

Case Report - Infection

# Echinococcal cyst affecting the mandible

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## ABSTRACT

Echinococcosis is a parasitic infection also called as hydatid disease or hydatidosis. Hydatidosis is a cyclo-zoonotic infection of the larvae form of *Echinococcus granulosus* (canine tapeworm). The majority of hydatid cysts are seen in the liver (65%) as most of the embryos are trapped within it. Infratemporal region is an unusual site for hydatidosis and has been sparsely reported in the literature. To the best of our knowledge, this is the fourth and perhaps the largest case report of hydatidosis involving the mandible in the literature in right infratemporal region involving the ramus condyle unit in a 35-year-old female with chief complaint of pain on right side of temporomandibular joint area on mastication, and opening and closing of mouth.

**Keywords:** Echinococcosis, hydatidosis, parasitic infestations

## INTRODUCTION

Echinococcosis (hydatid disease or hydatidosis) is a near cosmopolitan cyclo-zoonotic infection in humans caused due to infestation by the larval stages of taeniid cestodes of the genus *Echinococcus*. Six species have been recognized, but four are of health concern: *Echinococcus granulosus* (which causes cystic echinococcosis), *Echinococcus multilocularis* (which causes alveolar echinococcosis), and *Echinococcus vogeli* and *Echinococcus oligarthrus* (which cause polycystic echinococcosis). Two new species have recently been identified: *Echinococcus shiquicus* and *Echinococcus felidis*, but their zoonotic transmission potential is unknown.<sup>[1]</sup> It is endemic in regions where close and continuous contact exists among dogs and sheep. Highest prevalence of the disease is found in the Mediterranean and parts of Central Asia, South America, Australia, and Africa.<sup>[1]</sup> The life cycle of *Echinococcus* involves a definitive host (dog) and an intermediate host (usually sheep) with humans as an accidental host following ingestion of the larvae. Once ingested, the larvae pass into the blood stream through the intestinal mucosa, where they enter portal circulation to infest the liver as this is the first organ that they pass through. However, in all cases of echinococcosis, a thorough systemic investigation should be performed, as 20–40% may have multiorgan involvement.<sup>[1]</sup>

Although the occurrence of a single echinococcal cyst is common in most cases, it is atypical that a cyst will occur in the maxillofacial region without evidence of additional hepatic or lung involvement, although the embryos must have passed through the organs. Echinococcal cysts in the maxillofacial region are rare, and only a few cases (<0.1%) have been reported in the English literature.<sup>[1-3]</sup> Recommendations for diagnosis and management are substantially available for hepatic and pulmonary diseases, and maxillofacial surgeons are left with diminutive guidance when coming across such rare disease.<sup>[4]</sup> We present a case of a middle-aged woman with a single echinococcal cyst affecting the mandible and emphasizing the need to consider it in the differential diagnosis of slow-growing lesions of maxillofacial regions.

## CASE REPORT

A 35-year-old married female from countryside with poor socio-economic background reported to our department with a complaint of mild pain and pressure sensation on right side of temporomandibular joint (TMJ) area on mastication and opening and closing of jaws since 2–3 years. The associated pain was dull, localized, and aggravated on talking and taking food. She was unemployed and managed all domestic chores at home.

On examination, there was no gross facial asymmetry, deviation of jaw toward left side on opening and closing was appreciated with acceptable mouth opening. Clinical examination revealed a painless, soft mass in the right TMJ area with egg shell cracking and no evidence of a sound condylar unit. TMJ movements were diminished on right side. Intraorally, occlusion was good with healthy gingiva. The general health of the patient was normal with no cervical lymphadenopathy.

