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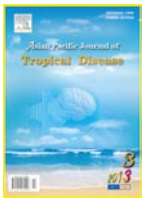

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A solitary facial nodular swelling - A case report of intramuscular cysticercosis in buccinator muscle

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PEER REVIEW

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ABSTRACT

Taenia solium, the larvae of pork tapeworm can cause the parasitic infection known as cysticercosis. It is commonly seen in developing countries. The condition rarely involves orofacial region and represents a difficulty in clinical diagnosis. We present a case report of a healthy middle aged female patient who had a painless swelling on right side of face. The ultrasound examination revealed an intramuscular cysticercosal cyst.

KEYWORDS
Cysticercosis, Differential diagnosis, Oral cysticercosis, *Taenia solium*

1. Introduction

Taenia solium (*T. solium*), is a cyclophyllid cestode belonging to the family of Taeniidae and is also known as the pork tapeworm. Life cycle of *T. solium* is very similar to *Taenia saginata*. It has three distinct morphological verities[1]. Cellulose cysticercus type has a fluid filled bladder with an invaginated scolex. The racemose type has no evident scolex but is believed to be larger and much more dangerous. The intermediate form has a scolex. Humans beings are infected by eating uncooked or partially cooked pork containing cysticercus cellulose. Inside the alimentary canal of the man, the scolex of the cysticercus evaginates and attaches to the gut wall by its suckers and then develops into an adult worm and the eggs or gravid proglottids are passed out with faeces[2]. Sometimes humans acts as intermediate host where eggs get into the stomach as a result of retroperistalsis or contaminated hands[3].

T. solium that causes cysticercosis is endemic to several parts of the globe including China, Southeast Asia, India, sub-Saharan Africa, and Latin America[2,4,5]. The World Health Organization estimates that over 50 000 deaths per year are caused by neurocysticercosis worldwide[5]. It is more commonly in cerebral tissue, subcutaneous tissue, muscles, liver, lungs and eye[4,6-8].

Oral cysticercosis is very rare in the oral and maxillofacial region although the muscular tissue is usually asymptomatic[2]. According to the literature reports, the prevalence of oral cysticercosis is 4.1%. The most commonly involved intraoral sites are buccal mucosa, tongue and lips[9].

Here, we highlight a case of persistent asymptomatic facial swelling after extraction of root stump leading to an unusual diagnosis of cysticercosis involving right buccinator muscle.

2. Case report

A 35-year old female patient reported to outpatient in the Department of Oral Medicine and Radiology, the

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Oxford Dental College and Hospital, Bangalore, with a chief complaint of swelling on right side of face since 10 days. Painless swelling was not related to any traumatic episode. There is no history of previous swelling in the same location.

Past dental history revealed pain in the right upper molar teeth from past one month for which she visited local dentist and extraction of root stump in relation to upper right molar region was performed without any complications followed by medication. Patient was under long term antibiotic medication for the same reason. Personal history revealed that she works in field and vegetarian.

Clinical examination revealed a diffuse extra oral swelling located 2.5 cm away from the anterior tragus of the right ear, measuring approximately 2.0 cm×2.5 cm. Skin over the swelling was normal in colour. On palpation swelling was firm in consistency, non tender, no rise of local temperature and non-fluctuant (Figure 1).

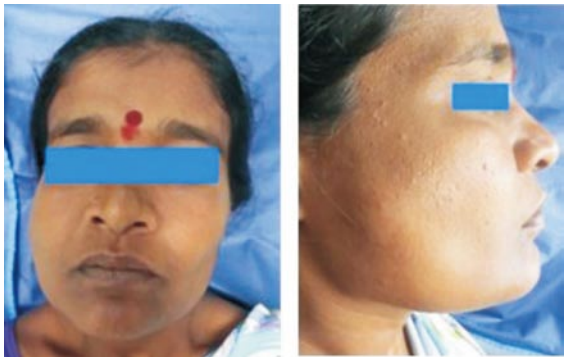

Figure 1. Swelling on the middle one third of right side of face.

