

Cysticercosis of the sunlingual region – a case report



MEDICAL SCIENCE

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ABSTRACT

Cysticercosis is a parasitic disease caused by Taenia solium. It is a major public health problem in developing countries. Infection is acquired through ingestion of raw or undercooked meat containing the cysticercus. Man is the intermediate host and pig is definitive host. Cases usually present with vague abdominal discomfort, indigestion and diarrhoea. Cysticerci can be found anywhere in the body, but are most commonly detected in brain, eye, skeletal muscle and subcutaneous tissue. Diagnosis is made by the demonstration of eggs or proglottids in faeces and definitive diagnosis is by biopsy of the lesion. We report an unusual case, which presented with a sublingual mass. Diagnosis of cysticercosis was confirmed by histopathology.

Introduction

Cysticercosis is the larval stage of the tape worm *Taenia solium* found in tissues whereas the adult worm form is found in the intestines [1]. The eggs, after ingestion of contaminated food, release larvae in the intestine, which penetrate the intestinal wall and migrate to various sites with an affinity for neck, tongue and trunk, subcutaneous tissues and brain [1]. Oral cysticercosis is a rare event, and it represents difficulty in clinical diagnosis [2]. The most frequently involved oral locations are the tongue, labial or buccal mucosa [3,4]. A case of sublingual cysticercosis is being reported.

Cysticercosis is caused by the larval stage of *Taenia Solium*. *Taenia solium* has a complex 2-host life cycle. It is a hermaphrodite 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 begins forming proglottids, which are frequently detached from the distal end of the worm and are excreted in the feces. 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 host), and oncospheres (embryos) are liberated by the action of gastric acid and intestinal fluids and actively cross the bowel wall, enter the blood stream and infest various other tissues and organs where they develop into larval vesicles or cysticerci. In humans, this potentially fatal parasitic disease mainly occurs as a result of the ingestion of contaminated food or polluted drinking water, but it may also develop by fecal-oral contamination in tapeworm carriers.

Although the disease is more common in endemic areas like Latin America, Asia, Africa and Eastern Europe, its incidence is also increasing in developed countries as a result of migration of infected persons and frequent travel to and from endemic areas. In humans, cysticerci are most commonly located within the central nervous system (CNS), where it produces a pleomorphic clinical disorder known as neurocysticercosis (NCC), but it may also localize primarily in a variety of tissues, including muscle, heart, eyes, and skin. Although oral involvement by cysticercosis

is common in swine, this location is rare in humans.^{1,3} We hereby present a case of cysticercosis in the sublingual region.

Case Report

24 /M patient presented with H/O sublingual swelling. Routine investigations including CBC , ESR were normal. Clinical diagnosis was ? sublingual mucous cyst .Excisional biopsy was performed. Gross : Specimen consists of a single cystic mass m.1.5 x 1.4 x 1.4 cm. Cut section shows a cyst filled with clear fluid along with a tiny nodule in the cyst wall m. 0.4 x 0.3 x 0.2 cm . Microscopy shows skeletal muscle bundles and fibrocollagenous tissue with an abscess which shows cross section of cysticercus cellulose.

Discussion

Taenia solium is usually acquired by the ingestion of eggs in the food contaminated with feces. Humans are the only definite host, with pigs serving as intermediate hosts only. The larvae hatch in the small intestinal lumen and penetrate the wall to reach their site of encystation commonly subcutaneous tissue, skeletal muscle, brain and head and neck region. The cysticerci in the brain are the most serious manifestation and may be life threatening. CNS lesions are diagnosed by imaging while other accessible lesions are usually diagnosed by excision and histology. Cysticercosis of the oral cavity is rare only a handful of cases have been reported earlier [5-7]. Floor of mouth lesions are rarest among all oral lesions. Oral lesions may appear as mucocele and may be diagnosed as minor salivary tumors, which is the most common differential clinical diagnosis. These patients have to receive medical therapy in the form of Albendazole or Praziquantel, with Niclosamide sometimes being added for CNS lesions. The patient described here underwent surgery as the definitive and diagnostic procedure and is doing well on anti helminthic agents.

Cysticerci are uncommon in the oral cavity of humans where they appear as cystic nodules that may rupture and heal uneventfully.⁴ In swine this location is common. Authors suggest that a high muscular activity and metabolic rate of oral tissues in humans might act against the lodgement and development of cysticercosis in this location⁵

According to literature, oral cysticerci usually elicit a clinical diagnosis of mucocele, or a benign tumour of mesenchymal origin,

such as lipoma, fibroma, hemangioma, granular cell tumour, or a minor salivary gland tumour.^{6,7} Delgado et al⁸ through experience proposed that oral cysticerci are firm nodules on palpation because of its high intraluminal pressure, and therefore considered neither lipoma nor hemangioma as clinical possibilities.

Routine sections stained with haematoxylin and eosin may be all that is required for diagnosis, although in later stages only an inflammatory response to dead larvae may be seen. Fine needle aspiration cytology (FNAC) can also aid in diagnosis as reported in some series, but is subject to sampling error and may be difficult to confirm the diagnosis. Studies have demonstrated that parts of the parasite have been identified in 45% to 100% of the aspirates, particularly when the aspirated material showed a speck of pearly white content that was confirmed to be the larva in acute and chronic inflammatory background by microscopic examination, which may include some eosinophils and palisading histiocytes. The larva has been identified by its lightly stained outer wavy membrane and multiple tiny ovoid nuclei in the fibrillary stroma beneath. In our case the aspirate revealed only RBCs.

