CASE REPORT

Cysticercosis of the upper lip

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ABSTRACT

Cysticercosis is a parasitic infestation caused by the larval stage of *Taenia solium*. It is common in regions where humans and animals live in close contact, with poor sanitation, and due to consumption of infected meat. The tissues affected are the subcutaneous layers, brain, muscle, heart, liver, lungs, and peritoneum. Oral manifestations are very rare. The most common intra-oral site is the tongue. Here, we present a case in a who sought treatment for an asymptomatic nodule in the upper lip. A gross specimen revealed a cystic cavity containing clear watery fluid and white membranous flecks. The histopathology showed features of *cysticercosis*.

Key words: Cysticercosis, upper lip, *Taenia solium*

INTRODUCTION

Cysticercosis is a potentially fatal parasitic disease that rarely involves the oral region in man. It is relatively common in developing countries of Central and South America, Asia, and Africa, especially in those areas where humans and animals live in close contact, and in those regions where inspection of meat is not strict. In humans, cysticerci are most commonly located within the central nervous system (CNS), where they produce a pleomorphic clinical disorder known as neurocysticercosis. It may also primarily be located in a variety of tissues, including muscle, heart, eyes, and skin. Although, oral involvement by cysticercosis is common in swines, this location is rare in humans.[1] Here we report a case of cysticercosis in an 11-year old-boy with only oral manifestation.

CASE REPORT

An 11-year-old boy reported to the Department of Oral Pathology with a chief complaint of swelling in the upper lip since one year. He complained of a slowly enlarging swelling on the upper lip since one year. The swelling gradually increased in size to the present dimensions.

There was no history of bleeding, trauma, paresthesia or pain. He had no relevant medical or dental history. Dietary history revealed that he was a vegetarian. Intraoral examination revealed a firm, nontender, well-circumscribed nodule on the upper lip. The size of the nodule was about 1 × 1 cm, the shape was circular, and the overlying mucosa was normal. [Figure 1] The extraoral examination had no relevant findings. A provisional diagnosis of fibroma was made. Differential diagnoses were given as mucocele, lipoma, and pleomorphic adenoma. The lesion was surgically excised. On gross examination, the specimen consisted of a well-circumscribed, nodular mass, about 1 × 1 × 1 cm in diameter [Figure 2]. On sectioning, the specimen appeared to be cystic and contained clear fluid. A pearly white mass was seen attached to the sectioned cyst [Figure 3].

Microscopic examination showed a cystic cavity containing the larval form of *Taenia solium* [Figure 4]. The scolex was seen at the cephalic region and caudal to it, a duct-like invagination was noted. This invagination was lined by a eosinophilic membrane [Figure 5]. Higher magnification of the caudal region showed few ovoid basophilic calcified corpuscles [Figure 6]. The diagnosis of cysticercosis cellulosae was made. The patient was symptomless after surgery, to date.

DISCUSSION

The *Taenia solium* parasite or tapeworm is found in the small intestine of man, the definitive host. The terminal segment of the parasite (proglottids) contains eggs and these are excreted in the feces. In areas where unhygienic conditions prevail, the feces are dispersed on the surface of the ground and may be ingested by pigs, the intermediate host. The gastrointestinal secretions of the pig dissolve the eggs and liberate the embryos or enchospheres. These embryos then penetrate the intestinal mucosa and gain access to either the vascular or lymphatic circulation and are thus distributed to various tissues or organs,

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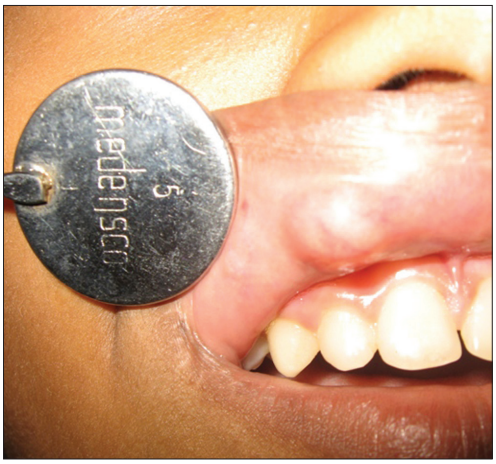

Figure 1: Intraoral photograph showing well-circumscribed nodule (arrow) on the upper lip

