CASE REPORT

Oral cysticercosis: a clinical dilemma

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SUMMARY

Cysticercosis is a potentially fatal parasitic disease caused by *cysticercus cellulosae*, the larval stage of *Taenia solium*. Oral cysticercosis is a rare entity and represents difficulty in clinical diagnosis. This article reports two cases of oral cysticercosis involving buccal and labial mucosa. Both the cases presented with solitary, nodular swelling that had been clinically diagnosed as a mucocele. Histopathology of excisional biopsy revealed it to be cysticercosis. Single, cystic nodular swelling of oral cavity may be the only evidence of cysticercosis and may present first to dentist. These cases emphasise the role of dentist and thorough histopathological examination in the early diagnosis of disease that can prevent potential systemic complication.

BACKGROUND

Cysticercosis is an infection with the larval stage of *Taenia solium*.^{1–3} *Cysticercus cellulosae*, the larval stage of *T solium* resides in muscles and other tissues in pigs that serve as an intermediate host.² Ingestion of inadequately cooked pork containing cysticercus leads to infection in human beings, the definitive host.⁴ Acquisition of cysticercosis can also be followed by faeco-oral route which explains cases of cysticercosis in vegetarians.⁴

Infestation by *T solium* is common in areas where a pig breeding is not controlled and sanitation is inadequate.⁵ Cysticercosis in human is common in cerebral tissue, subcutaneous tissue, muscle and the eye.^{3 6 7} Cysticercosis in the oral tissue such as tongue, labial mucosa, buccal mucosa and the floor of mouth have also been reported; however, incidence is very rare and correct and precise diagnosis is infrequently established.^{6 8 9} We report two cases of oral cysticercosis that emphasises the importance of routine microscopic examination in diagnosis of innocuous lesions of oral cavity.

CASE PRESENTATION

This paper presents two cases of cysticercosis of oral cavity reported to the department. Clinical presentation for both the cases was of diffuse, non-tender, soft to fluctuant swelling. Case 1 showed involvement of right cheek in lower third of face in line with outer canthus of the right eye (figure 1); however, intraorally overlying mucosa was intact and normal in colour. While case 2 revealed involvement of right labial mucosa in relation to maxillary right anterior region (figure 2). No sub-mandibular or cervical lymphadenopathy was detected in both the cases. General, systemic and hard tissue examination of dental structure was

Figure 1 Case 1 showing extraoral submucosal nodule involving right cheek.

within normal limits. Both the patients were vegetarian but not related with each other.

The age, sex, site, duration, number and investigations of two cases are shown in table 1.

Fine needle aspiration cytology (FNAC) in both cases revealed approximately few drops to 0.5 ml of clear watery fluid. In both the cases provisional diagnosis of mucocele was considered. Differential diagnosis included lipoma, fibroma, benign salivary gland tumours and parasitic cysts, for example, cysticercosis cellulosae or hydatid cyst. To rule out parasitic cysts stool examination was carried out. Case 1 was positive for proglottids and eggs in stool examination (figures 3 and 4) while case 2 was negative. Histopathological examination of excisional biopsy specimen revealed external dense, fibrous capsule derived from host tissue surrounding a cystic cavity which contained larval stage of *T solium*-cysticercus cellulosae (figure 5). The

Figure 2 Case 2 showing intraoral submucosal nodule on right gingival mucosa of maxillary labial anterior region.

To cite: Wanjari SP, Patidar KA, Parwani RN, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2012-007482

Table 1 Clinical features of oral cysticercosis in the present series

No.	Age	Sex	Site	Duration	Number	Size	FNAC	Stool examination
1	53 years	M	Rt cheek, lower third of face	5–6 months	Solitary	3×3 mm	1/2 ml of clear watery fluid	Positive
2	10 years	M	Rt labial mucosa, anterior region	10–12 months	Solitary	3×2 cm	few drops of clear watery fluid	Negative

FNAC, fine needle aspiration cytology; M, male; Rt, right.

capsule showed intense inflammatory infiltrate consisting of plasma cells and few eosinophils. The larva consisted of a scolex, where suckers and duct-like invaginated segment could be identified at the caudal end. A digitiform coating of homogenous eosinophilic membrane was evident lining the cystic area (figure 6). A final diagnosis of oral cysticercosis was made. MRI and CT scan were negative which ruled out neurocysticercosis (NCC).

INVESTIGATIONS

The following investigations were performed:

- ▶ Routine haemogram, which was within normal limits in both cases.
- ▶ FNAC from both the lesions revealed few drops to 0.5 ml clear watery fluid.
- ▶ Stool examination was positive for proglottids and eggs in one case.
- ▶ Excisional biopsy of both the cases confirmed it to be oral cysticercosis.
- ▶ CT scan and MRI were negative in both the cases indicating other areas to be free from disease.

DIFFERENTIAL DIAGNOSIS

- ▶ Lipoma
- ▶ Fibroma
- ▶ Pyogenic granuloma
- ▶ Benign salivary gland neoplasm
- ▶ Parasitic cysts—cysticercosis cellulosa/hydatid cyst

TREATMENT

Patients in the present report were asymptomatic except for awareness of swelling. Surgical enucleation was performed in both the cases. Case 1, who was showing positive stool examination, was prescribed with oral corticosteroids and antihelminthic drugs, while case 2 was kept under observation.

