Case Report

Labial hydatid cyst – A rare entity

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Abstract

Hydatid cyst is a parasitic infestation by tapeworm *Echinococcus*. This endemic disease is prevalent in the Middle East and Southeast Asian populations. This zoonotic disease is mainly transmitted by cattle and dogs. Few studies have been reported in the head-and-neck region. There are very few isolated cases have been reported within the oral cavity region. No case until now has been isolated from the labial mucosa. We now present one such case here.

Keywords: Hydatid cyst, labial mucosa, labial swelling

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Received: 16.02.2019, **Accepted:** 27.06.2019

INTRODUCTION

Hydatid cyst is a parasitic zoonosis otherwise known as human cystic echinococcosis.[1] The Greek word "Hydatid" means "drop of water."[2] This disease is endemic in the areas where dogs and cattle are found in abundance. The incidence of hydatid disease is around 1–200 per 100,000 population in India.[3] The host plays an important role in disease transmission. Humans mostly act as an accidental or end host. *Echinococcus* E. chinococcosis granulosus is the most common etiology behind the formation of hydatid cysts.[1]

Hydatid cyst in human is mostly encountered in the major organs such as lungs and liver (80%), followed by the other parts such as the peritoneum, spleen, kidney, skin and muscles.[4] In the maxillofacial and head-and-neck region, it is reported to as less as 1%–2%.[4-6] This disease is mostly restricted to population having specific food trends and unique lifestyles. The clinical presentation, diagnosis and management of the disease vary according to the localization, duration, shape, size and the number of cysts.

The diagnosis of the hydatid cyst in the oral and maxillofacial region creates accost among the clinical oral pathologists, oral surgeons, microbiologists and general pathologists reporting oral pathology cases due to its rare occurrence. There are very few published literature in the maxillofacial region as per the meticulous research done in various search engines such as PubMed, Cochrane Library and Semantic Scholar. There are a total of six published reports of occurrence in the oral region that involves buccal mucosa, floor of the mouth and tongue and three cases in the maxillary region, i.e., maxillary sinus.[5,7-11] Our case holds a worthy citation as it was localized in the labial mucosa of the maxillary region of the oral cavity in a 62-year-old male.

Access this article online

Quick Response Code:

Website: www.jomfp.in

DOI: 10.4103/jomfp.JOMFP_322_18

How to cite this article: Banerjee A, Elangovan E, Mitra KS, Saha A. Labial hydatid cyst – A rare entity. J Oral Maxillofac Pathol 2019;23:418-21.

CASE REPORT

A 62-year-old male reported to the department of oral pathology and diagnosis after being referred by an oral surgeon. The patient gives a very interesting history of an appearance of an asymptomatic small elevation on the labial mucosa in the maxillary region in relation to 13 and 14. The swelling is recurrent and disappears within 1–2 days. There was no noticeable extraoral swelling, lymphadenopathy or fever associated. Numerous clinical diagnoses of mucocele, epulis and salivary ductal obstruction were rendered, but since the entity appeared for short duration and used to get resolved by itself, it was never indicated for surgical intervention.

On inspection, there was a mild diffuse oval elevation of 0.5 cm × 0.7 cm having normal mucosal color and was barely noticeable [Figure 1a]. There was no sign of inflammation or discharge. On bidigital palpation, the swelling was firm to hard in consistency, nonfixed to the underlying tissue. A provisional diagnosis of minute salivary duct calculi was given and was posted for surgery after hematological examination.

Surgical intervention revealed no association or presence of minor salivary glands or salivary ducts along with the mass. The mass was firm in consistency resembling a small rubber ball measuring 0.5 cm × 0.6 cm, dark creamish black in color surrounded by loose tissue [Figure 1b]. The mass was not fixed to underlying deeper structures. The entire mass was sent for histopathological evaluation. The mass was slightly compressible on bidigital pressure. Grossing of the sample was done [Figure 2]. The entire mass was embedded into one block, and gradual sections of 4 μm were obtained.

