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Hydatid Cyst of the Floor of the Mouth: A Case Report

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Abstract **Case Report**

The occurrence of a hydatid cyst in the face is rare. We report the case of a 24-year-old male with a slow-growing swelling in the floor of the mouth. The patient's history revealed chest drainage from a ruptured pulmonary abscess four years ago, without a documented bacteriological diagnosis. Clinical examination found a non-inflammatory and encapsuled swelling lifting the tongue without adenopathy. MRI scan showed a well-encapsuled multivesicular cyst (05 x 04 cm). Needle aspiration for cytology and hydatid serology were inconclusive. A complete blood count (CBC) and chest radiography revealed no abnormality. The cyst was entirely removed, and the histopathological examination conclusion was compatible with hydatid disease, which confirmed our thoughts regarding the patient's history.

Keywords: Hydatid cyst, Cervicofacial hydatid.

INTRODUCTION

Hydatid disease, hydatidosis, or cystic echinococcosis (WHO) is an anthropozoonosis caused by the canine taenia parasite, Echinococcus granulosus (E. G.). The Mediterranean basin is an endemic area where human infection occurs through direct or indirect contact with the canine-cattle cycle. Hydatid disease can occur in one or more organs. In theory, it can be found in all organs of the human body. The most common sites of hydatid disease are the liver and lungs. Head and neck sites are rare. This report presents a case of intraoral hydatid disease.

OBSERVATION

Mr. M. S., 24 years old, from and living in Timimoun (Southern Algeria), was referred to our department in May 2017. He had a chronic mass on the floor of his mouth that had been developing for several years.

Medical history revealed that the patient was admitted to the pneumological department of the EPH in Adrar in September 2013 for ruptured right lung abscess, pyo-pneumothorax, and bronchial dilatation. The patient had received antibiotic treatment and rehabilitation, and the cyto-bacteriological study of the pleural puncture was not found.

Clinical examination revealed a cystic, flexible, non-pulsatile medial mass of the floor of the mouth, 05 cm long. There was no erythema, pain, or fistulisation. Tongue is movable with dysphagia on solid food, without dyspnea or dysphonia. No motor, sensory, or sensory damage to the tongue was noted (Fig 1).

Examination of the submandibular glands and Wharton's ducts showed no abnormalities. The lymph nodes were clear, and other ENT and general examinations were unremarkable.

An MRI of the face showed a rounded, homogeneous, fluid-filled, multivesicular sublingual mass measuring 54 mm in width, surrounded by a thin wall without enhancement. This was suggestive of a hydatid cyst (Fig 2).

The examination of the submandibular glands and Wharton's ducts was unremarkable; the lymph nodes were clear; and the remaining ENT and general examinations were unremarkable.

MRI of the face showed a rounded, homogeneous, fluid-filled, multivesicular sublingual mass measuring 54 mm in width, surrounded by a thin wall without enhancement, suggestive of a hydatid cyst (Fig 2).

The blood count showed no hypereosinophilia. Cytopuncture of the mass and serology were negative. Chest computed tomography showed diffuse interstitial syndrome with bronchial dilatation and scarring. A mixed ventilatory defect with predominantly obstructive airways was noted on respiratory function tests (RFTs). The abdominal ultrasound was unremarkable.

Surgical removal of the mass was performed through a vertical midline floor incision, the cleavage plane was well identified, and pericystectomy was performed without cyst rupture or lavage with hypertonic saline. Intraoperative puncture revealed a citrine-yellow aqueous fluid (Figs 3 and 4).

Histological analysis of the specimen confirmed the diagnosis of a hydatid cyst. The 8-month follow-up was satisfactory.

