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OTHER ARTICLES

Pterygopalatine-Infratemporal Fossa Hydatid Cyst Resembling Cystic Tumor

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Abstract
Hydatid cyst is an endemic disease in Mediterranean and Middle Eastern countries, Eastern European countries, East Africa, China, New Zealand, Australia. We aimed to present this educational case, which is endemic in our country and seen in a very rare localization, with the combined surgical approach, within the literature.

Keywords Echinococcus · Infratemporal fossa · Maxillary sinus · Cysts · Middle East · Skull base neoplasms

Introduction

Hydatid cyst (HC) is a zoonotic parasitic disease, commonly caused by the larvae of Echinococcus granulosus. A small tapeworm that can cause disease in intermediate hosts, usually herbivorous animals, and incidentally infected individuals. Affects 1–220 people in 100,000 depending on the region. It is an endemic disease in Mediterranean and Middle Eastern countries, Eastern European countries, East Africa, China, New Zealand, Australia [1].

In this disease, humans become intermediate hosts by ingestion of the larva. Larvae pass through the intestinal mucosa into the systemic bloodstream using the intestinal mesenteric veins. From there, it comes to the liver via the portal venous system. Every patient with echinococcosis should have a whole-body scan because 20–40% of patients may have multiple organ disease [2]. Although hydatid cysts can be seen all over the body, the most frequently involved organ is the liver (65%), the first organ they come from via the portal system. Other affected sites are the lungs (25%) and less commonly the spleen, kidneys, heart, bone, central nervous system, and soft tissues [3]. Head and neck involvement by echinococcosis is a rare entity, and involvement of the infratemporal region is extremely rare even in endemic areas. Very few cases of hydatid cyst located in the pterygopalatine-infratemporal fossa (PIF) have been reported in the literature [4, 5]. We aimed to present this zoonotic infection, which has a clinical presentation in a rare localization and is confused with an infective cystic tumor, in the light of the literature.

Case Presentation

A 39-year-old female patient applied to the dentist because of swelling in the right half of her face. Gingiva biopsy was performed in the patient whose complaints did not regress after two weeks of amoxicillin clavulanic acid treatment. Gingiva biopsy result was interpreted as normal mucosa. Afterwards, the patient who applied to our clinic was given intravenous (iv) ampicillin sulbactam treatment for two weeks. The swelling on the right malar region of her face regressed. There was no history of contact with cats, dogs or wild animals and she had no additional disease. Gadolinium-enhanced maxillofacial magnetic resonance imaging (MRI) was performed. A cystic mass of 19 × 13 millimeters (mm) with peripheral enhancement was detected, which was hypointense on T1 examination, heterogeneous hyperintense on T2 examination. Surgical exploration was planned since malignancy could not be ruled out. After elevation of the mucosal flap from the gingivobuccal area, a small window was created

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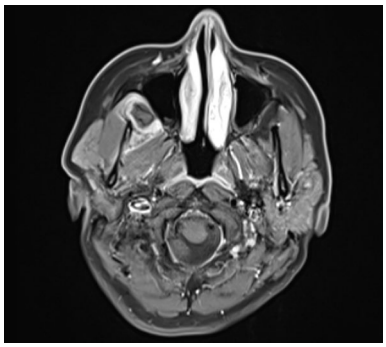

Fig. 1 The appearance of the cyst on T1 MRI sections

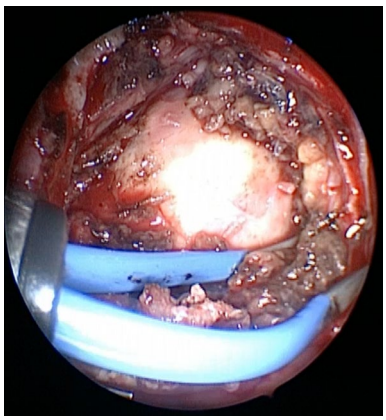

Fig. 2 Intraoperative view of hydatid cyst in the pterygopalatine-infratemporal fossa

in the bone in accordance with the Caldwell-Luc procedure. Unsinectomy, partial resection of the lower turbinate, medial maxillectomy were performed under guiding of endoscopic approach. Through Caldwell-Luc window and enlarged maxillary sinus ostium, posterior wall of maxillary sinus was removed and reached to cyst area. The internal maxillary artery was ligated with a clip. The cyst was separated from the surrounding adipose tissue and completely excised. There was not any complication postoperatively. Patient was discharged on postoperative third day. Pathologic examination of excised material was reported as hydatid cyst. Intraoperative view and radiological view are shown in the figures. (Figures 1 and 2) Physical and radiologic examination of abdomen, thorax, and other systems were normal. Patient was sent to infectious disease department for further evaluation and treatment. Albendazole treatment has been prescribed for the patient and she comes to regular examination. There is no recurrence or symptoms during follow-ups periods.

Written and verbal consent was obtained from the patient, and ethics committee approval was not obtained because it was a case report.

Discussion

It is very rare for hydatid cyst disease to involve the head and neck regions [6, 7].

The infratemporal fossa and the pterygopalatine fossa are extremely rare and unusually surprising localizations for this disease. If parasite embryos pass through the liver and lung systems, they can form infratemporal and pterygopalatine hydatid cyst disease [8].

In our patient, HC seen in PIF localization was detected and treated.Symptoms of the disease are variable and may vary depending on the location and extent of the cyst.Generally, the symptoms of HC disease located in PIF occur due to the pressure of the mass.History, imaging and clinical suspicion may help in the differential diagnosis.Serological tests can also help in diagnosis.Casoni tests and eosinophilia are not specific for HC.The use of fine needle aspiration cytology is controversial due to the risk of anaphylaxis and dissemination of daughter vesicles.Some authors reported that cyst HC is a safe diagnostic method [9].

In the differential diagnosis of PIF masses, diseases such as epidermoid cyst, meningocele, carcinoma, melanoma, schwannoma, neurofibroma, neurofibrosarcoma, chordoma and teratoma should be kept in mind [10]. Surgical removal of HC is the most effective definitive treatment method in the maxillofacial region.It is very important that the cyst does not rupture, because of the risk of anaphylaxis and the spread of larvae and causing recurrence.Mebendazole and albendazole can be preferred as medical treatment [5]. In our rare case with PIF localization, we cured our patient with a combined surgical technique and the participation of three surgeons without complications. The patient's medical treatment continues in the infectious diseases clinic.

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Declarations

Conflict of interest The authors have no conflict of interest to declare.

Informed Consent Written informed consent was obtained from the patient who agreed to take part in the study.

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