The H&E-stained section revealed a trilaminar cystic structure surrounded by the loose fibrous tissue [Figure 3a and b]. The outermost layer or pericyst , intermediate layer was avascular dark eosinophilic refractile layer or exocyst and the inner most layer or endocyst projecting toward the lumen can be appreciated [Figure 4a and b]. The lumen was filled by amorphous, light eosinophilic substance showing branching spaces with small vesicles or brood capsules [Figure 4c]. These brood capsules represent developing daughter cysts filled by clear fluid. However, in this case, hooklets were not appreciated much. The surrounding tissue was loose and fibrous and showed the presence of chronic inflammatory cells. Histopathological interpretation of the hydatid cyst of the oral cavity was rendered.

The patient was sent for further investigations such as ultrasonography (USG) of the upper lip and lower abdomen, chest radiographs and blood investigations which showed no abnormality. The recommended dosage of albendazole was prescribed for 1 month, and the patient was kept under thorough follow-up. There was no such entity found elsewhere in the body and neither any recurrence happened in the same site.

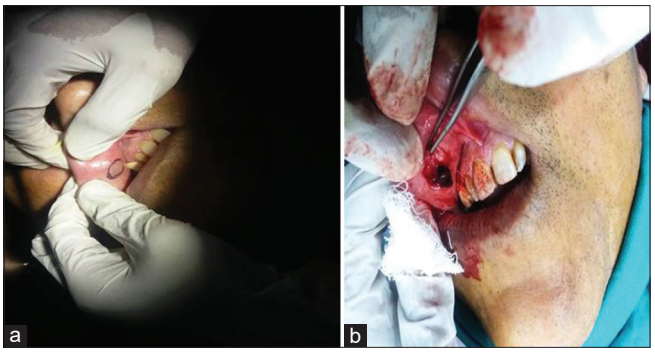

Figure 1: (a) On inspection, a diffuse small swelling on the upper labial mucosa. (b) Excision of the mass

Figure 2: Grossing of the sample, sample along with the peripheral tissue and hemorrhage

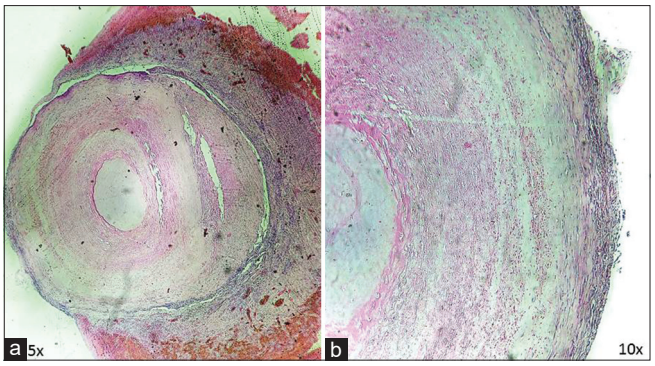

Figure 3: (a) The scanning histological view of the entire specimen showing trilaminar structure. (b) The attached surrounding tissue is loose well-arranged fibrous tissue with chronic inflammatory component

DISCUSSION

Only a few research efforts have been put forth in studying the parasitic cysts of the oral cavity as these entities pose themselves as a dilemma to the diagnosticians.