Figure 3 Stool examination of case 1 revealed fragmented proglottids of cysticercosis cellulosa (Iodine stain ×600).

OUTCOME AND FOLLOW-UP

Case 1 was reviewed every week for 3 months with stool examination which appeared negative at the end of 1 month. Thereafter he was reviewed every month for another 3 months. While case 2 was reviewed every month for 2 months and thereafter with an interval of 3 months. Periodic follow-up of both the cases for 1 year at the interval of 3 months revealed prognosis to be excellent with no recurrence.

DISCUSSION

Parasitic infections, once restricted to certain parts of the world are becoming worldwide in their distribution as a result of jet age travel.^{9 10} Nevertheless, parasitic oral problems are rare.¹⁰

Oral cysticercosis is one of the rare parasitic infestations.¹¹ Cysticercosis is an infection with *Cysticercus cellulosa*, the larval stage of *T. solium* or pork tape worm.^{1 8 12} *T. solium* passes its life cycle in two hosts, the definitive host is human who harbours adult worm and intermediate host is pig which harbours the larval stage.^{1 13} The adult worm may be ingested in inadequately heated or frozen pork or humans may ingest the cysticerci themselves from infested pork and these develop into the adult worm.^{13 14} This lives attached to the wall of small intestine where it is fully grown and may reach a length upto 7 m. Proglottids are frequently detached from the distal end of the worm and are excreted in the faeces.¹⁵ Each proglottid contains 50 000–60 000 fertile eggs, which remain viable for long time in water, soil and vegetation.^{12 15} These eggs may infect pigs and cycle goes on^{1 16} or alternately human being gets infected by parasite in three ways:

1. Ingestion of food or water contaminated by infected human faeces containing *T. solium* eggs;
2. Oral transmission of eggs via the hands or carriers of adult worm;
3. Internal autoinfection by regurgitation of eggs into the stomach after reverse peristalsis.^{1 8}

Figure 4 Stool examination of case 1 revealed eggs of cysticercosis cellulosa (Iodine stain 600×).

Figure 5 Photomicrograph showing longitudinal section of cysticercosis larva projecting in the lumen from the wall in the form of duct like invagination(D) with scolices(S) and outer fibrous capsule(C) of cyst (H&E stain, $\times 40$).

The larval form penetrates the intestinal mucosa and is then distributed through the blood vessels and lymphatics to all parts of the body where they develop into cysticerci completing the life cycle.^{14 17}

The most serious involvement of cysticercosis is that of central nervous system (CNS) followed by ocular involvement.⁵ However oral and maxillofacial region is not a frequent site of occurrence.⁵ In the present case, the patient complained of asymptomatic nodule on buccal and labial mucosa, respectively. Elias *et al* reported only 65 cases of oral cysticercosis whereas Saran *et al* analysing 120 cases reported a prevalence of 4.2% in the mouth. Both of them found a greater incidence in the tongue followed by labial and buccal mucosa.² Nigam S *et al* (2000) reported three cases each in the buccal mucosa and lip.¹⁴ The most frequently affected age range is believed to be between 20 and 50 years¹⁸ which is almost similar in our case. In the present case, duration varied between 5 and 6 months to 10 and 12 months. In previous published case reports the duration varied between 1 month and 4 years.¹⁸ Mostly in all the reported cases swelling was non-tender with the exception of one case reported by Kinnmen *et al*.¹⁸ Similarly present cases were also asymptomatic except for awareness of swellings.

Figure 6 Photomicrograph showing caudal end of larva lined by cellular, homogeneous eosinophilic membrane (H&E Stain, $100\times$).

Mukesh *et al*¹⁸ reported 9 of 11 as a solitary lesion similar to our report.

The clinical presentation of present case suggested to be mucocoele. The differential diagnosis considered was lipoma, fibroma, pyogenic granuloma, Benign Salivary Gland Neoplasm, for example, pleomorphic adenoma or Parasitic cysts—cysticercosis cellulosa/hydatid cyst.

MRI and CT scan of the brain are the investigations of choice in patients with suspected NCC.⁹ Present cases were negative on CT scan and MRI suggesting no other involvement.

