

Orofacial Cysticercosis: A Review

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Abstract

Cysticercosis is a common healthcare problem, especially in developing countries. Orofacial presentation of the disease is rare. It usually manifest as an asymptomatic nodular swelling that is difficult to differentiate clinically, from other orofacial swellings. Diagnosis of cysticercosis is usually not possible clinically owing to its rarity and asymptomatic presentation in orofacial region. Ultrasonography (USG) is the initial and most reliable diagnostic modality for cysticercosis. This review discusses the various oral manifestations, differentials and investigations for oral cysticercosis.

Keywords: Orofacial; Cysticercosis; Nodules; Echogenic foc

Introduction

Cysticercosis was first described in pigs by Aristophanes and Aristotle in 3rd century BC. Latter it was noticed in human by Parunoli in 1550 [1]. Cysticercosis is an infection caused in humans by the larval form of the pork tapeworm *T. solium* (i.e. *Cysticercus cellulosae*) [2]. *T. solium* exists worldwide but is most prevalent in Latin America, sub-Saharan Africa, China, southern and Southeast Asia, and Eastern Europe [3]. The pork tapeworm eggs, when ingested through the contaminated food, water or dirty hands leads to the parasitic infestation [4].

Life Cycle of Cysticercosis

Life cycle of *Taenia solium* comprises two natural hosts, humans as the definite and swine as the intermediate host. When pork containing cysticerci is consumed by the humans, the larva form enters the small intestine and develops into an adult worm. The adult worm attaches itself to the intestinal mucosa by scolex equipped with four lateral suckers and a rostellum, which bears 25-50 hooklets. Aided by their hooklets, the oncospheres cross the intestinal wall and local venules, enter systemic circulation and are carried to different organs of the host (skeletal muscles, central nervous system, subcutaneous tissue, eye, etc.) [1].

Route of Infection

The route of entry is predominantly oral. The eggs of *Taenia* enter the gastrointestinal tract of the humans through consumption of contaminated water and improperly cleaned raw fruits and vegetables or by the process of autoinfection due to reverse peristalsis in the people infected with its adult form and harboring eggs in the stomach [5].

Manifestations in Human

Living larvae can easily evade immune recognition and may not elicit inflammatory reaction. When these larvae die, a vigorous granulomatous inflammatory response is induced and this may be

responsible for producing the clinical symptoms [2]. Generalized symptoms include headache, fever and myalgia. Multiple tissues of the body may be involved but, the most serious involvement is that of the central nervous system, followed by ocular involvement. Clinical spectra of the disease depend upon the localization of the cyst. Literature review reveals that neurocysticercosis (cyst lodged in the CNS) is the commonest form of cysticercosis, with the brain parenchyma most commonly involved. Ophthalmic cysticercosis (intraocular) manifests symptoms like proptosis, diplopia, and loss of vision while extraocular cyst resembles slow growing tumour or nodule with focal inflammation. The larva has a strong affinity for muscular tissue. Cysts in muscles may manifest as muscular pain, weakness or pseudohypertrophy. Subcutaneous cysticercosis is frequently asymptomatic but may manifest as palpable nodules [1].

The most common site for occurrence of subcutaneous nodule is trunk, followed by upper arm, eyes, neck, tongue, face and breast [6].

Oral Manifestations

In the maxillofacial region, the locations of calcified cysticerci present on muscles of mastication and facial expression, the suprahyoid muscle, and the posterior cervical as well as the tongue, buccal mucosa, or lip [7]. But, despite the abundance of muscular tissue in the oral and maxillofacial region, this is not a frequent site of occurrence [8]. Whenever, orofacial cysticercosis is present, multiple foci may be involved. So, every case of oral cysticercosis should be thoroughly investigated for presence of multiple foci.

The orofacial lesions usually present as insidious, benign, asymptomatic, nodular swellings that are well tolerated by the patients. Rarely, these may be painful when the larva dies and there is a leakage of fluid from the cystic cavity. Alternatively, when the implanted larvae die as a result of immunological defense of the host, the cystic fluid may become turbid due to signs of hyaline degeneration of the scolex (colloidal stage). This is followed by the calcification of the larvae and thickening of the capsule (granular stage). The remnants of the dead larvae may become mineralized (calcified stage) and appear radiographically as calcified nodules [5].

