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## **Oral Cysticercosis: A Diagnostic Rarity Oral Cysticercosis**

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### **Abstract**

Helminth parasites comprise a complex and diverse group of organisms frequently found in developing countries like India, sub-Saharan Africa, Eastern Europe and Latin America. The species of Echinococcus and Taenia cause innocuous to potentially fatal disease. Cysticercosis is caused as a result of infestation with the larval stage of the pig tape worm Taenia solium. The larval stage of the pork tapeworm infests the human central nervous system causing neuro-cysticercosis, one of the leading causes of epileptic seizures in developing and under-developed countries. Extra-neural and ophthalmic organs are other harbors of the pork parasite. Therapeutic measures include drugs preferably Praziquantel and Albendazole, steroids, prophylactic anti-parasitic treatment of endemic populations and improving sanitization and vaccination. Although oral cysticercosis is a rare finding and presents as an asymptomatic nodule, generally affecting the tongue followed by buccal mucosa and lower lip, multiple oral involvements have been reported in few cases. This study reports a case of oral cysticercosis in the lower labial mucosa. Excisional biopsy was performed and histopathology was suggestive of larval form of Taenia solium.

**Introduction:** The twenty first century is a period of random change and evolution but still changes in advances in public health, sanitation, vaccination strategies and improvement in medical facilities are stagnant. Taenia solium infection and associated disease occurs only in the human host and are endemic

in under-developed countries.<sup>1</sup> Cysticercosis is more prevalent in developing countries, mainly the sub-Asian continent, Africa, Peru and Mexico. In non-endemic areas of UK, Scandinavian region and USA, cysticercosis is associated with increased immigration and travel related tapeworm

infestation.<sup>2</sup> Taeniasis (adult worm) occurs only in human host after ingestion of undercooked pork infected with cysticerci. The worm attaches strongly to the mucosa of upper small intestine bringing mild inflammation at implantation site although without significant damage to the intestine and characterized by very few to nil symptoms, if at all seen.<sup>3</sup> Humans are either the definitive or intermediate host of the adult tape worm *Taenia solium* and the life cycle of this platyhelminthes requires various hosts that harbor the proglottids (eggs), oncospheres, larve and adult forms. Dissemination of the oncospheres occurs through vascular or lymphatic circulation and develops into cysticerci.<sup>2</sup> A characteristic difference is observed in the motility of the organisms of *T. solium* where tapeworm segments are not noticed in stools where as in patients infested with *T. saginata*, passage of proglottids is seen which are motile, numerous and larger than *T. solium*.<sup>4</sup> Oral involvement of cysticercosis is unusual and the exact incidence is unknown thereby making a precise clinical diagnosis not readily established. Thus we report a case of oral cysticercosis for a precise understanding of this unusual parasite.

**Case report:** A 21 year old Indian male with average health attended the oral clinics for management of a solitary painless swelling on the lower labial mucosa present since 5-6 months. [Figure.1] Medical and dental history was not significant. General and systemic examination did not reveal any sign of a disseminated or systemic disease. The patient was a non-vegetarian in diet and belonged to low socio-economic strata and dwelled in non-sanitized rural surroundings. The lesion increased gradually to attain the

present size, approximately 1- 1.5 cm, with no history of similar swelling or trauma in the past. The swelling was freely movable with no discharge or change in the size during lip movements. On examination, the solitary swelling was round to oval in shape, soft, elastic, and fluctuant and translucent in appearance with well-demarcated borders and a smooth surface. Based on the clinical appearance, a provisional diagnosis of a Mucocoele was arrived at although fibroma, lipoma, haemangioma and glandular entities were considered in the differential diagnoses. Blood chemistry revealed components within normal references. The lesion was enucleated and a smooth well-encapsulated, pearly white fibrous, globular tissue was obtained and sent for histo-pathological analysis. [Figure.2]

**Histopathology:** The Hematoxylin and Eosin stained sections on light microscopic examination revealed part of the cystic lumen with a delicate double-layered membrane containing an outer acellular and an inner sparsely cellular layer loosely attached to the fibrous capsule; [Figure.3] at the cranial end, the shrunken larve containing scolex with suckers and crown of hooks was observed and at the caudal end, inverted papillary spaces were observed confirming the presence of larval stage of *Taenia solium*. [Figure.4] The inner aspect of the fibrous capsule consisted of dense aggregation of eosinophils and polymorphonuclear leucocytes. [Figure.5] Thus, a final diagnosis of cysticercosis was established and the patient was referred for a through medical investigation. No signs and symptoms of systemic or disseminated disease were although detected. The patient is still for regular follow up since two years and is

without any neurological, muscular and orbital symptoms.