Serological investigations such as enzyme-linked immunosorbent assay (ELISA) and immunoblot assays for the detection of antibodies to *T solium* in serum and cerebrospinal fluid confirm the diagnosis although they are not 100% sensitive.⁴

Although laboratory tests and radiological imaging can be used to help for the diagnosis of cysticercosis but definitive diagnosis can only be confirmed by excisional biopsy and its histological examination.^{6 15} Some other diagnostic tools that must be considered to detect its presence are FNAC and stool examination.¹⁵ Case 1 on parasitological examination of stool revealed *T solium* proglottids and eggs as mentioned by Romero De Leon.^{9 11}

Symptomatic disease usually results from invasion of the CNS, in particular when parenchymal neurocysticerci die years after infection, resulting in strong inflammatory reaction that leads to epileptic seizures. NCC is the most common cause of acquired epilepsy in developing world. Signs of raised intracranial, motor deficits and encephalitis are other manifestations of cerebral involvement.^{4 19}

NCC has been implicated in the pathogenesis of some types of human cancer such as cerebral gliomas, astrocytoma, etc.¹⁵ Oncogenic effect of cysticerci could be explained by transfer of genetic material from the parasites to the host cell. Persistent antigenic stimulation increases the frequency of aberrations in chromosomes 7 and 14, may lead to Hodgkin disease, multiple myeloma, etc. Persistent chronic inflammation surrounding dying parasites may inhibit the immunological surveillance against cancer leading to malignant transformation of adjacent cells.¹⁵

Treatment of cysticercosis may be unnecessary in asymptomatic patients. An accessible solitary symptomatic nodule is usually removed surgically.¹⁸ Anthelmintics like praziquantel and albendazole may be reserved for disseminated cysticercosis, NCC, etc. In the present case anthelmintics and corticosteroids were prescribed to case 1 who showed positive *T solium* proglottids and eggs in stool. As the patients in the present case were asymptomatic surgical enucleation was performed with periodic follow-up.

Learning point

While diagnosing innocuous nodular swelling in the oral cavity, cysticercosis should also be considered in the differential diagnosis. Transmitted via the orofecal route, cysticercosis is potentially eradicable. An efficient eradication programme which include, pork inspection, properly washed vegetables and efficient hand washing before meal and food preparation is the fundamental factor in basic sanitation to extinguish the infection. Our cases could add to the series of rare manifestation of cysticercosis, in the form of oral involvement.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- Patel K, Shah M, Patel B, *et al.* Subcutaneous oral cysticercosis. *Natl J Community Med* 2011;2:311–13.
- Ribeiro AC, Luvizotto MC, Soubhia AM, *et al.* Oral cysticercosis: case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007;104:e56–8.
- Mohan H. Infectious and parasitic diseases. In: Mohan P, Mohan T, eds. *Textbook of pathology*. New Delhi, India: Jaypee Brothers Medical Publishers Ltd, 2005:195–6.
- Jay A, Dhanda J, Peter L, *et al.* Short communication-oral cysticercosis. *Br J Oral Maxillofac Surg* 2007;45:331–4.
- Elias FM, Martins MT, Foronda R, *et al.* Oral cysticercosis: case report and review of the literature. *Rev Inst Med Trop S Paulo* 2005;47:95–8.
- Junior HM, Melo Filho MR, Nogueira dos Santos LA. Oral cysticercosis. *Braz J Oral Sci* 2006;5:1109–11.
- Lee KH, Cepeda L, Miller M, *et al.* Mucocoeles not-oral cysticercosis and minor salivary gland adenocarcinoma: two case reports. *Dermatol Online J* 2009;15:5.
- Mukheh S, Kacker SK, Kapilla K. Cysticercosis of the oral cavity—a clinico pathological study of ten and a half years. *JIDA* 1986;58:257–9.
- Dhaif GA, Al-Hadi AA. Oral cysticercosis: a case report. *Saudi Dental J* 2000;2:100–2.
- Shafer WG, Hine MK, Levy BM. Mycotic infections of the oral cavity. In: Rajendran R, Sivapathasundharam B, eds. *Shafer's textbook of oral pathology*. New Delhi, India: Elsevier, 2006:513.
- Romero DL, Aguirre A, Monterrey NL. Oral cysticercosis. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1995;79:572–7.
- Kinger A, Kawatra M, Choudhary TS. Case of lingual cysticercosis and review of literature. *J Lab Phys* 2012;4:56–8.
- Kumar V, Abbas AK, Fausto N. Infectious diseases. In: Sklar E, eds. *Robbins and Cotran pathologic basis of diseases*. Philadelphia, PA: Saunders Elsevier, 2004:406–7.
- Nigam S, Singh T, Mishra A, *et al.* Oral Cysticercosis: report of six cases. *Head and Neck* 2001;23:497–9.
- Delgado-Azner WA, Mosqueda-Taylor A, Carlos-Bregni R, *et al.* Oral cysticercosis: a collaborative study of 16 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007;103:528–33.
- Deshmukh A, Avadhani A, Tupkari JV, *et al.* Cysticercosis of the upper lip. *JOMFP* 2011;15:219–22.
- Rubin E. Infectious and parasitic diseases. In: Gorstein F, Rubin R, Schwartzing R, *et al.* eds. *Rubin's pathology: clinicopathologic foundations of medicine*. Philadelphia, PA: Lippincott William and Wilkins, 2004:469–70.
- Saran RK, Rattan V, Rajwanshi A. Cysticercosis of the oral cavity: report of five cases and a review of literature. *Int J Paediatr Dent* 1998;8:273–8.
- Gutierrez Y, Smith JH. Metazoan diseases. In: Damjanov J, Linder J, eds. *Anderson's pathology*. Missouri, StLouis: Mosby, 1996:1034–5.

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