Based on above history and clinical findings, a provisional diagnosis of antibioma of right cheek was suspected with differential diagnosis of soft tissue abscess and right parotid gland sailadenitis.

Aspiration of swelling was performed yielding 0.2 m blood mixed pus. Complete hemogram revealed increased number of eosinophils suggestive of parasitic infection.

Ultrasound examination revealed a partially collapsed intramuscular cystic lesion measuring about 7.9 mm×4.2 mm seen in right cheek and mural calcified nodule was noticed within the lesion. A diagnosis of intramuscular cysticerosal cyst was given by radiologist (Figure 2).

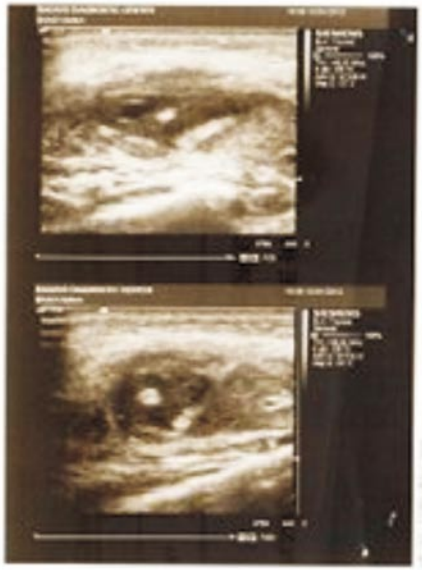

Figure 2. Ultrasound showing partially collapsed intramuscular cystic lesion measuring about 7.9 mm×4.2 mm with mural calcified nodule within the lesion.

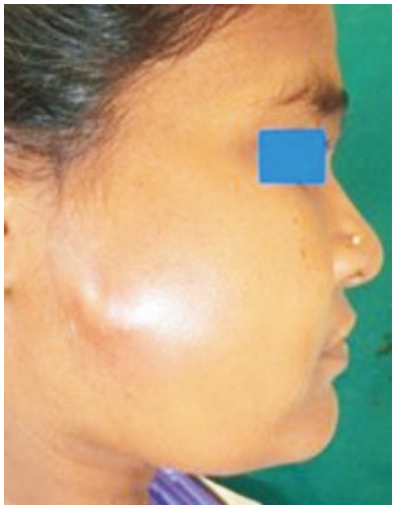

Figure 3. Second visit revealing reinfection of the swelling.

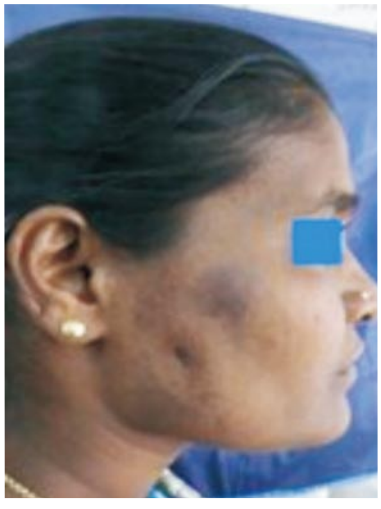

Figure 4. Post therapy complete resolution of swelling.

Patient was referred to the physician at Saint John's Hospital, to rule out cysticercosis in other locations of the body. Since both symptoms of CNS, GI disturbances and general physical findings were negative patient was prescribed albendazole 400 mg once daily for 21 d.

After 2 d, patient noticed increase in the size of the swelling associated with pain. Skin over the swelling was tense and shiny (Figure 3).

Consistency of swelling was firm. Due to reinfection of swelling, incision and drainage was performed along with curettage of granulation tissue and it was sent for histopathological examination which revealed presence of increased number of eosinophils.

After completion of anthelminthic therapy, patient revealed complete resolution of the swelling (Figure 4). Ultrasound examination also revealed resolving cysticercosal lesion (Figure 5).

