Case Report

An atypical case of cutaneous cysticercosis in buccal mucosa

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Abstract

Cysticercosis is a recognized fatal disease to the humans. *Taenia solium,* a larval stage of pork tapeworm that causes cysticercosis which is an important cause of morbidity in the world. This case report shows the clinical presentations of an atypical case of cutaneous cysticercosis on the right buccal mucosa and its management. It presents the importance of thorough knowledge, proper investigation, symptomatic management with improved treatment regimens and important role of minimally invasive surgery. Its outcome and the treatment options mainly depend on the number, location, size and stage of parasites, as well as on the immune response of the host.

Keywords: Buccal mucosa, cysticercosis, oral cysticercosis, *Taenia solium,* taeniasis, treatment

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INTRODUCTION

Cysticercosis is a potentially fatal parasitic disease that seldom involves the oral region in humans.[1] *Taenia solium* (*T. solium*), larval stage of tapeworm that causes cysticercosis.[2] The larval stage of *T. solium* resides in tissues and muscles of pigs that serve as an intermediate host. Accidental ingestion of inadequately cooked pork containing *T. solium* leads to infection in human beings. Pigs are the primary host, whereas human beings can act as both definitive and intermediate hosts. Cysticercosis is acquired mainly by the fecal–oral route and not necessarily by eating pork, thus even vegetarians can acquire this disease.[3,4] Commonly affected sites include subcutaneous tissue, brain, muscle, heart, liver, lungs and the eyes.[5] The disease is a potential threat because the ingested eggs develop into embryos (oncospheres) that can penetrate the intestinal wall and disseminate through the vascular or lymphatic circulation to develop into cystic larvae (cysticercus cellulosae).[1]

Oral cysticercosis is a rare quintessence, which is a diagnostic challenge as it mimics other benign tumors. In this case report, we have highlighted the importance of thorough knowledge, radiographic and histopathological evaluation for diagnosis and surgical management of oral cysticercosis.

CASE REPORT

In this case report, a 26-year-old female reported to Shri Sathya Sai Medical College and Research Institute with a chief complaint of swelling inside the cheeks for about 6 months with no history of pain, tenderness and discharge. The swelling initiated slowly and gradually increased in

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size for the past 1 month. The patient did not report any past medical or dental history. On clinical examination, no cervical lymph nodes were palpable. There was a swelling present on the right buccal mucosa which was soft in consistency, well encapsulated, present subcutaneously and nonfixed [Figure 1]. Based on clinical examination, the preoperative provisional diagnosis was narrowed down to fibroma, lipoma and adenomatoid odontogenic tumor. Magnetic resonance imaging (MRI) of buccal mucosa, chest X ray, ultrasonography (USG) of the abdomen, pelvis, parotid gland, cheeks and thyroid was advised as a part of the investigation. The chest X-ray and USG of the thyroid, abdomen and pelvis revealed no abnormality. On USG, there was a benign, well-defined hypoechoic mural focus deep to masseter muscle measuring 1.0 cm × 1.1 cm [Figure 2]. To exclude the possibility of involvement of any other structure, MRI was carried out. The MRI of buccal mucosa showed the presence of a well-defined T1W hypointense T2W hyperintense lesion in the premaxillary fat posterior to the right zygomaticus major muscle. The lesion appears to abut the zygomaticus major muscle suggestive of fluid within the lesion. The scolex of the lava was better seen in the T1 image as a hyperintense nidus [Figure 3]. The findings of the MRI supported cysticercosis. A simple buccal incision was carried out with number 15 size BP blade and the cystic mass was completely enucleated from the zygomaticus major muscle [Figure 4]. 3'0 vicryl sutures were placed following the surgery. The specimen was sent for histopathological evaluation [Figure 5]. Gross grayish white soft-tissue specimen measuring 1.5 cm × 1.8 cm × 1.3 cm with the cut surface of cyst measuring 0.5 cm × 0.5 cm was firm in consistency. On microscopic evaluation, it showed an outer dense fibrous layer with inner mixed inflammatory cell collection predominantly neutrophils, lymphocytes, eosinophils, granulomatous reaction and central irregular cavity. Few foci shows small cyst with thin middle cellular layer and a thick inner layer containing loosely packed myxoid matrix with scattered small canaliculi. One of the foci shows two thickened cyst walls with scolex transforming into coarse mineralized granules with areas of mature adipose tissue. This draws the final diagnosis of cysticercosis on the right buccal mucosa. The patient was prescribed with antihelminthic drug albendazole 400 mg twice daily for 28 days and a periodic follow-up was advised. The patient was under follow-up for 7th day, 3 months and 6 months with no recurrence [Figure 6].