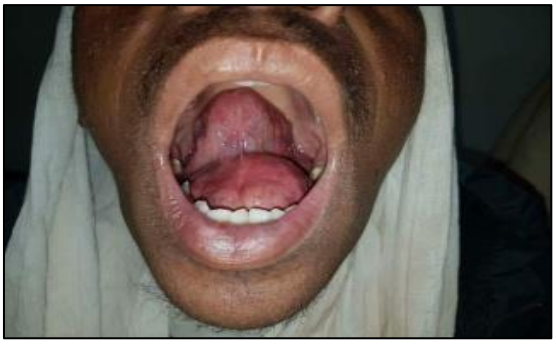

Fig 1: A chronic non-inflammatory cystic mass of the floor of the mouth in a 24-year-old man

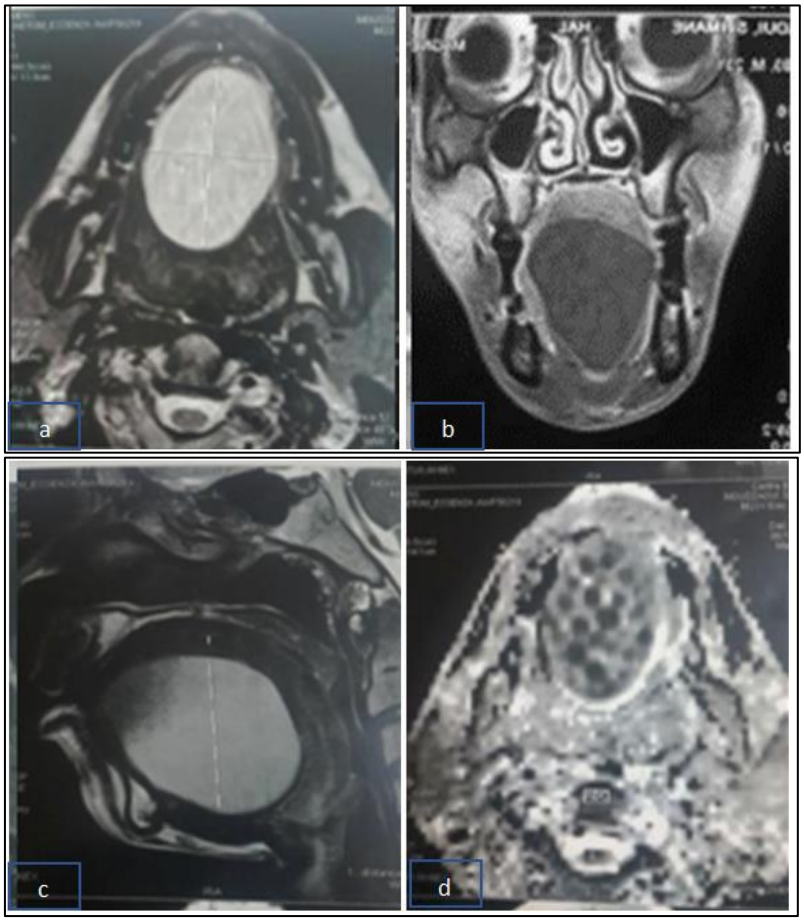

Fig 2: (MRI of the face): a rounded, homogeneous, fluid-filled, multi- vesicular, sublingual mass measuring 54 mm in width, surrounded by a thin wall with no elevation and no infiltration of neighbouring structures, suggestive of a hydatid cyst

Fig 3: Median incision of the mucosa of the floor of the mouth, dissection of the cyst and removal of the pericyst

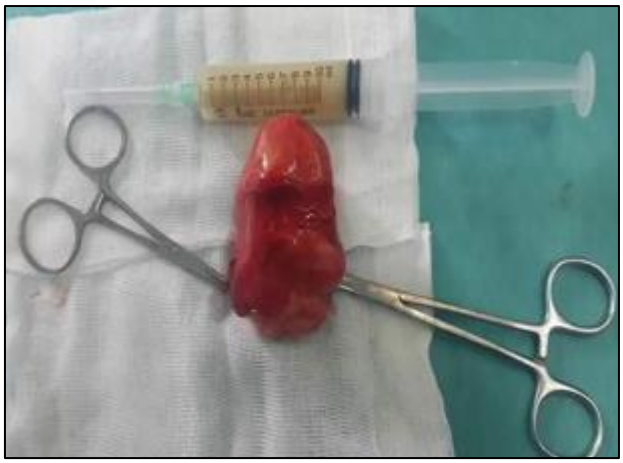

Fig 4: Removal of the cyst with pericystectomy, puncture: aqueous citrine yellow fluid

DISCUSSION

Hydatidosis is an anthropozoonosis caused by a cosmopolitan metazoan of the genus Echinococcus Granulosus (E. G.). The infection is widespread in cattle-breeding regions such as China (33 cases/100,000 population/year), Latin America (143 cases), and sub-Saharan Africa (220 cases).

