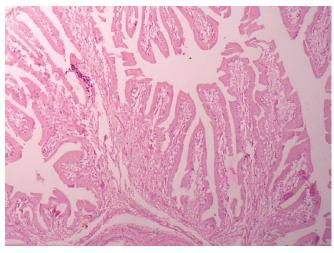


Fig. 3: Photomicrograph (10×) shows larva-cysticercosis cellulosae

of pain in the swelling. There were no history of difficulty in eating or speaking, fever or facial paralysis. Patient noticed that initially swelling was small in size and gradually increased to the present size of 5×4 cm.

Clinical examination revealed that the oval-shaped swelling was present and that measured 5×4 cm in diameter. The swelling extended from the lower border of the mandible to the thyroid cartilage. Border of the swelling was regular, well-defined, smooth surfaced, and color was normal as that of the adjacent side. Tenderness was negative on palpation and temperature was afebrile to touch. Consistency of swelling was soft to palpation and fluctuation was absent. There was no fixity of swelling to overlying skin and diagnosis was as benign nodule. There was no discharge or redness. Physically, the patient's status was good and she had no remarkable medical history. The differential diagnosis included fibroma, lipoma, and pleomorphic adenoma.

After that, the patient was advised an investigatory workup including complete hemogram and ultrasonography and fine needle aspiration cytology or biopsy. Routine hematological investigations were within normal limits. In ultrasonography, there was well-defined cystic lesion with hyperechoic eccentric tiny nodules (Fig. 2).

The lesion was enucleated under local anesthesia and submitted to histopathological examination. In histopathological examination, there was a thin-to-thick fibrous capsule (Fig. 3).

In the capsule, there was a cyst containing the larval stage of *T. solium*. Computed tomography images showed no abnormal density lesion. The final diagnosis of cysticercosis was established based on typical invaginated segments with papillary projections of the larval body.

Clinical information about the case—as no signs or symptoms of disseminated cysticercosis were detected, additional therapy was not performed. Patient was recalled and follow-up was done without signs of cysticercosis for 2 years.

DISCUSSION

Taenia solium is endemic to many parts of the world including Latin America, India, Eastern Europe, Asia, and sub-Saharan Africa. The life cycle of the tapeworm is characterized by different stages of development, which require several species of hosts to appropriately harbor the eggs, oncospheres, larvae, and adult worms. Cystic larvae develop by penetrating the human intestinal wall and may disseminate through vascular or lymphatic circulation. The cycle is ended by the development of an adult worm in the small intestine of the host.⁶⁻⁸

Most severe manifestation of cysticercosis is neurocysticercosis that involves the central nervous system characterized by symptoms, such as headache, fever and myalgia or more severe signs and symptoms, such as convulsions, increase in intracranial pressure, meningitis, and mental disorders.^{1,9} The World Health Organization estimates that more than 50,000 deaths per year are caused by neurocysticercosis worldwide. 8,10 In the present case, the patient denied any symptoms, and a lack of systemic involvement was confirmed by laboratory tests and diagnostic imaging carried out by the medical team. The patient had a single asymptomatic nodule in the submandibular region. Oral cysticercosis was considered to be unusual by Elias et al. Only 65 cases are reported in literature. Saran et al examined 120 cases and noticed 4.2% in the mouth. Both researchers found a greater incidence in the tongue, followed by labial mucosa and buccal mucosa.1

The factors that are responsible for the clinical appearance of cysticercosis are number, location, and extent of associated inflammatory response or scarring. Neurocysticercosis is characterized by seizures, hydrocephalus,

and raised intracranial tension.¹¹ Three distinct forms of muscular types are the myalgic type, mass-like, abscess or pseudotumor type, and the rare pseudohypertrophic type. During the death of the larva, there is leakage of fluid from the cyst. The resulting acute inflammation may result in local pain and myalgia. Alternatively, degeneration of the cyst may result in intermittent leakage of fluid, eliciting a chronic inflammatory response, with collection of fluid around the cyst, resulting in the mass-like type, the pseudotumor type or the abscess-like type, as was seen in our case. Alternatively, the cyst retracts, its capsule thickens, and the scolex calcifies.¹²

The present case had submandibular region involvement; there is no gender preference for oral cysticercosis. The clinical appearance connotes the presence of lipoma, pleomrphic adenoma. The location of the lesion decides the differential diagnosis of cysticercosis. Nodules on the lips and cheeks may be considered as fibroma, lipoma, mucocele, pyogenic granuloma, or pleomorphic adenoma. If there are nodules on the tongue, then differential diagnosis will be considered as pyogenic granuloma, fibroma, rhabdomyoma, or granular cell myoblastoma. The final diagnosis of cysticercosis is dependent on histologic examination, but before that radiologic imaging and laboratory tests can be used as initial imaging tools. The submandance of the presence of lipoma, and laboratory tests can be used as initial imaging tools.

Poor personal and household hygiene, frequent consumption of pork, and history of passing tapeworm proglotids in feces are risk factors for human cysticercosis. Once a person becomes the intermediate host, cysticercosis can develop in various organs and tissues. The types of tissue affected by cysticercosis are subcutaneous layers, brain, muscles, heart, liver, lungs, and peritoneum in this order. The treatment of oral cysticercus is surgical excision and the biopsy specimen will allow confirmation of the tentative diagnosis.¹⁴ Surgery is the treatment for localized lesions, which have obvious symptoms. Medical treatment with praziquantel or albendazole has been recommended for neurocysticercosis and subcutaneous cysticercosis. 16 Preventive measures are important and include the thorough cooking of pork and all vegetables and early detection and complete removal of the worm, including the head.

Noninvasive and nonionizing, high-resolution ultrasound is important to establish the diagnosis in patients

with muscular cysticercosis. If lesions with the morphological characteristics described earlier are encountered on ultrasound, the diagnosis of cysticercosis can be made with great confidence, and in muscular and subcutaneous cysticercosis, no further investigation is required.

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