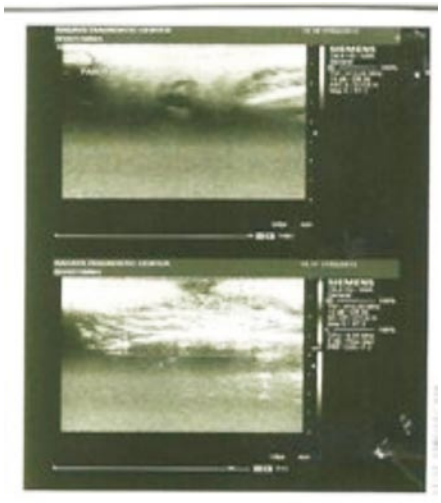

Figure 5. Ultrasound showing resolving cysticercosal lesion.

3. Discussion

The worm passes its life cycle in two hosts: the definitive host and the intermediate host. Man is the definitive host and pig is the intermediate host.

The terminal segments of the parasite (proglottids) contain eggs, and these are excreted with the faeces. In areas where hygiene is not maintained, the faeces are dispersed on the surface of the ground and the animal become infected by swallowing these eggs(the intermediate host). The gastro-intestinal secretions of the pig dissolve the eggs and liberate the embryos or onchospheres. The eating of partially cooked and contaminated pork by humans results in the larvae reaching the intestine, where they develop into the adult worms (Figure 6).

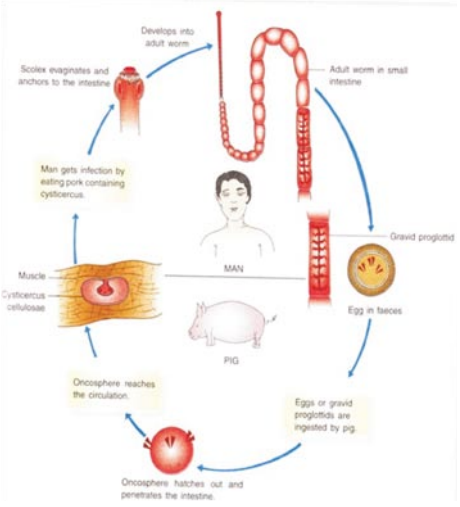

Figure 6. Life cycle of *Taenia solium*.

These embryos hatches out and penetrate the gut and gain access to either the vascular, or lymphatic circulation and are thus lodged into various tissues and organs, mainly muscle. They are transformed into cysticercus stage in the muscle and these are known as the cysticercus cellulosae[10].

The growing larva in cysticercosis may provoke a series of inflammatory reactions including infiltration of neutrophils and eosinophils, lymphocytes, plasma cells and at times giant cells, followed by fibrosis and necrosis of capsule with eventual caseation or calcification of the larva[4]. Cysticerci may remain alive for many years. The first stage of involution of cysticerci is the colloidal stage, in which a viscous, turbid fluid replaces the transparent vesicular fluid. Additionally, the scolex shows signs of hyaline degeneration. There-after, the cyst wall thickens and the scolex is transformed into coarse mineralised granule is termed the granular stage[2,4].

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 calcifications of the parasite debris[4].

This pattern of reaction is the same regardless of the organ involved, and this evolution indicates the age of the infestation. The exact duration of each or these stages is highly variable, mainly because of considerable differences in the immune response of the host[4].

The most frequently affected decade was the third (32%), followed by the fourth decade (20.6%). There was an equal distribution between genders[2,4].

The most frequently involved site were the tongue (42.15%), followed by the lips (26.15%) - with the lower lip 64.7% and the buccal mucosa (18.9%). Several reports revealed multiple foci in the same patient[2].

Once a person becomes the host of cysticercus cellulosae, cysticercosis can develop in various organs

and tissues[5]. Clinical effects vary depending on site of larval lodging, larval burden and host reaction. Intestinal infestations with *T. solium* may be asymptomatic.

Epigastric discomfort, nausea and diarrhea are GI symptoms[11]. Generalised symptoms include headache, fever and myalgia[5,10].

Patients with neurocysticercosis can present with several signs and symptoms, the most frequent being seizures, increased intracranial pressure, obstructive hydrocephalus, meningitis and mental disorders[2,12,13].

Delgado-Azanero *et al.* suggested that the possibility of longer exposure of neurocysticercosis may promote the malignant transformation of glial and lymphoid cells[4].