The Mediterranean basin is an intermediate endemic zone (10 to 14 cases/year/100,000 inhabitants) between Algeria, Tunisia, and Morocco [1-4].

Canids (dogs) are the reservoir and final host of the parasite cycle. The infective form (eggs) is excreted in the dog's feces. The intermediate host (cattle) or human (accidental definitive host) is infected by direct contact (with the dog) or indirect contact (soiled objects or food) and ingestion of the expelled eggs [5, 6].

Ingested eggs release their embryos, which cross the intestinal barrier and enter the portal venous system. The liver is the first obstacle (60–75% of cases). It then progresses to the lungs (in 15 to 30 percent of cases). Rarely, the embroyon crosses these two barriers, embolizes into the general circulation, and invades all other organs (10% of cases) [1, 7].

One month after ingestion, the embroyon transforms into its larval (cystic) form in the affected organ; its growth rate depends on the organ affected (0–05 cm/year) [5].

The most commonly affected organs are the liver (60 to 80% of cases), lungs (25 to 40% of cases), and spleen (2 to 5% of cases). Cervicofacial involvement is rare (1 to 2% of cases). Between 1967 and 2002, six (06) cases of submaxillary hydatid cysts were reported in the literature [8].

Clinically, the condition may remain asymptomatic for a long time. In most cases, it is a cystic mass with a local compressive effect depending on the organ (dyspnea, dysphonia, dysphagia, or paralysis). An infectious syndrome (abscess) or an immunological syndrome (urticaria, anaphylactic shock) may be observed.

As in our case, the patient's medical history was studied in detail (origin, occupation, abdominal, pleuropulmonary, or musculoskeletal signs). The complete blood count (CBC) shows hyper-eosinophilia in 30% of cases [9].

MRI plays an important role in establishing the diagnosis by showing the exact location of the cyst, its type (multilocular or septate), its wall, and other features. The contents of the cyst are unchanged in the T1 hypersignal after intravenous contrast injection and in the T2 hypersignal. The septa show T1 and T2 hypersignals.

According to recent studies, MRI can differentiate between the parasitic, non-parasitic, or traumatic origin of the cystic mass by the "low signal intensity rim". This rim sign is considered pathognomonic for hydatidosis [10].

A CT scan shows a fluid mass, well demarcated by a slightly raised rim, with peripheral calcifications, unilocular or multilocular, unenhanced after contrast injection, and without evidence of locoregional aggression.

If the condition is suspected, a systemic radiological assessment (TLT, ultrasound) is mandatory.

From a biological point of view, aspiration puncture can find protoscolex or internal membrane debris but is not very sensitive (false negative) and dangerous (anaphylactic shock) [11].

Among the various techniques (ELISA, haemagglutination, latex agglutination, etc.), immunoelectrophoresis is the most sensitive (90–95%), with a positive result up to 1 year after surgery [6]. It is an important follow-up parameter considering the decrease in diagnostic sensitivity for extra-hepatico-pulmonary localisations (only 50%) [11].

In practice, serological diagnosis should be based on a combination of two techniques, one quantitative and the other qualitative.

Surgical treatment of the hydatid cyst with pericystectomy, removal of the residual cavity, and lavage with hypertonic saline is the preferred option (90% cure rate).

Percutaneous aspiration and irrigation (PAIR) are currently under investigation [12]. Medical treatment with benzimidazoles (albendazole) and praziquentel is indicated for multiple lesions or when surgery is contraindicated and results in resolution of the cyst in 30% of cases and a reduction in cyst size in 50% of cases [12-14].

CONCLUSION

The rarity of ENT hydatid disease can lead to misdiagnosis. This pathology should be suspected in the presence of an asymptomatic cystic mass, especially in endemic areas.

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