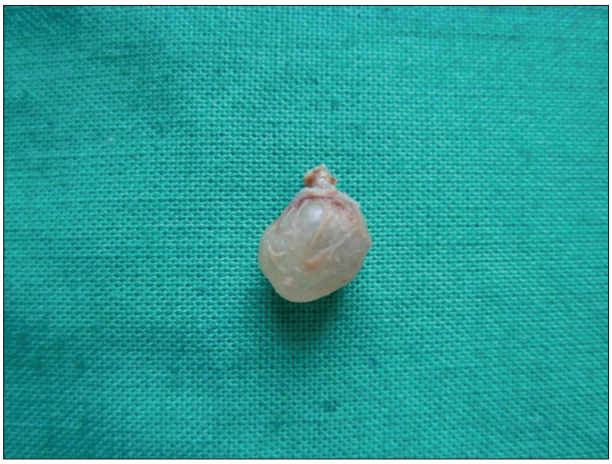

Figure 2: Gross specimen showing a well-circumscribed nodule

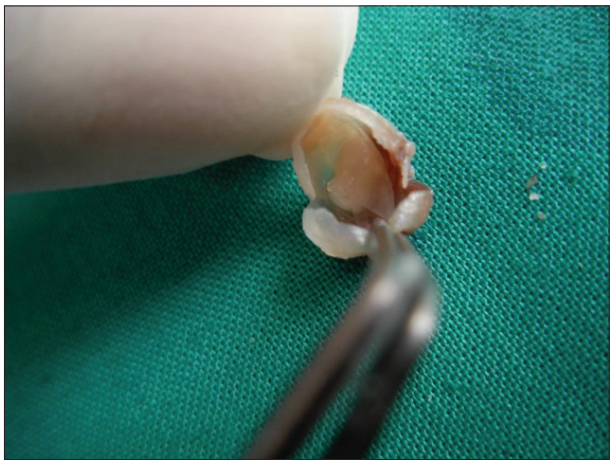

Figure 3: Sectioned specimen showing pearly white mass attached to the cyst

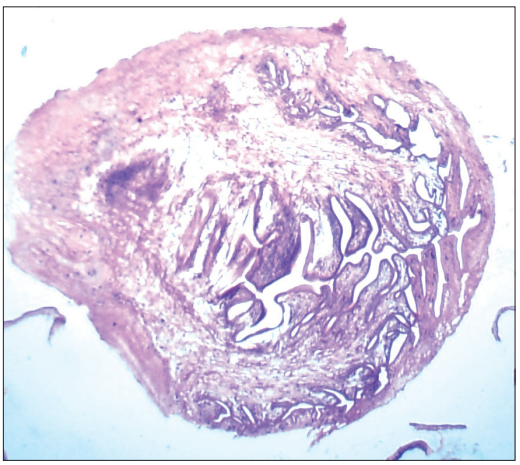

Figure 4: Scanner view showing cystic cavity containing larval form of cysticercus cellulose (H and E stain, 4×)

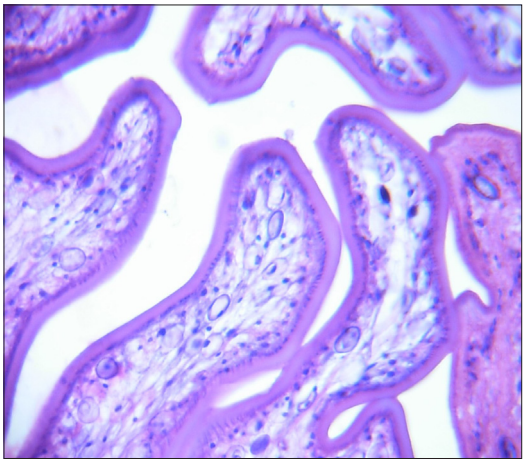

Figure 5: Duct-like invagination lined by eosinophilic membrane (H and E, 10×).

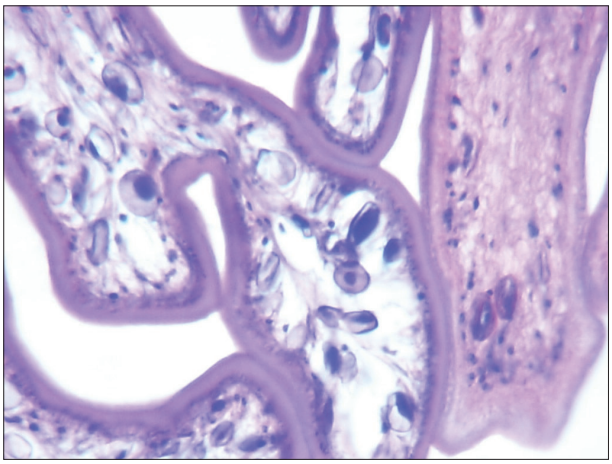

Figure 6: Higher magnification of the caudal region shows few ovoid basophilic calcified corpuscles (H and E section, 40×)

particularly muscles. Here they develop into the larval form known as cysticercus cellulosae. The eating of undercooked and contaminated pork by man results in the larvae reaching the small intestine, where they develop into the adult stage of *Taenia solium*. This is the normal lifecycle of the tapeworm.

In rare circumstances, and mainly as a result of poor hygienic conditions, man may ingest either the eggs or proglottids of the parasite. The larval stage, the cysticercus cellulosae, will then develop in the human. This is likely to happen when carriers of the *Taenia* infect themselves through the

feco-oral route. The eggs or proglottids can gain access to either the duodenum or the stomach through regurgitation. Subsequent penetration of the mucosa by the embryos results in the cysticerci developing in human organs and tissues. Man thus becomes the intermediate host.[2] In the present case, the patient followed vegetarian dietary habits and was reported to be from a region with poor sanitation.