**Discussion:** Cysticercosis develops when humans accidentally ingest eggs of *Taenia solium* via contaminated food, polluted water or unclean hands.<sup>5</sup> It is highly common in areas of poor sanitization where animals and humans are in close proximity and were rearing domestic animals especially pigs at rural households and not particularly taken care of unlike raised and precisely maintained industrial herds that are subject to strict sanitary control.<sup>6</sup> Humans also acquire cysticercosis through faeco-oral contamination with *T. solium* eggs from tape worm carriers.<sup>3, 7</sup> Thus, vegetarians and non-pork eaters are also prone to acquire cysticercosis.<sup>8</sup> Contaminated water, flies and aerosols are other means of indirect transmission and do play a vital role in spread of the infection.<sup>9</sup> The infective oncospheres are eventually liberated by gastric acids and enter the blood stream by crossing the bowel wall and are carried to the harboring tissues.<sup>10</sup> The tape worms in their mammalian host then elicit immune response dominated and coordinated by T helper 2 (Th2) cells and their associated cytokines.<sup>1</sup> The clinical manifestations are named in accordance to the specific visceral involvement as neuro-cysticercosis, a pleomorphic clinical disorder affecting the central nervous system and eliciting inflammatory changes, being protected by the blood brain barrier and active immune evasion mechanisms by the cysterci.<sup>11</sup> They cause symptoms in such cases by blocking the circulation of cerebrospinal fluid and most of the results are seen as an effect of inflammatory changes leading to cystic degeneration.<sup>10</sup> Epileptic seizures are the

most common presentation of neuro-cysticercosis occurring approximately in 50-80% of patients especially in endemic regions where sudden onset of seizures in a healthy individual is strongly suggestive of neurocysticercosis.<sup>12</sup> Extra-neural cysticercosis outside the nervous system does not cause major symptoms. Subcutaneous cysticercosis presents as small, painless movable nodules particularly noticed on arms or chest while muscular cysticercosis seen as ellipsoidal calcifications across muscle bundles in thighs or arms. Majority of the patients with neuro-cysticercosis show muscular calcifications while cardiac cysticercosis is usually asymptomatic.<sup>13</sup> Ophthalmic cysticercosis occurs in approximately 1-3% of all infections. *T. solium* is seen as the most common intra-orbital parasite with visual disturbances and proptosis being some of the most common symptoms noted. Cysts are found intra-ocularly and are freely flowing in the vitreous humor or in the sub-retinal space, anterior chamber or may affect the extra-ocular muscles also in rare of the cases.<sup>14</sup> Lesions may be asymptomatic when involving the oral cavity. It was found that the lesion on the tongue could interfere with movement, causing discomfort during speaking and eating.<sup>15</sup> Oral cysticercosis indicates disseminated infestation although systemic complications are not demonstrated in most of the patients with oral lesions. This may be due to the fact that generally disseminated larvae are located in the deeper tissues and may remain alive throughout the life of the host without giving clinical manifestations. Despite of the lesser degree of severity, it is however important to constantly evaluate the medical status and maintain

judicious follow-up in such patients.<sup>16</sup>The oral lesions in these patients may present a variety of benign lesions. Tongue lesions may be provisionally diagnosed as neurofibromas, lipomas, leiomyomas, schwannomas, fibromas, lymphangiomas or haemangiomas and several others. Lesions on the buccal mucosa may be diagnosed as fibromas, mucoceles and a plethora of other benign mesenchymal tumors and even benign salivary gland neoplasms cannot be left misdiagnosed. Several laboratory tests and radiologic imaging techniques have been implemented to confirm a definite diagnosis of cysticercosis however histopathology and advanced molecular techniques have emerged as tools for conformational diagnosis. Fine Needle Aspiration Cytology also remains a reliable conventional procedure for pre-operative diagnosis of cysticercosis in treatment planning and efficient management.<sup>16,17</sup> Cytology usually consists of multiple well-vascularized fragments and dense diffuse infiltration of polymorphous lymphoid cells, plasma cells, macrophages, and epithelial histiocytes with inflammatory foreign body granulomatous reactions. At times, segmental material with nuclear bodies and refractile elements may also be noticed.<sup>18</sup> Serology and neuro-imaging and eosinophilia contributes majority in neuro-cysticercosis by sampling of cerebrospinal fluid.<sup>19</sup> The enzyme linked immune-blot assay contributes to have sensitivity of 98% and specificity of 100% and performs better than ELISA in clinical settings and serves as an excellent modality in diagnosis and treatment planning.<sup>20</sup> Computed imaging procedures which include CT and MRI are vaguely used presently leaving behind arteriography and pneumo-