Oral lesions are nodular swellings and may be asymptomatic. It was found that the lesion on the tongue could interfere with movement, causing discomfort during speaking and eating. Although, oral cysticercosis indicates disseminated infestation, systemic complications are not demonstrated in most of the patients with oral lesions.

The differential diagnosis of oral cysticercosis depends on the location of the lesion. Nodules on the lips and cheeks may be considered as fibroma, lipoma, mucocele, pyogenic granuloma or pleomorphic adenoma[2,5]. Nodules on the tongue may be considered as fibroma, pyogenic granuloma, granular cell myoblastoma or rhabdomyoma[2,10,11].

Saran *et al.* and Mazhari *et al.* proposed that FNAC is also a well-accepted procedure for reliable and quick preoperative diagnosis of cysticercosis to help the clinician in planning the treatment[2]. Other diagnostic tools such as radiologic imaging and serology can be used. Besides, normal radiographic examination, other modalities of imaging are very effective in the detection of cysticerci such as computerised tomography, ultrasonography and magnetic resonance[8,9].

As the cysts can lodge in multiple locations, all patients with cysticercosis should have an ophthalmologic examination to rule out ocular involvement, and all patients with extraneurologic cysticercosis should have computed tomography or magnetic resonance imaging of the brain to rule out neurocysticercosis[9].

Laboratory findings of blood profile noticed eosinophilia, raised Immunoglobulin E and most importantly, a positive enzyme-linked immunosorbent assay (ELISA) test against cysticercus cellulose[7].

Immunodetection of cysticercosis can be achieved in sera, cerebrospinal fluid (considered diagnostic for neurocysticercosis, although they are not 100% sensitive) and saliva, by ELISA or enzyme-linked immunoelectrotransfer blot, but it is important to consider that individuals living in an endemic area may have antibodies because of an exposure instead of an established infestation[14].

Superficial cysticerci and solitary asymptomatic nodule of oral cysticercosis were managed by simple surgical excision and the periodic follow up[15].

Anthelminthic drugs such as praziquantel and albendazole are given in the cases, where surgical treatment is not possible, risky or neurocysticercosis condition. Corticosteroids are used as adjuvant to counteract the development of inflammatory reaction after the death of larvae. To prevent epileptic seizures anticonvulsants are used[5,8-11,16]. As the patient in the present case had no occurrence of cysticercosis at any other site or any other symptoms, no addition treatment was prescribed except the periodic follow-ups.

Cysticercosis can be eradicated. An efficient preventive program, which incorporates pork inspection, well washed vegetables, consumption of filtered or boiled water, and efficient hand washing before meals and food preparation, is the key factor in basic sanitation to eliminate the infection[17].

Dentists should be aware of the existence of such lesions and have important role in detection of oral cysticercosis, which may resemble clinically benign connective tissue lesions. Primary treatment for oral cysticercosis is simple surgical excision; however, it is important to carry out a detailed medical evaluation in every case, in order to exclude the presence of the parasite at other sites. The patient should be followed up for ophthalmologic, neurologic signs and symptoms of cysticercosis cellulosae. The prevention and complete eradication of disease is feasible through education programmes on the life cycle of the parasite and importance of personal hygiene.

Conflict of interest statement

We declare that we have no conflict of interest.

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Comments

Background

Infection caused by taeniasolium has more prevalent in developing countries where unhygienic condition prevails. Cysticercosis is a condition that occurs when human are infested by the larvae of *T. solium*. Oral cysticercosis

is a rare event, and it represents a difficulty in clinical diagnosis.

Research frontiers

Usually oral cysticercosis is diagnosed on the bases of histopathology examination. But in this case, it was diagnosed on the bases of ultrasound examination. The lesion is commonly seen in non vegetarians and in present case report the patient was purely vegetarians.

Related reports

There is no case reports of oral cysticercosis diagnosed on the bases of ultrasound examination, and which is in agreement with Vijayraghavan *et al*. presented the ultrasound appearance of the cysticercosis lesions.