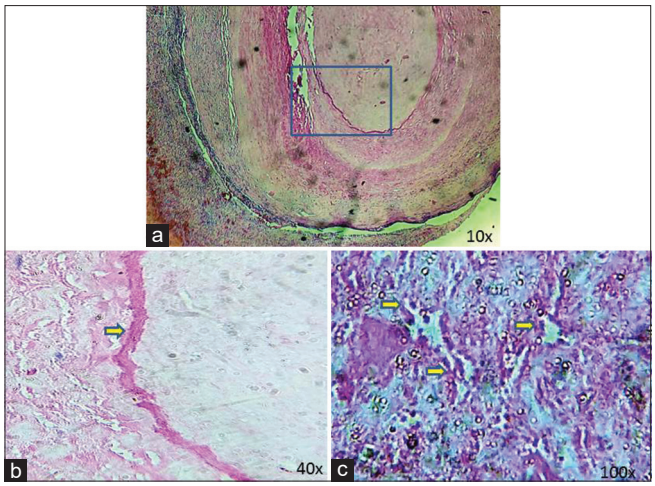

Figure 4: The higher magnification views (a) the pericyst attached to the outer fibrous layer, intermediate layer or exocyst and the innermost layer toward the lumen is endocyst. (b) The intermediate layer depicting refractile eosinophilic substance sandwiched between the outermost layer and the innermost layer and is well demarcated. (c) The lumen depicting small vesicles or brood capsules along with branched lumen space

Echinococcosis is a parasitic infection occurring in humans and other animals such as dogs, sheep, cattle and fox. This parasitic disease is caused by cestodes of genus *Echinococcus*.[12] It is found to occur mostly in the Mediterranean countries, South America, Australia, central parts of Asia and East African countries.[13] The parasitic infestations result in hydatid cyst which is formed by the larval form *Echinococcus granulosus*.[14] The life cycle of this parasite occurs in two hosts, wherein the adult tapeworms reside in the intestine of carnivores, i.e., dogs and foxes, which are the definitive hosts, and the larval forms reside in the intermediate hosts, i.e., man, sheep and cattle. Humans acquire this disease if he/she accidentally ingests the ova passed in the stool of dog or the food contaminated by the ova or by contact with the infected dogs.[12,14] The larva from the ova penetrates the intestinal mucosa and reaches the liver through the portal vein and develops into a cyst. Majority of the larvae affect the liver, followed by lungs, abdominal cavity, kidneys and bone. These cysts are very rarely positioned in the head-and-neck region,[15] and few reported cases in literature showed site predilections such as the right side submandibular region,[16] infratemporal region,[17] tongue[15] and floor of the mouth.[18]

The disease often starts without symptoms and this may last for a year. The signs and symptoms depend on the cyst's location and size. The diagnosis of this condition can be made by histopathology, various imaging techniques (USG, computed tomography and magnetic resonance imaging) and serological tests and antigen assays.[18]

The presence of an antigen specific for *E. granulosus* that appeared immunoelectrophoretically as a band of characteristic morphology and location when tested against sera from human patients has been reported.[19]

In a study, they reported a case of intraoral hydatid cyst of the buccal mucosa which was previously diagnosed as calcified buccal lymph node based on history and clinical examination. Later, only after the surgical removal of the lesion was done, histopathology confirmed it to be a hydatid cyst. No recurrence was noted.[7]

In another study, again, a case of hydatid cyst occurring in the buccal mucosa was reported. A provisional diagnosis of mucocele was made based on history and clinical examination. A histopathological diagnosis of hydatid cyst of the buccal mucosa was given later. Routine hematological examinations revealed no abnormalities. The patient was then started on albendazole at 15 mg/kg/day for a month. A chest radiograph and abdominal sonography scans were requested. No hydatid cysts were identified anywhere else in the body. The patient is on follow-up till date with no recurrences.[5]

The occurrence of the cyst intraorally has been well-documented. Involvement of the buccal mucosa is rare, and only two other cases have been reported in the literature. The latter occurred in a 6-year-old male and was asymptomatic with no organ involvement.[20] In all of these cases, a history of association with sheep or animals could not be elicited.

Case reports localized from the antrum reveal aspiration tests which are positive for the demonstration of protoscolices. However, they do carry the danger of dissemination or anaphylaxis. Again, histopathology remains the gold standard for diagnosis in such cases.[21]

In the present study, based on history and clinical examination, multiple provisional diagnoses of mucocele, epulis and salivary duct obstruction were given. Only after histopathology, a conclusive diagnosis of intraoral hydatid cyst of the labial mucosa was made.

Till date, no intraoral hydatid cyst localized to labial mucosa has been reported. This would be the first case to be reported. We would also like to mention that proper eliciting of history and making a habit of including hydatid cyst as a differential diagnosis in any cystic swellings of the head-and-neck region will help the clinicians to report such cases in future.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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