About 97 cases have been reported in English and Spanish literature. There was no definite sex predilection. Of these 50 were males and 47 were females. The age ranged from three to seventy years with a mean of 23.9 years. There were 28 pediatric cases. In order of frequency, the tissues affected by cysticercosis were the subcutaneous layers, brain, muscle, heart, liver, lung, and peritoneum. Its occurrence in the oral cavity was rare. The most common intraoral site found was the tongue. There were only eight cases involving the upper lip. The case reported in our discussion was seen on the upper lip of an 11-year-old boy.[1,3]

Differential diagnosis depends on the location of the lesion. Nodules on lips and cheeks may be considered as fibroma, lipoma, mucocele, pyogenic granuloma, or pleomorphic adenoma. Nodules on the tongue may be considered as fibroma, pyogenic granuloma, granular cell myoblastoma or rhabdomyoma.[2] According to Wilson *et al.*, oral cysticerci are firm nodules on palpation because of their high intraluminal pressure, and therefore, neither lipoma nor hemangioma should be considered as clinical possibilities.[1]

Preoperative diagnosis of cysticercosis can be made by FNAC. R.K. Saran *et al.*, in 1998, reported five cases of cysticercosis diagnosed by fine needle aspiration cytology (FNAC).[3] Parts of the parasite have been identified in 45 – 100% of the aspirates, particularly when the aspirated material shows a speck of pearly white content that is confirmed to be the larva in an acute and chronic inflammatory background, by microscopic examination. The larva has been identified by its lightly stained outer wavy membrane and multiple tiny ovoid nuclei in the fibrillary stroma beneath.[1] In the present case FNAC was not used, as the nodule was firm in consistency and was provisionally diagnosed as fibroma. Fibroma, being a very common lesion, surgical excision was performed.

Other modes of diagnosis are examination of serum, saliva, cerebrospinal fluid (CSF) anlaysis (in case of neurocysticercosis) for antibodies to Cysticercus cellulose, by enzyme-linked immunosorbent assay (ELISA) or enzyme-linked immunoelectro transfer blot (EITB). Plain film radiography can be used when calcifications are present.[3]

When blood investigation was carried out, eosinophil count in the present case was found to be within normal limits. A normal blood profile count was also reported by E Romero De Leon *et al*.[4] However, in cases reported by Timosca G *et al.*, eosinophilia was observed in the blood profile.[5] Thus, it can be said that cysticercosis, although a parasitic condition, may exhibit a blood eosinophil count within normal limits.

Nevertheless, the definitive diagnoses in all the previous published cases have been made by histopathology.

Symptomatology depends on the organs involved. When CNS is involved, the symptoms are related to the anatomical location of the cysticerci, the lesions can produce pressure on different brain centers. The muscular lesions are symptomless. When the lesions are calcified, they can be detected radiologically. In case of oral and subcutaneous forms they are symptomless. They are easily detected clinically because of their superficial location.[2] Cysticerci may remain viable for years. The first stage of involution of cysticerci is the colloidal stage, in which the transparent vesicular fluid is replaced by a turbid, viscous fluid. Additionally, scolex shows signs of hyaline degeneration. Thereafter, the cyst wall thickens and the scolex is transformed into coarse mineralized granules. The stage in which cysticercus is no longer viable, is termed as the granular stage. Finally, a granulomatous reaction develops characterized by histiocytes, epitheloid cells, and foreign body giant cells, leading to fibrosis of the supporting stroma and calcification of the parasitic debris.[1]

The exact duration of each of these stages is highly variable, mainly because of differences in the immune response of the host. In subcutaneous and muscular locations, calcification occurs after about five years. In the present case, the lesion exhibited histopathological changes compatible with viable cysticercus, which suggests that the case was diagnosed shortly after the infestation was produced. This may explain to some extent why the patient had no other clinical manifestations of the disease.

Treatment of choice was surgical excision.[3,6]

The treatment in the other locations is dependent on the symptomatology and the accessibility of the lesion to the surgical intervention. Multicysts and neurocysticercosis are treated therapeutically with drugs like Pranziquantel and Albendazole.[3]

Prognosis in the maxillofacial region is excellent with no recurrence. In contrast to the gravity of the disease in the cerebral, ocular, or cardiac sites, it is often well-tolerated. When associated with other localizations, the prognosis depends upon the site and number of other larval localizations and may be very severe.[5,6] The present case has shown no recurrence to date.

The incidence of oral cysticercosis is rare and it should always be considered as a possibility when nodular formations appear in the mouth, especially when the affected individual lives or comes from a geographical area of high incidence of cysticercus. One cannot rule out the possibility of cysticercosis

even though the patient has a normal eosinophil count and vegetarian dietary habits.

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