Innovations & breakthroughs

This case report has suggested that oral cysticercosis can be considered in differential diagnosis of oral painless swelling. Ultrasound examination can be modality of diagnosing this condition.

Applications

Specially, people in developing countries need to be educated on the parasitic infection and importance of maintain hygiene so that this infection can be prevented.

Peer review

This paper presents a rare case and it is in vegetarian patient. Only few reports are present in literature. Authors have mentioned about importance of early diagnosis of this condition and can be diagnosed on the bases of ultrasound examination which has the potential to be completely eradicated by patient education on the life cycle of teaniasolium and importance of maintaining sanitation.

References

[1] Rabiela M, Rivas A, Flisser A. Morphological types of *Taenia solium* cysticeci. *Parasitol Today* 1989; **5**: 357-360.

[2] Elias FM, Martins MT, Foronda R, Jorge WA, Arajio NS. Oral cysticercosis : case report and review of the literature. *Rev Inst Med Trop São Paulo* 2005; **47**(2): 95-98.

[3] Garcia HH, Del Brutto OH. *Taenia solium* cysticercosis. *Infect Dis Clin North Am* 2000; **14**: 97-119.

[4] Delgado-Azañero WA, Mosqueda-Taylor A, Carlos-Bregni R, Del Muro-Delgado R, Díaz-Franco MA, Contreras-Vidaurre E. Oral cysticercosis: a collaborative study of 16 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007; **103**: 528-533.

[5] Ribeiroet A, Luvizotto M, Soubhia A, de Castro A. Oral cysticercosis: case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007; **104**: e56-e58.

[6] Dhaif GA, AI-Hadi AA. Oral cysticercosis: A case report. *Saudi Dent J* 2000; **12**(2): 100-102.

[7] Rao SC, Sharma H, Vinay KN, Vidya KC. Oral cysticercosis - A case report. *Int J Clin Dent Sci* 2011; **2**(4): 36-39.

[8] Shetty RM, Ramprasad, Kalra A, Goyal S, Sachin BM, Shetty S, et al. Cysticercosis cellulosae in labial mucosa: a rare case report. *J Indian Acad Oral Med Radio* 2010; **22**(4): S48-S50.

[9] Jay A, Dhanda J, Chiodini PL, Woodrow CJ, Farthing PM, Evans J, et al. Oral cysticercosis. *Brit J Oral Max Surg* 2007; **45**: 331-334.

[10] Kumar BP, Mohan AP, Col Kumar KAJ, Rao JB, Kumar HR. Oral cysticercosis- Review of literature. *Indian J Dent Adv* 2011; **3**(1): 438-441.

[11] Júnior HM, Filho MR, Santos LN. Oral cysticercosis. *Braz J Oral Sci* 2006; **5**(18): 1109-1111.

[12] Garcia HH, Gonzalez AE, Evans CAW, Gilman RH. *Taenia solium* cysticercosis. *Lancet* 2003; **361**: 547-556.

[13] Yeh J, Jeanne S. Cysticercosis: a zebra in the neighborhood. *Virtual Mentor* 2008; **10**(4): 220-223.

[14] Garcia HH, Harrison LJ, Parkhouse RM, Montenegro T, Martinez SM, Tsang VC. A specific antigen-detection ELISA for the diagnosis of human neurocysticercosis. The Cysticercosis Working Group in Peru. *Trans R Soc Trop Med Hyg* 1998; **92**: 411-414.

[15] Gadbail AR, Korde S, Wadhwan V, Chaudhary M, Patil S. Oral cysticercosis: report of two cases with review of literature. *Oral Surgery* 2010; **3**(1-2): 51-56.

[16] Del Brutto OH, Roos KL, Coffey CS, Garcia HH. Meta-analysis: cysticidal drugs for neurocysticercosis: albendazole and praziquantel. *Ann Int Med* 2006; **145**(1): 43-51.

[17] Garcia HH, Del brutto OH. Cysticercosis working group in peru neurocysticercosis: updated concepts about an old disease. *Lancet Neurol* 2005; **4**: 653-661.