Paediatric Oral Cysticercosis: A Misdiagnosed and a Rare Entity
Varsha Omprakash Bhatia, Ashwini Arvind Natekar, Arvind Govind Valand

Abstract
Cysticercosis cellulosa is a common disease in developing countries, but oral lesions caused by this parasitic infestation are rare. Specifically, buccal mucosa is an uncommon site for cysticercosis cellulosa. There are very few cases of solitary cystercosis of buccal mucosa in paediatric patients which have been reported in the literature. In this report we document a case of oral cysticercosis in an eight year old girl who sought treatment for asymptomatic nodule of the right buccal mucosa that had been clinically misdiagnosed as submucosal retention cyst.

Keywords: Cellulosa; Cyclophyllidcestode; Paediatric; Misdiagnosis; Oral Cysticercosis; Taenia Solium

Introduction
Taenia solium (T. Solium) is a cyclophyllidcestode belonging to the family of Taeniidae and is also known as the pork tapeworm. Human beings are infected by eating uncooked or partially cooked pork containing cysticercus cellulose. T. Solium that causes cysticercosis is endemic to several parts of the globe including China, Southeast Asia, India, Sub Saharan Africa and Latin America especially in those areas with poor sanitation where humans and animals live in close contact. Oral cysticercosis is very rare in the oral and maxillofacial region.

According to the literature reports, the prevalence of oral cysticercosis is 4.1%. The most commonly involved intraoral sites are tongue followed by lips and buccal mucosa. Subcutaneous tissue, brain, muscle, heart, liver, lungs and peritoneum are more frequently affected but oral cysticercosis is a rare event. It is often a diagnostic challenge to the clinicians and the diagnosis is usually made on microscopic examination.

Case Report
We report a case of eight year old Hindu female and a resident of slum in Mumbai. The patient came with swelling inside the oral cavity on the right buccal mucosa since one year which gradually increased in size. On local examination the swelling was non tender, firm and mobile measuring 1x0.5cm. She had bilateral cervical lymphadenopathy since many months. She did not give any history of fever/ cough/ contact with tuberculosis/ loss of weight/ reduced appetite. Her vital parameters were within normal limits. Systemic examination did not reveal any significant findings.

She was referred to ENT department for cervical lymph nodes to rule out tuberculosis and was advised chest x-ray which was within normal limits. Her complete blood counts were within normal limits. She was referred to paediatric surgery department for removal of swelling under local anaesthesia. The probable clinical diagnosis was given as submucosal retention cyst and the tissue was sent for histopathological examination. We received a cyst measuring 1x0.5x0.3cm, externally smooth and pearly white in appearance. On cutting open clear fluid was seen.

On microscopic examination, Haematoxylin and Eosin (H&E) stained sections showed external fibrous capsule with mononuclear cell infiltrate comprised of lymphocytes, histocytes and plasma cells. A double layered membrane consisting of an outer acellular hyaline eosinophilic layer and an inner sparsely cellular layer with aggregates of eosinophils was seen. The cyst contained larval form of T. Solium (Figure 1). The cephalic extremity of larva (scolex) with suckers could be identified suggesting the diagnosis of cysticercosis of right buccal mucosa (Figure 2). Patient was given anthelmintic therapy and on follow up there was complete resolution of cervical lymphnode swelling and no neurological deficit was seen.
Discussion
Kuchenmaister in 1855 established that human cysticercosis is caused by larval stage (Cysticercosis cellulosae) of the pork tapeworm (T. solium). It is the “biological marker” of the social and economic development of a community.\textsuperscript{4} Inspite of the high prevalence of cysticercosis in the developing countries of world, oral and perioral lesions are relatively rare. A thorough search in the English literature showed less than 70 cases have been reported till date,\textsuperscript{3,5} of which only 28 paediatric cases (<12 years) have been described.\textsuperscript{5,6} Our case was of an eight year old female. Also amongst sites of oral cysticercosis, buccal mucosa involvement is relatively rare as compared to tongue and lip.\textsuperscript{1,4} Only 14 cases of buccal mucosa cysticercosis is reported (six cases worldwide and eight cases in India).\textsuperscript{4} Only two paediatric cases\textsuperscript{6,7} involving buccal mucosa have been reported as per our knowledge. Our patient also had swelling over buccal mucosa.

Cysticercosis is a potentially fatal parasitic infestation that rarely involves oral regions in humans. Oral cysticercosis usually manifests as a well circumscribed, soft swellings. Cysticercosis is rarely included in the pre-operative differential diagnosis due to relative rarity of the lesion, inadequate knowledge of oral manifestations of parasitic infections and negligence while taking medical history.\textsuperscript{8} It is usually misdiagnosed as a mucocele or a benign tumor of mesenchymal origin such as lipoma, fibroma, hemangioma, granular cell tumor or minor salivary gland adenoma.\textsuperscript{9} In our case the probable clinical diagnosis was given as submucosal retention cyst. Thus, the diagnosis of cysticercosis requires histopathological examination, although few cases have been diagnosed on Fine Needle Aspiration Cytology.\textsuperscript{10}

As cysticercosis involves multiple locations, other investigations to be carried out are ocular examination to rule out ocular involvement and magnetic resonance imaging of brain to rule out neurocysticercosis, computed tomography to rule out extra-neurologic cysticercosis.\textsuperscript{1} Cysticercosis outside CNS is a benign condition and usually does not require specific treatment. Cysticercosis is mostly seen in population having non-vegetarian diet and less commonly seen in Hindu religion as there is no intake of pork meat in their diet as was seen in our case of young Hindu female. Thus to prevent and eradicate human cysticercosis supportive measures such as improvement in the sanitary conditions, pork inspection, consumption of boiled water, well washed vegetables, mass education about personal hygiene should be undertaken along with medical treatment which includes larvicidal drugs, corticosteroids, surgical procedure for removal of the cyst.\textsuperscript{3,4}

Conclusion
Buccal mucosa is a very rare site of involvement by cysticercosis in a paediatric patient, even in an endemic area. Histopathological findings of the excised swelling are diagnostic of the lesion. However detailed evaluation should be done to exclude the presence of the parasite at other sites.

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Author Affiliations
1. Dr. Varsha Omprakash Bhatia, Assistant Professor, 2. Dr. Ashwini Arvind Natekar, Senior Resident, Department of Pathology, 3. Dr. Arvind Govind Valand, Professor & Head, Grant Government Medical College and Sir JJ Group of Hospitals, Byculla, Mumbai, India.

References

Corresponding Author
Dr. Varsha Omprakash Bhatia, 8/a, Jaya Apartments, Cama Lane, Ghatkopar (west), Mumbai-86, India. Ph: +91 9867571769 E-mail: drvarshabhatia@gmail.com

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