



A NOTEWORTHY PRESENTATION OF HYDATID CYST - CASE REPORT

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ABSTRACT

In general Swelling over sublingual regions will be provisionally diagnosed as Dermoid cyst / Ranula / Mucous retention cyst / Salivary gland tumours. But in rarest of rare cases, there may be chances of a Hydatid cyst. Hydatid disease or Echinococcosis is a zoonotic disease caused by Echinococcus granulosus as seen in this 29years old female with swelling over the sublingual region extending into the neck. Clinically appeared as dermoid cyst and radiological investigations suggestive of the possibility of ranula, so planned for surgery but surprisingly on table after opening the swelling there was a tough fibrous layer with underneath thick whitish lamellated membrane, anticipated as hydatid cyst which was also confirmed by the histopathological report.

KEYWORDS :**INTRODUCTION:**

Echinococcosis or Hydatid disease is a zoonosis, occurring primarily in sheep grazing areas of the world, commonly with dogs as definitive hosts, and sheep as the intermediate host. Humans are accidental dead-end hosts, which contract the disease through the ingestion of echinococcus eggs through contaminated water, foods, and direct contact with dogs, but there is no human to human transmission. Most commonly affects the liver and lungs (80%); rarely involves kidneys, spleen, pancreas, uterus, skin, and muscles[1]. Less than 1-2% of reported cases are seen in head and neck, and maxillofacial regions[1,2,3]. Involvement in the oral cavity is seen over buccal mucosa, the floor of the mouth, tongue, and maxillary sinus[2,4,5,6,7,8]

Here, we report a rare case of sublingual hydatid cyst in a 29-year-old female, managed at our institution.

CASE PRESENTATION:

A 29-year-old female presented with complaints of swelling on the floor of the mouth for 2 years, extending into the neck pushing the tongue to the left - since 8 months, gradual onset and progressive in nature associated with difficulty in chewing and talking. No history of a sudden increase in the size of swelling/discharge from swelling / fever/deviation of mouth/dribbling of saliva.

No history of smoking/tobacco-chewing/alcohol intake. No history of loss of weight/loss of appetite/halitosis. No history of diabetes / hypertension / thyroid illness / tuberculosis. On examination - A solitary cystic swelling of 5x3 cm, non-tender, non-warm, fluctuant is palpated over the floor of the mouth extending into the neck 5cm below the angle of the mouth, with limited mobility side to side and in the vertical direction. A 1x1 cm firm, the non-tender lymph node was palpated in level I-B location.



Figure 1: Clinical presentation of the patient.

CLINICAL COURSE:

Blood Tests	Hb - 12.6 g/dl WBC - 8000/mm3 Platelets - 3.1 lakhs/mm3 Creatinine - 0.56 mg/dl Serum amylase - 66.77 U/L Calcium A - 8.8 mg/dl K - 4.31 mEq/L CL - 104.9 mEq/l TSH- 1.42 mIU/ml
Ultrasonography Neck	Well defined thin-walled 4.1 x 3cm cystic lesion extending from the floor of the mouth to the adjacent right submandibular space posterior to the submandibular gland - likely Ranula
Computed Tomography Of Head And Neck	Well-defined oval thin-walled cystic lesion of fluid attenuation noted in the right sublingual space measuring 45.4 x 34.2 x 40.3 mm. Parotid, Submandibular, Sublingual salivary glands are normal. Features suggestive of Simple Ranula
MRI Head And Neck	Well-defined thin-walled T2 hyperintense cystic lesion measuring, 4.0[CC] x 3.8[T] x 4.2 [AP] cm with dependent debris not suppressing the parotid duct. Fat noted in the right sublingual space abutting and displacing the right genioglossus to the contralateral side. Features suggestive of Ranula
Gross Pathology	Unilocular cyst measuring 4 x 4 cm, along with grey-white membranous bits, showing multiple papillae.
Histopathology Report	Multiple sections from the cyst wall shows acellular eosinophilic laminated membrane, inflammatory granulation tissue, along with muscle bundles and inflammatory cell infiltrate composed of lymphocytes, plasma cells and eosinophils. Suggestive of Hydatid Cyst floor of the mouth

DISCUSSION:

Hydatid disease or Echinococcosis is caused by tapeworm *Echinococcus granulosus*, *Echinococcus multilocularis* or *Echinococcus ligartus*. It affects both males and females, equally with an average age of about 45 years. It is endemic in the Mediterranean, Middle East, Asia, Australia, South America, and Southeast African countries [9]. The adult tapeworm is attached to the villi of the ileum, of the definitive host (dog). Up to thousands of ova are passed daily and deposited in the dog's faeces. Sheep are usually intermediate hosts but humans are accidental intermediate dead-end hosts. An individual, who is infected by eating food or drinking water contaminated with matured eggs, is considered an accidental host [10]. In the human duodenum, the parasite embryos release an oncosphere containing hooklets that penetrate mucosa allowing access to the bloodstream. In the blood, the oncosphere reaches the liver (most commonly) through a portal vein or lungs through the capillary bed, where the parasite develops in the larval stage - Hydatid Cyst. Three weeks after infection, a visible Hydatid cyst develops, which slowly grows in a spherical manner. Other mechanisms for spreading are the systemic release through the lymph nodes and direct spread to adjacent sites [11]. The involvement of the head and neck is rare and involvement of the sublingual glands is extremely rare [12,13]. It contains 3 layers:

- 1 . Pericyst / Fibrous capsule - derived from host tissues, develops around the Hydatid cyst.
2. Ectocyst - outer gelatinous membrane of cyst wall
3. Endocyst - inner germinal membrane of cyst wall.

