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

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.³⁴ 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).⁶

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...
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 larva 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].

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. *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. 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
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. 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.

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.

Parasitic infections are endemic diseases, but parasitic oral infections are rare cases. Cysticercosis is endemic in developing countries such as Russia, China, India, Mexico, Latin America, Eastern Europe and southern Africa. 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.

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%). 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.

In a rare scenario, mild pain may be associated with superimposed infection. Differential diagnosis of intraoral
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Conflicts of interest
There are no conflicts of interest.

REFERENCES

1. García HH, Del Brutto OH. Taenia solium cysticercosis. Infect Dis Clin North Am 2000;14:97-119.
2. Ribeiro AC, Luvizotto MC, Soubbia AM, de Castro AL. Oral cysticercosis: Case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;104:e56-8.
3. Mohan H. Infectious and parasitic diseases. In: Mohan P, Mohan T, editors. Textbook of pathology. New Delhi, India: Jaypee Brothers Medical Publishers Ltd. 2005. p. 195-6.
4. Jay A, Dhandha J, Chiodini PL, Woodrow CJ, Farthing PM, Evans J, et al. Oral cysticercosis. Br J Oral Maxillofac Surg. 2007;45:331-4. Epub 2006 Jan 18.
5. Saran RK, Rattan V, Rajwanshi A, Nijawan R, Gupta SK. Cysticercosis of the oral cavity: Report of five cases and a review of literature. Int J Paediatr Dent 1998;8:273-8.
6. Patel K, Shah M, Patel B, Doshi N. Subcutaneous oral cysticercosis. Natl J Community Med 2011;2013:311-3.
7. Mukheh S, Kacker SK, Kapilla K. Cysticercosis of the oral cavity—a clinico pathological study of ten and a half years. JDA 1986;2013:257.
8. Kinger A, Kawaatra M, Chaudhary TS. Case of lingual cysticercosis and review of literature. J Lab Physicians 2012;4:56-8.
9. 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;103:528-33.
10. Sah K, Grover N, Chandra S, Gula S. Oral cysticercosis in a vegetarian female: A diagnostic dilemma. J Oral Maxillofac Pathol 2019;23:289-91.
11. Singh A, Gautam P, Handa AC, Handa KK. Oral cysticercosis: A case series and review of literature. J Maxillofac Surg 2018;76:2572-6.
12. Shafer WG, Hine MK, Levy BM. Mycotic infections of the oral cavity. In: Rajendran R, Sivapathasundharam B, editors. Shafer's textbook of oral pathology. New Delhi, India: Elsevier; 2006. p. 513
13. de Souza PE, Barreto DC, Fonseca LM, de Paula AM, Silva EC, Gomes RS. Cysticercosis of the oral cavity: Report of seven cases. Oral Dis 2000;6:253-5.
14. Krishnamoorthy B, Suma GN, Dhillon M, Srivastava S, Sharma ML, Malik SS. Encysted Taenia solium larva of oral cavity: Case report with review of literature. Contemp Clin Dent 2012;3:S228-32.
15. Jay A, Dhandha J, Chiodini PL, Woodrow CJ, Farthing PM, Evans J, et al. Oral cysticercosis. Br J Oral Maxillofac Surg 2007;45:331-4.
16. Deshmukh A, Avadhani A, Tupkari J, Sardar M. Cysticercosis of the upper lip. J Oral Maxillofac Pathol 2011;15:219-22.
17. Dhaff GA, Al-Hadi AA. Oral cysticercosis: A case report. Saudi Dental J 2000;2013:100-2.
18. Mittal A, Das D, Iyer N, Nagaraj J, Gupta M. Masseter cysticercosis - a rare case diagnosed on ultrasound. Dentomaxillofac Radiol 2008;37:1113-6.