Histopathological examination makes up a diagnosis of cysticercosis by the detection of a cystic space containing the cysticercus cellulosa. The scolex has four suckers and a double crown of rostellar hooklets.^{1,4} 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. After a period within three and five years the larva dies and the cyst undergoes calcification.²

Cysticerci may remain alive for many years. The first stage of involution of cysticerci is the colloidal stage, in which the transparent vesicular fluid is replaced by a viscous, turbid fluid. Additionally the scolex shows signs of hyaline degeneration. Thereafter, the cyst wall thickens and the scolex is transformed into coarse mineralized granules. This stage, in which the cysticercus is no longer viable, is termed the granular stage, which anyway does not seem to modify its clinical outcome.^{5,6} Finally, a granulomatous reaction develops that is characterized by histiocytes, epithelioid cells, and foreign body giant cells, leading to fibrosis of the supporting stroma and calcification of the parasitic debris. This pattern of reaction is the same, regardless of the organ involved, and this evolution indicates the age of the infestation.⁸

Laboratory findings in patients with cysticercosis reveal eosinophilia, raised immunoglobulin E (IgE), and most importantly, a positive enzyme linked immunosorbent assay (ELISA) test against cysticercus cellulosa. Anti cysticercus cellulosa antibodies are important in the immunodiagnosis of the disease. This procedure may be performed in serum or cerebrospinal fluid, the latter is considered a diagnostic test for neurocysticercosis.⁸

Drugs as albendazole and praziquantel are potent antihelmintics used in the treatment of cysticercosis^{1,2,3}, replacing niclosamide, which was the drug of choice for the treatment of the disease for a long time.^{1,2} Drugs should be used especially in cases where surgical treatment is risky or not possible, as in neurocysticercosis.

Serological

Antibodies to cysticerci can be demonstrated in serum by EITB (Enzyme Linked Immunosorbent Assay) assay and in CSF by ELISA. An immunoblot assay using lentil-lectin (agglutinin from *Lens culinaris*) is highly sensitive and specific. However, Individuals with intracranial lesions and calcifications may be sero-

negative. In the CDC's immunoblot assay, cysticercosis-specific antibodies can react with structural glycoprotein antigens from the larval cysts of *Taenia solium*.¹⁵ However, this is mainly a research tool not widely available in clinical practice and nearly unobtainable in resource limited settings.

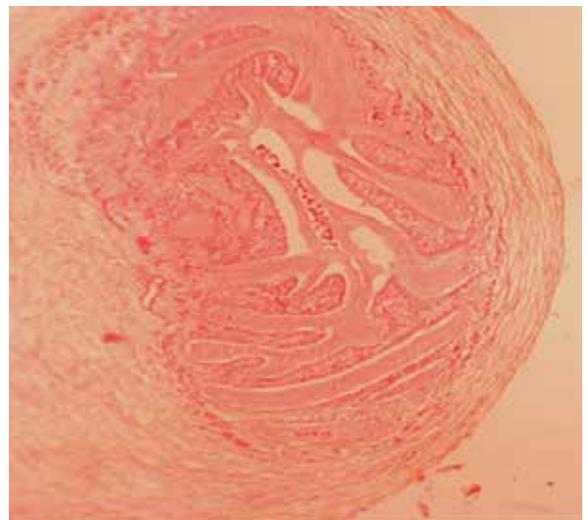
Conclusion:-

Cysticercosis of sunlingual region is rare. Due to its rarity, its presentation as sublingual cystic mass pose diagnostic dilemma even after clinical and radiological investigations. This entity must be kept in mind while dealing with cases of sublingual cystic lump

In summary, we have showed the clinical and histopathological findings in a man with oral cysticercosis, emphasizing the need to consider cysticercosis along with other causes of cystic lesions, particularly in areas with a high incidence of this condition.

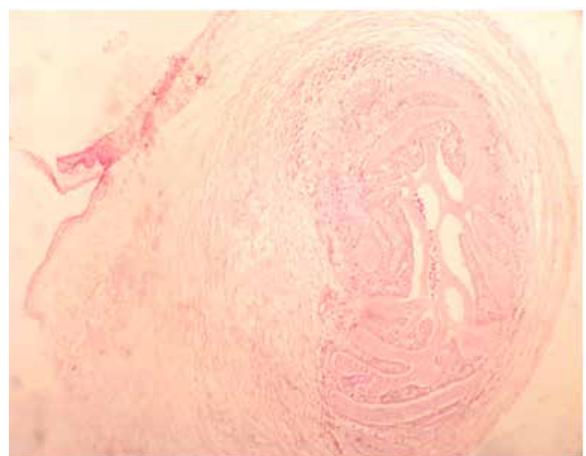
Legends to figures

Figure 1} Microphotograph showing cross section of cysticercus cellulosa. (H & E stain , 40 x).



Legends to figures

Figure 2} Microphotograph showing cross section of cysticercus cellulosa. (H & E stain , 10 x).



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