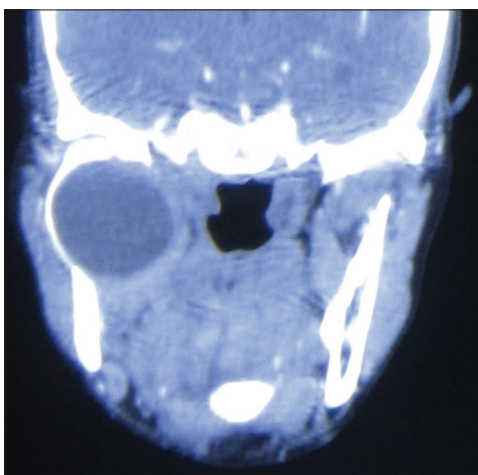
An orthopantomogram of the region revealed an ill-defined osteolytic lesion involving the ramus condyle unit of the right side of size 3 cm × 1.5 cm. Computerized tomographic scan showed a well encapsulated fluid filled cystic lesion of size 4.9 cm × 3.8 cm with osteolysis and thinning out of the condylar unit encroaching the infratemporal region and abutting the pterygoid plates on right side [Figures 1-3]. USG liver and plain posteroanterior view chest radiograph were normal with only a slight increase in eosinophil count. Surgical procedure under general anesthesia was planned. The region was approached via an extended Risdon's approach extending and curving near the lower ear lode. Dissecting sub platysmally, pterygomassetric sling was incised. The cyst was judiciously dissected out of the

surrounding tissues in toto. The intraoperative and postoperative course were uneventful.

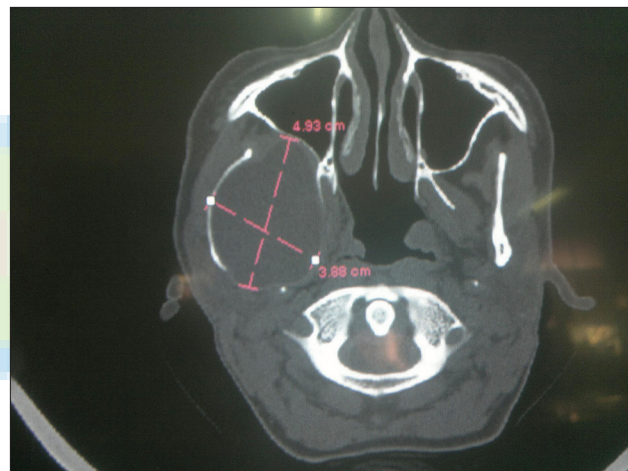
The gross macroscopic specimen revealed two cystic cavities. The outer was tough reddish-brown and the inner was thin, slimy and fragile [Figure 4]. In histopathology, scanner view, ×10 view and ×40 view showing daughter cysts with scolex and germinal layers [Figures 5-7]. The patient was followed up for 2 year with no recurrence.

## DISCUSSION

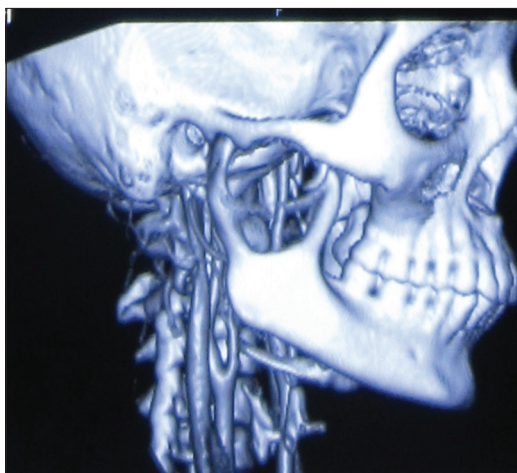
Maxillofacial zoonosis with an echinococcal cyst is a rare event. The Greek word "*Echinococcus*" meaning, "Hedgehog Berry" is a term to describe the gross pathology of the lesion, meaning "a drop of water." Hippocrates very well described the disease affecting the liver masking it's appearance and aptly described it as "liver filled with water." Apart from the present case there are only 3 other cases of echinococcal cyst of the mandible reported in the literature.<sup>[5-7]</sup> other than mandible, echinococcal cyst has been reported to affect the neck, maxilla, pterygopalatine fossa,



