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Calcified Cystic Echinococcosis in Masseter Muscle-A Rare Case and Literature Review

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Abstract: ***Introduction:*** ***Echinococcus granulosus is a small intestinal tapeworm found in dogs and occasionally in other carnivores. In the literature, information on the incidence of Echinococcus manifestations in the head (noncerebral) and neck region is relatively rare.*** ***Purpose:*** ***To present a case of an unusual case of echinococcus in the masseter muscle*** *of a young female patient at the Department of Oral and maxillofacial surgery, University Hospital 'St. .Anna', Sofia.* ***Material and Methods:*** ***The medical history of 14 years old female patient revealed a painless swelling of the left masseter muscle. Bimanual palpation showed two mobile thick lesions in the left masseter muscle. Pain and neurologic deficits were absent. Salivary flow was normal from the left Stennoni's duct. The patient denied any prior medical problems. All hematologic parameters were within normal limits- no eosinophilia was evident. A computed tomography (CT) scan was obtained, and it showed two, well-defined lesion - parasite cysts in the left masseter muscle which were with high degree of calcification. The patient was scheduled to undergo excisional biopsy of the lesion under general anaesthesia. Result: Pathohystological examination showed the presence of the cyst parasite hooklets fully calcificated with characteristic for E. granulosus which were non active lesions (Hematoxylin and eosin stain original magnification _10.) showed absence of scolexes).*** ***Conclusion:*** ***Although a rare event, echinococcosis must be considered in the differential diagnosis of head and neck tumours.***

Keywords: *Echinococcus granulosus*, masseter muscle, maxillofacial region

1. Introduction

Echinococcus granulosus is a small intestinal tapeworm found in dogs and occasionally other carnivores. Shedding gravid parasite proglottids or eggs passing in the dog feces occurs within 4 to 5 weeks after infection of the definitive host. Ingestion of eggs by intermediate host animals (e.g., cattle, sheep, pigs) or humans results in the release of an oncosphere into the gastrointestinal tract, which will migrate to primary target organs, such as the liver most frequently, followed by the lungs and other organs, such as kidney, spleen, brain, heart, and bone.(11) Usually a fully mature hydatid cyst is formed after several months or years. The average increase in cyst diameter varies from a few millimeters to 5 cm per year. Tissue damage and organ dysfunction result mainly from this gradual process of space-occupying repression, or from displacement of vital host tissue, vessels, or organs.(8) Consequently, clinical manifestations are primarily determined by site, size and number of cysts, and are thus considerably variable.(13) In the literature, information on the incidence of Echinococcus manifestations in the head (noncerebral) and neck region is relatively rare. Cases have been described with cystic lesions located in the mandible, maxillary sinus, submandibular and parotid gland, neck, mastoid, the infratemporal and pterygopalatine fossa.(1, 2, 4, 7, 16, 17, 18, 19) When located at these sites, the disease often has a long history before echinococcosis is correctly diagnosed. The primary clinical diagnosis includes nonspecific symptoms also encountered in tumours at the same site and should be followed by specific morphologic features detected by imaging techniques (such as computerized tomography [CT], magnetic resonance imaging, or ultrasound, and by supporting specitic immunodiagnostic results.(13) CT is preferable for the detection of extrahepatic lesions and for voluminometric follow- up assessment. Calcification of variable degree occurs within the periphery of the cyst in about 10% of cases. Aspiration cytology appears particularly helpful in detecting pulmonary, renal, and other nonhepatic lesions for which imaging techniques and serology do not provide appropriate diagnostic support.(13)

Immunodiagnostic screening tests such as the enzymelinked immunosorbent assay (ELISA) using E granulosus hydatid fluid antigen are diagnostically relatively sensitive (85% to 98%) with the exception of pulmonary cyst localization (50% to 60%).(5,14) The specificity of ELISA is relatively low. Thus, primary ELISA-positive sera are retested in a confirmation test such as antigen-5-precipitation (arc-5-test: diagnostic sensitivity, or immunoblotting for a metacestodespecific S-kd hydatid fluid polypeptide antigen. (9,15) After surgical resection of parasitic lesions, the etiologic proof of *E granulosus* as the infecting organism may be achieved microscopically by the demonstration of characteristic protoscolices and free hooklets.(6) If these structures are absent, a specific diagnosis may be achieved by immunohistologic identification of the parasitic laminated layer or by the demonstration of *E granulosus* - specific DNA on polymerase chain reaction. However, PCR requires nonfixed, nativetest samples(10,13).

2. Case Report

Here, we highlight a case of persistent asymptomatic facial swelling in the right masseter muscle leading to an unusual diagnosis of calcified echiniccocus cysts.

We present a case report of a healthy young woman who came with a chief complaint of swelling on right side of face since some weeks. She became aware of it accidentally. The painless swelling was not related to any traumatic episode. There was no history of previous swelling in the same location. Clinical examination revealed a localized extraoral asymetry. Skin over the site was normal in colour. On palpation we distinguished a swelling which was firm in consistency. The ultrasound investigation showed two well defined calcifying formations. Few rounded hyperechoic calcific densities were seen within masseter muscle without

cyst formation on CT imaging (Fig.1). Routine laboratory findings were all normal; no eosinophilia was evident. We performed a surgical removal of the calcified cysts with extraoral approach and the pathohystological result was dead calcifying echinococus cysts. ELISA test was negative. The postoperative period was without any problems.