Imaging

Diagnosis of cysticercosis is usually not possible clinically owing to its rarity and asymptomatic presentation in orofacial region. The initial manifestation of cysticercosis is usually a soft tissue swelling that makes Ultrasonography (USG) as the initial and most reliable diagnostic modality for these swellings. Vijayaraghavan SB described four different pattern of muscular cysticercosis on USG. The first pattern is a cystic cavity with an inflammatory response around it, as a result of the death of the larva. The second ultrasonographic pattern is an irregularly defined cystic cavity with very minimal fluid on one side, indicating the leakage of fluid. An eccentric echogenic mass within the cyst is not seen. The third appearance is a large irregular cyst within the muscle with an eccentric placed echogenic foci within the collection. This appearance is similar to an intramuscular abscess. The fourth sonographic appearance is that of calcified cysticercosis. With the use of high resolution modern USG machines, these appearances on USG may be considered pathognomic for confirmed diagnosis of Cysticercosis [5].

MRI is also being helpful in diagnosis of soft tissue cysticercosis. Cysticercosis is seen as a cystic lesion that appears hyperintense on T2W and hypointense on T1W images [5]. MRI is also considered as the best tool for the investigation of degenerating and innocuous (viable) cysticerci in nervous system, while Computed Tomography (CT) has been considered best for calcified lesions. The additional benefit of prescribing MRI is that the different stages of the parasite can be identified in contrast to CT [1].

Plain radiography is rarely helpful to visualize cysticerci in the active phase, but in the chronic cases, the calcified lesions can be identified. Calcified intramuscular cysticerci appear as small elliptical lesions in the soft tissue parallel to muscle fibres [5].

Other Investigations

Parasitological examination is more reliable in revealing *T. solium* eggs in the collected stool sample [7]. Body fluids like sera, cerebrospinal fluid and saliva, can be used for Immunodetection of cysticercosis by ELISA (enzyme-linked immunosorbent) assay or EITB (enzyme-linked immunoelectro transfer blot). However, this investigation may give false positive results in the individuals living in an endemic area, in patients with solitary lesions and old calcified disease [5,6]. The specificity and sensitivity of EITB are superior to ELISA for the diagnosis of cysticercosis [7].

Although excisional biopsy has been considered as the only definitive diagnostic procedure to demonstrate the presence of the parasite, there are some other diagnostic tools that must be considered to detect its presence in the diverse tissues that may be affected, including the oral region [9]. Saran et al. advocated the use of fine-needle aspiration cytology for identification of the tegument layer of the larva. Histopathological examination formulates a definitive diagnosis of cysticercosis by the detecting the organism within the cystic spaces [10].

Previous Studies on Oral Cysticercosis

In various reviews on the topic, it was observed that there is almost equal distribution between the genders, with the mean age range of second decade and most common site involvement of tongue.

The findings of previous studies have been summarized in Table 1 [5,9,11-14].

Review/ Study	Total no. of cases	Gender distribution	Age distribution	Site distribution
Ambika et al. (2016)	16 cases	68.75% were females and 31.25% males	10-55 years with a mean of 25.69 years	Masseteric region (37.5%), zygomatic region (18.75%), infra-orbital (12.5%), tongue (12.5%) and temporal region (12.5%)
Wilson A (2007)	97 cases	50 males, 47 females	3-70 years, Mean- 23.9 years	Tongue (44 cases), buccal mucosa (24 cases), lower lip (19 cases), upper lip (8 cases)
Fernando et al. (2005)	65 cases	1:1	3-70 years Mean 23.7 years	Tongue (42.15%), lips (26.15), buccal mucosa (18.9%).
Nigam S et al. (2001)	6 cases	-	-	3 in lips, 3 in buccal mucosa
De Souza PE et al. (2000)	7 cases	M/F=0.4	12-56 years. Mean 25 years	3 in tongue, 2 in lip, 1 in floor of mouth, 1 in retromolar area
Saran RK et al. (1998)	5 cases	M/F=4:1	3-12 years	4 in tongue, 1 in buccal mucosa
Timosca G, Gavriilita L (1974)	5 cases		9-30 years	2 in submandibular region, 2 in cheek, 1 in lip and chin

Table 1: Findings of previous studies on oral cysticercosis

Management

Treatment includes medical and surgical modalities. The management of cysticercosis is also site-dependant. Drugs such as albendazole (given in a dose of 15 mg/kg/day for 28 days) or praziquantel (administered in a dose of 50 mg/kg/day in three doses for 15 days) are used as effective antihelminthics for cysticercosis.

Praziquantel, however, has no effect on calcified parasites. Low dose steroid is sometimes given along with cysticidal drugs to prevent inflammatory reaction following death of larva [5].

Conclusion

With the use of high resolution modern USG machines, it may be considered for confirmed diagnosis of Cysticercosis. The prognosis of the disease is excellent if timely and adequate medical treatment is provided. Death from cysticercosis is rare, but it can occur. Cysticercosis can be easily prevented and eradicated. To achieve this goal, intensive health care programs, mass education and adequate medical facilities need to be undertaken in the endemic areas.

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