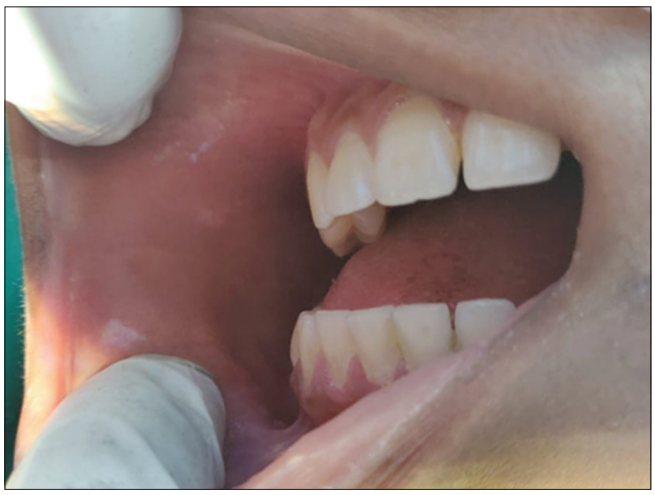

Figure 1: Preoperative

DISCUSSION

Oral cysticercosis is a rare infection that can elicit a clinical diagnosis of a mucocele or benign tumors. Cysticercosis is caused by cysticercus cellulosae, the larval stage of *T. solium* or pork tapeworm.[6-8] *T. solium* passes its life cycle in two hosts. The intermediate host is pig which harbors the larval stage and the definitive host is human who harbors the adult worm.[1] The adult worm which is 3 m can grow up to 7 m long and lives in the small intestine of man for years. Each worm can bear 1000 proglottids (eggs). Proglottids are frequently detached from the distal end of

