Case-report

SUBCUTANEOUS ORAL CYSTICERCOSIS

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INTRODUCTION

Cysticercosis is an infection with the larval stage of Taenia solium. Infestation by Taenia solium is common in area where pig bredding is not controlled and sanitation is inadequate. Latin America, Southern Africa, India, Southeast Asia and Europe are the most frequent locations of occurrence. The larval form of cyst is commonly seen in the brain, meninges and eyes. The remainder are located in the muscles, heart, lungs and peritoneum. Cases in the maxillofacial region including oral and cheek muscles, are rarely reported.

We present a case of cysticercosis as a solitary cystic nodule in the oral cavity of a forty year male, a very rare occurrence. Very few cases have been reported in the world literature. This is therefore a diagnostic and therapeutic dilemma for clinicians. Solitary cystic nodular swelling inside the lower lip is usually not suspected clinically for cysticercosis. The diagnosis is made on histopathological examination.

The ensuing clinical disorder is named after the name given to the organism at this larval stage, cysticercosis cellulosae, larvae of pork tapeworm Taneia Solium.

CASE HISTORY

A forty year old non vegetarian male presented with a painless solitary cystic nodular swelling near the left corner of inner aspect of lower lip. The nodule was gradually increasing in size and had lately caused discomfort while eating and talking. Examination revealed a 1cm x 0.5 cm well defined , smooth, moveable, non tender swelling at left corner of inner side of lower lip. General and systemic examination was within normal limits. The swelling was excised under local anesthesia at private surgical hospital. On excision it was suspected of a a retetion mucocel and sent for histopathological examination at surgical pathology department, Shree Krishna Hospital, Karamsad.

Grossly it was small, soft and cystic, containing fluid. Microscopic examination revealed a capsule of fibrous connective tissue surrounding a cystic cavity, which contained the cysticercosis cellulosae, Taenia solium larval form.



Figure – 1: Photomicrograph of excised cyst show cystic space with cysticercus larva projecting in lumen from the wall, with scolices and outer fibrous wall of cyst. (H and E, x40)

The capsule showed intense inflammatory infiltrate, consisting mainly of lymphocytes and plasma cells, eosinophils and giant cell reaction. The larva consisted of a scolex, where a sucker and a duct-like invaginated segment- the caudal end could be identified. Both larva and cystic structure were lined by a homogenous eosinophilic membrane.



Figure – II : Photomicrograph show cysticercus larva with scolices of Taenia.(H and E , x100)

The postoperative period was uneventful. The patient was being followed up regularly and was monitored for any neurological or ophthalmic signs and symptoms. He was given Albendazole 200 mg TDS for thirty days.

DISCUSSION

Cysticercosis is an infection with cysticercus cellulosae, the larval stage of Taenia solium or pork tape warm. Taenia solium passes its life cycle in two hosts. The definitive host is human who harbours the adult worm and intermediate host is pig which harbours the larval stage. The adult worm lives in the small intestine of man. Usually one adult worm is present which lives for years. It is about three meters long with 1000 proglotids. The gravid segments have about 50,000 eggs in each segment.¹ The worm sheds gravid segments laden with eggs in the stool which infect pigs. On reaching the alimentary canal of the intermediate host these eggs rupture and oncospheres are liberated. They penetrate the gut wall and reach the systemic circulation and are lodged in the different organs and muscles of the intermediate host.1

Here, they develop into larvae referred to as cystecercosis cellulosae. Human beings may accidentally or incidentally become the host of

parasite in three ways : 1. Ingestion of food or water contaminated by infected human feces containing Taenia Solium eggs. 2.Oral transmission of eggs via the hands or carriers of the adult worm; and 3.Internal autoinfection by regurgitation of eggs into the stomach after reverse peristalsis. These are partially digested in the stomach, evolving to oncospheres and subsequently penetrating the small intestinal mucosa to disseminate throughout the body via arteriovenous channels and lymphatics, frequently encysting in subcutaneous tissue; striated muscles, brain and ocular tissue.

Clinical manifestations of intestinal infection by Taenia solium could be asymptomatic or may present with epigastric pain, nausea and loose motions. In cysticersosis manifestations are different and depends on the location of cysticercosis in the body, not only this but also the number of cysticercosis at a particular site and the associated inflammatory response or scarring decides the clinical presentation.

In 87% of cases cysticercosis presents as solitary lesion. Presentation as subcutaneous nodule on trunk, upper arm, eyes, neck, tongue, face and breast has been reported in this order of frequency. Neurocysticercosis is most often presented with seizures and may be associated with subcutaneous nodules comprised of extraneural cysts. Disseminated cysticercosis with huge muscle hypertrophy reported by Bandyopadhyay D.2 Oral cysticercosis can be another component of disseminated cysticercosis. Dixon and Lipscomb examined 450 cases of cysticercosi and found oral involvement is only 1.8% of the cases.³

Our case is one more addition to this series of rare manifestation of cysticercosis. In spite of the abundance of muscular tissue in the oral and maxilloafacial region, this is not a a frequent site of occurrence for cysticercosis. We have found few cases repoted in the literature.⁴

The most frequently affected age was third to fourth decade with equal distribution between genders, but age was varied from three to seventy years. Authors from the school in Brazil, reported a rare and asymptomatic intraoral nodule in the right buccal mucosa of seven years boy and biopsy of nodule proved the diagnosis as cysticercosis.⁵

Usually, the patient complains of a swelling. Although pain is not a frequent feature, it had been reported in secondarily infected cases. It was suggested that the lesion on inner aspect of lip could interfere with movement of tongue, causing discomfort during speaking and eating.⁶ Differential diagnosis of oral lesion depends on the site involved. In this case the differential diagnosis would be retention mucocele and oral focal mucinosis.

Histopathological examination makes up a diagnosis of cysticercosis by the detection of a cystic space containing the cysticercus cellulosae. The scolex has four suckers and double crown of rostellar hooklets. A duct like invaginated segment, lined by a homogenous anhistic membrane, comprises the caudal end. The eosinophilic membrane that lines the capsule is double layered, consisting of an outer acellular and inner sparsely cellular layer. After a period within three and five years the larva dies and the cyst undergoes calcification.

Currently other diagnostic tool as radiologic imaging and serology can be used. Besides normal radiographic examination, computerized tomography and magnetic resonance are very effective in the detection of cysticerci.⁷

SARAN et al proposed the use of fine needle aspiration cytology, which identifies the tengument layer of the larva, to help the clinician in planning the treatment.⁸ Kamal et al reported ten cases of cysticercosis on cytology by cysticercosis cellulose, hooklet and fragments of wall with inflammation^{.9}

Immunodetection of cysticercosis can be achieved in sera of cerebrospinal fluid and saliva by ELISA (enzyme-linked immunosorbent assay) or EITB (enzyme-linked immune electrotransfer blot),but it is important to consider that individuals living in endemic area may have antilbodies because of an exposure instead of of an established infection.

Every case of oral cysticercosis should be thoroughly investigated to determine the involvement of multiple foci, since there is high incidence of such feature. Drugs as praziquantel and albendazole are potent antihelminthics used in the treatment of cysticercosis.¹⁰

As the patient in the present case had no occurrence of cysticercosis at any other site or any other symptoms, no addition treatment was prescribed except the periodic follow-ups.

CONCLUSION

It is important to consider the diagnosis of cysticercosis in oral solitary cystic nodular lesion presenting in patients living in an endemic area. This case emphasizes the role of the consultant doctor in the detection of disease that can have more serious involvement, as well as the importance of routine histological examination.

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