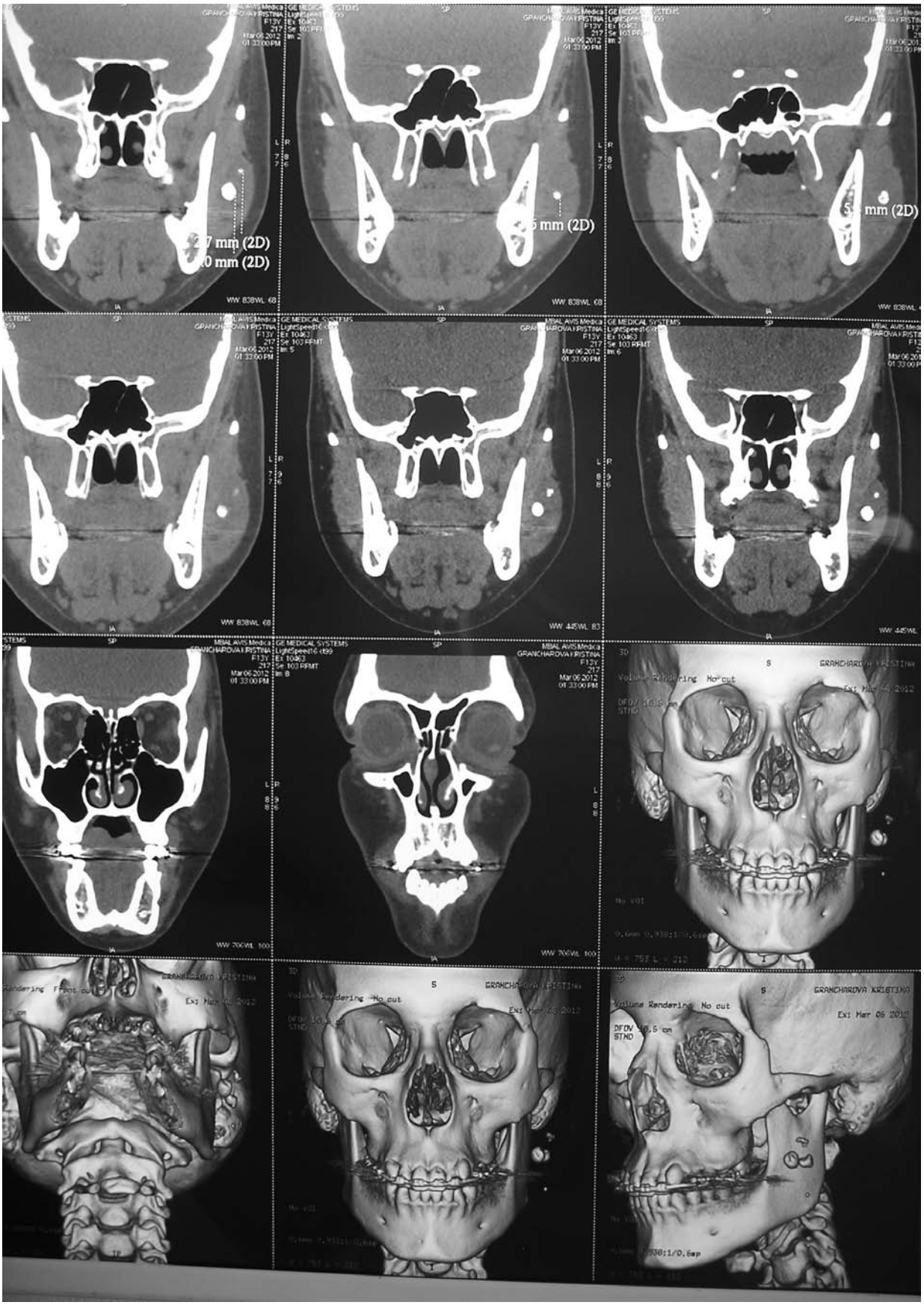

Figure 1: CT image of echinococcus calcified cysts in left masseter muscle

3. Discussion

Although a rare event, echinococcosis must be considered in the differential diagnosis of head and neck tumours and calcified cyst should be distinguished from phlebolities.(12) Typically, these patients have a long history of relapsing lesions with many unsuccessful therapeutic interventions, especially in cases in which the specific diagnosis remains unsuccessful and in areas of low endemicity where clinicians lack the appropriate experience; however, our patient had no such medical hystory. In doubtful cases, an appropriate immunodiagnosis can significantly support the clinical diagnosis of cystic hydatid disease. For follow-up studies, imaging procedures may provide appropriate information of progressive or regressive processes within the parasitic area. Consequently, an optimized assessment of treatment efficacy must be based on a multidisciplinary approach including clinical, parasitologic, and immunologic parameters, and each case needs to be considered individually. Under certain conditions (e.g., pregnancy), asymptomatic patients may undergo clinical observation only, with close follow-up every 3 months.(20) Specific indications for surgery include superficial, viable cysts at risk of spontaneous or traumatic rupture, spinal, bone, and infected cysts among others. A very small or calcified dead cyst is a relative contraindication to surgery. Preoperative chemotherapy with albendazole (10 to 15 mg/kg/d) may be indicated to reduce the risk of a secondary echinococcosis after the operation and should begin at least 4 days before surgery and be continued for at least 1 month, preferably several months. Today, continuous therapy may be preferred over therapy in two or three cycles.(3,4) Chemotherapy also should be used after any accidental hydatid fluid spillage, such as during surgery.(20)

4. Conclusion

Our patient with calcified degenerated echinococus cyst opted for surgical treatment because she complained of some cosmetic asymmetry but no other somatic reasons. She did agree to return for evaluation for further follow-up.

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