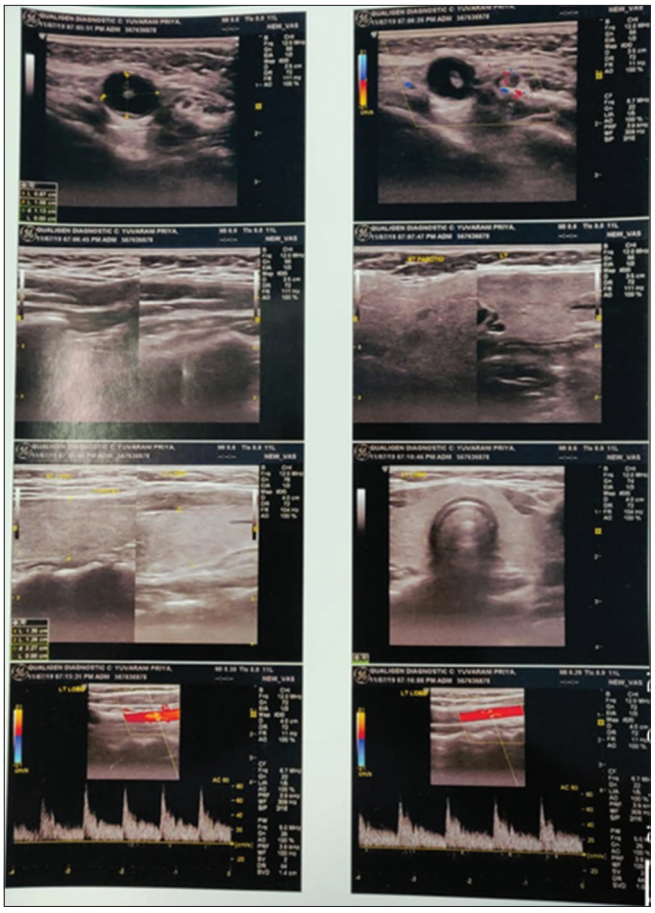

Figure 2: Ultrasound of the parotid gland and cheeks

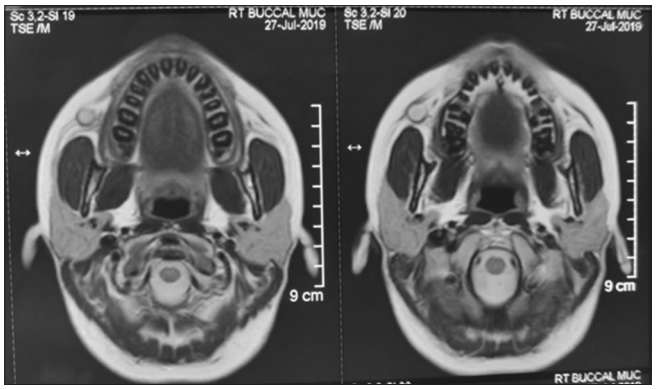

Figure 3: Magnetic resonance imaging of the head

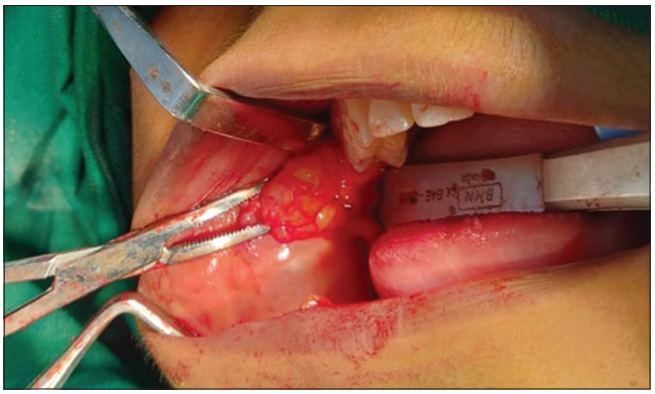

Figure 4: Surgical removal of the lesion

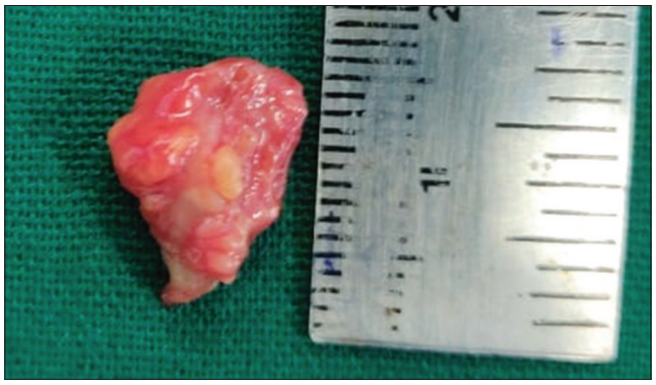

Figure 5: Surgically excised lesion tissue specimen measuring 1.5 cm × 1.8 cm × 1.3 cm