encephalography, MRI being the most accurate technique to assess the degree of systemic infestation, location and evolutionary stage of the disease.<sup>19</sup>A set of objective diagnostic criteria has also been proposed which includes absolute and minor criteria. Oral cysticercosis contributes into minor criteria for diagnosis in combination with neuro-imaging and compatible clinical manifestations although its significance in diagnosis cannot be underestimated in addition to its contribution for a continuous follow-up of the patient for possible future systemic or disseminated disease.<sup>21</sup>

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Figure.1 A 21 year old Indian male with average health presenting with a solitary painless swelling on the lower labial mucosa

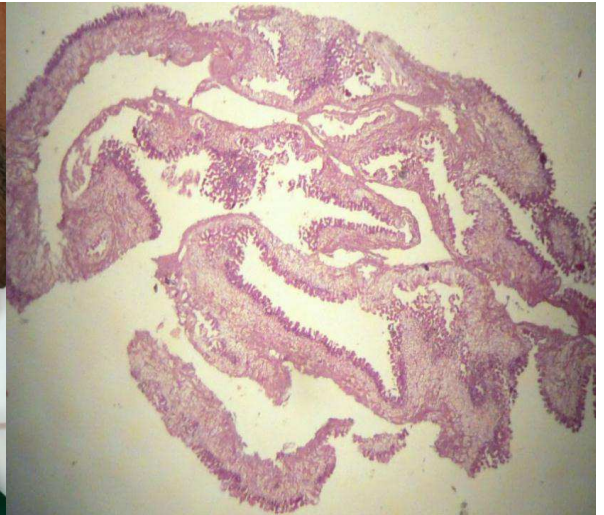


Figure.3 Double layered eosinophilic membrane consisting of outer acellular layer(o) and an inner sparsely cellular layer(i) ( H & E,original magnification  $\times 400$  )



Figure.2 The enucleated lesion appearing as a smooth well-encapsulated, pearly white fibrous, globular mass

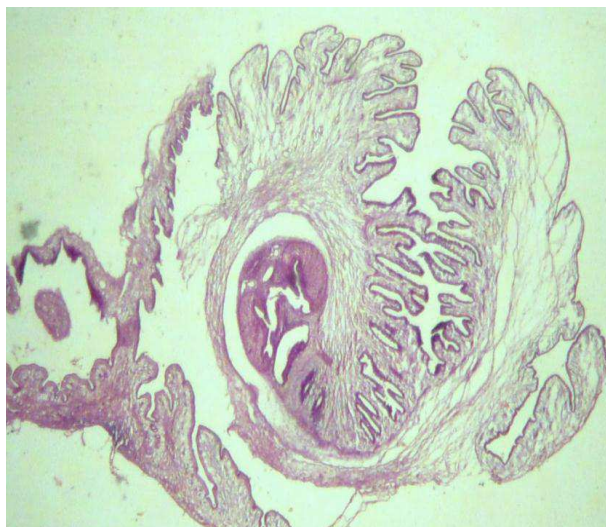


Figure.4 Invaginated segments of the caudal part of the larval body showing papillary projections and a part of the cyst wall ( H & E,original magnification  $\times 100$  )

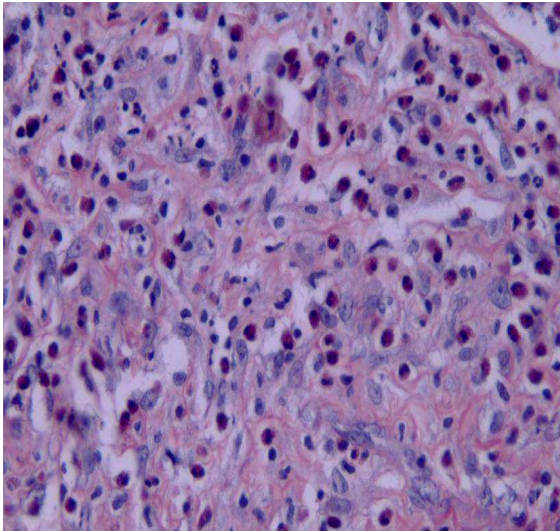


Figure.5 Eosinophilic infiltration along with polymorphonuclear leucocytes in the capsule ( H & E,original magnification  $\times 40$  )