Brood capsules are small, intracystic cellular masses in which future worm heads develop into scoleces. In the definitive host, the scoleces develop into adult tapeworms but in the intermediate host, they can differentiate only into a new Hydatid cyst.

Free rupture of cysts can result in disseminated *Echinococcus* or a potentially fatal anaphylactic reaction.

The best diagnostic modalities are sonography, CT scan, Magnetic Resonance Imaging (MRI), serologic tests (Enzyme-Linked Immunosorbent Assay (ELISA), Casoni test, latex agglutination and direct hemagglutination, Fine Needle Aspiration (FNA), and histopathology [9].

Imaging studies reveal a simple hydatid cyst to be well-circumscribed with budding signs on the cyst membrane and may contain free-floating hyperechogenic hydatid sand on ultrasonography. Rosette appearance and Water Lily signs are seen when daughter cysts are present. Calcifications in the wall of the cyst are highly suggestive of Hydatid disease and can be helpful in the diagnosis. Similar findings are seen on CT /MRI scans.

Treatment of Echinococcosis with Albendazole or Mebendazole is effective at shrinking cysts in many patients with *E. granulosus* infection, but cyst disappearance occurs in < 50% of patients. The dosage scheme for Albendazole, endorsed by World Health Organisation [WHO] is three 28 days courses of 10mg/kg/day in divided doses separated by 2 weeks intervals [16]. Preoperative treatment may decrease the risk of spillage and is a reasonable and safe practice [14].

Surgical options encompass excision (or pericystectomy), Marsupialization, leaving the cyst open, and drainage of the cyst. To avoid intraoperative contamination, proscolicidal solutions like 75% - 90% ethanol, 20% hypertonic saline

solution, and 0.5 % cetrimide are used. Incomplete removal of the germinal membrane from the cyst cavity is the major cause of recurrence [8.5%-25%].

Medical therapy without definitive resection or drainage should be considered only for widely disseminated diseases or poor surgical candidates. Chemotherapy in the postoperative period is essential for preventing recurrence. (15)

The present case was provisionally diagnosed as a dermoid cyst clinically, but radiological investigations and anatomical location revealed suspicion of Plunging Ranula. Excisional biopsy was done with the intraoral approach under general anaesthesia, intraoperatively after incising the buccal mucosa over the floor of the mouth, tough fibrous layer was present underneath the thin elastic whitish laminated membrane which was anticipated as Hydatid Cyst, confirmed by histopathological examination. The postoperative period is uneventful. The patient was prescribed tablet Albendazole for 3 months and had no symptoms.

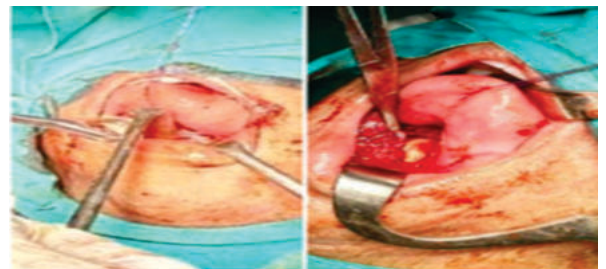


Figure 2: Intraoperative picture depicting excision of swelling over the floor of the mouth.

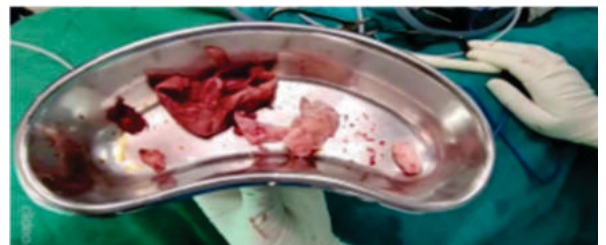


Figure 3: Intraoperative picture, after excision of the swelling.

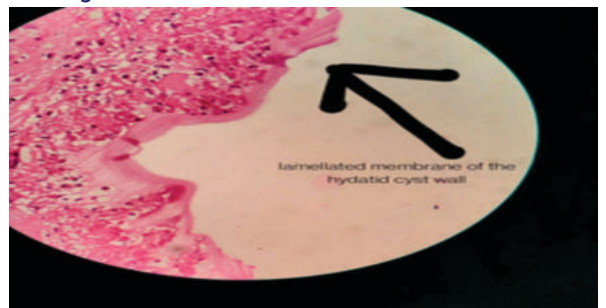


FIGURE 3: Histopathological image showing lamellated membrane of the hydatid cyst wall.

CONCLUSION:

The present case illustrates the presence of hydatid cysts in atypical anatomical locations in endemic regions. Based on the presentation and Imaging studies a differential diagnosis of dermoid cyst and ranula were made. Intraoperatively, it was diagnosed as a Hydatid cyst and confirmed by histopathology. Postoperative chemotherapy with albendazole was given.

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