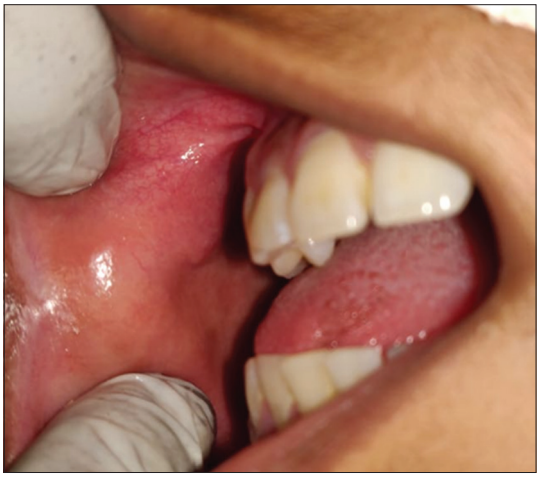

Figure 6: Postoperative

the worm and are excreted in the feces. Each proglottid contains 50,000–60,000 fertile eggs, which remain viable for a prolonged time period in water, soil and vegetation.[8,9] These eggs may infect pigs and its lifecycle goes on as humans being the intermediate host. This ingestion of the eggs can occur not only with the ingestion of infected cook but also with the consumption of contaminated food or water in case of vegetarians.[10]

Fecal–oral infection by hands contaminated with eggs or retrograde reflux of proglottids into the stomach can have a similar effect. The life cycle initiates by the ingestion of tapeworm "eggs" releasing embryos (oncospheres) in the intestine of the intermediate host. Due to the action of gastric and intestinal juices, these oncospheres are released and they penetrate the intestinal wall. The oncospheres travel through the blood and lymphatic stream to reach and get attached in various body organs, where they develop into cysticercus (the encysted larvae) harboring the larval stage of the worm. The cysticercus, when ingested by humans eating undercooked pork, gets released in the human intestine and its scolex attaches to the mucosa. The larval stage further develops into the adult tapeworm and the life cycle completes.[1,11]

Parasitic infections are endemic diseases, but parasitic oral infections are rare cases.[12] Cysticercosis is endemic in developing countries such as Russia, China, India, Mexico, Latin America, Eastern Europe and southern Africa.[13] Subcutaneous tissue, brain, muscle, heart, liver, lung, peritoneum and oral cavity being the least common is the order of inclination of the cysticercus attachment in the human body.[1]

Oral cavity and perioral involvement are rare in humans. A case report by Krishnamoorthy *et al*. in 2012 showed a total of 65 cases of oral (including lingual, buccal, labial and gingival; excluding masseter and parotid) cysticercosis. On a thorough review of the literature, the most common site of involvement was the tongue (45 cases, 50.6%), followed by lip (24 cases, 27%) and buccal mucosa (17 cases, 19.1%).[14] The clinical presentation of the infection is with a solitary, painless nodule or multiple nodules. Jay *et al*. reported a case of multiple intraoral cysticercus nodules involving buccal, labial and lateral tongue border.[15]

In a rare scenario, mild pain may be associated with superimposed infection. Differential diagnosis of intraoral

cysticercosis is site-specific and involves mucocele, benign tumors of mesenchymal origin including lipoma, fibroma, benign minor salivary gland neoplasms, hemangioma and lipoma. The latter two have a softer consistency and on the basis of this feature, may be differentiated from the tense cysticercus swelling.[16]

Localization in the brain due to neurocysticercosis is generally symptomatic with headache, raised intracranial pressure or epilepsy. Thus, it is easier for diagnosis through MRI and computed tomography scan of the brain.[17] However, most of the oral cysticercosis is asymptomatic. The only subjective symptom by the patient is growth in the subcutaneous tissue which is disturbing. The definitive final diagnosis can be drawn by the demonstration of cysticercus cellulosae on histopathology. To determine the intraoral location, ultrasonography (USG) can be used as an adjunctive aid to MRI to locate the presence of the cyst.[18]

Under the differential diagnosis of this case report, lesions such as fibroma, lipoma and peripheral adenomatoid odontogenic tumor were considered. Fibroma is the most common benign connective tissue tumor and can occur anywhere in the oral cavity as a firm, painless swelling and it most commonly occurs on the buccal mucosa and tongue as a pedunculated mass. Lipoma also appears as a painless swelling, with a slip sign on a smooth surface. Peripheral odontogenic lesions are very rare but can occur close to soft tissues adjacent to the tooth structure, but all the signs and the results of the investigation proved the presence of oral cysticercosis on the right buccal mucosa. To conclude, a thorough knowledge and prompt treatment should be carried out to prevent cysticercosis which may be fatal.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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