**Figure 1:** Presurgical coronal computed tomography showing well-encapsulated lesion



**Figure 2:** Presurgical axial computed tomography showing well-encapsulated lesion

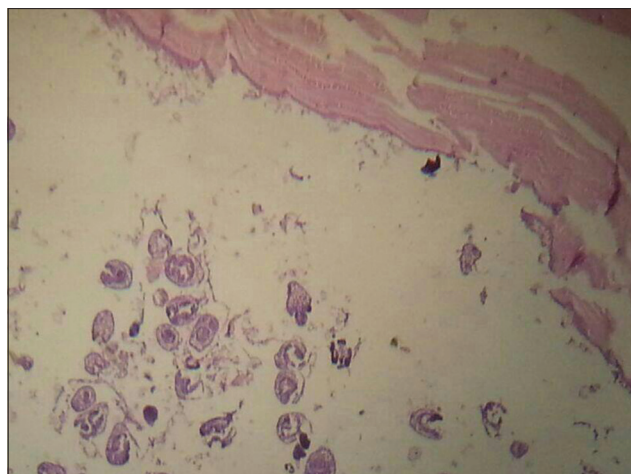


**Figure 3:** Presurgical three-dimensional reconstruction computed tomography showing thinning out of the condylar ramus unit on right side

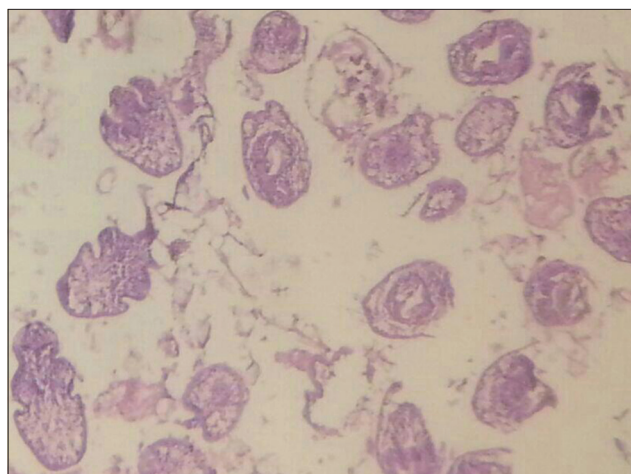


**Figure 4:** The gross macroscopic specimen revealed two cystic cavities. The outer was tough reddish brown and the inner was thin, slimy and fragile

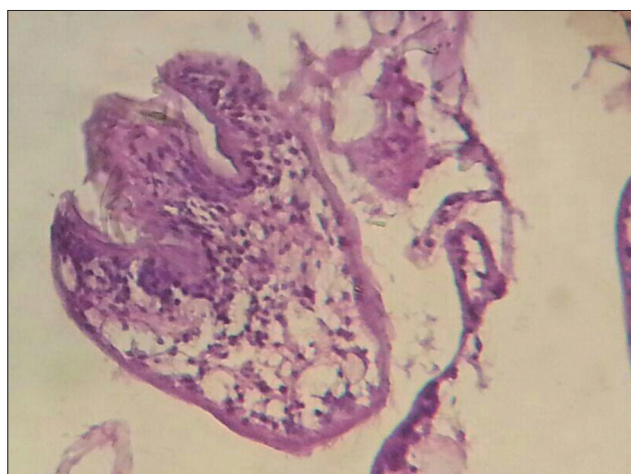




**Figure 5:** Scanner view showing daughter cysts with scolex and germinal layers



**Figure 6:** ×10 view showing daughter cysts with scolex and germinal layers



**Figure 7:** ×40 view showing daughter cysts with scolex and germinal layers

infratemporal fossa, parotid gland, and base of the skull. In majority of these cases, the disease appeared asymptomatic, docile, and benign in growth with secondary symptoms depending on their location. However, even in areas where the disease is endemic,

echinococcal cyst in the head and neck region is very rare, and is not usually considered in a differential diagnosis of a cystic swelling in the head-neck and maxillofacial region.<sup>[8,9]</sup>

Humans acquire the disease accidentally due to close association with either the definitive hosts, like dogs, or intermediate hosts like sheep, goats, or through contaminated foods. As an accidental host men and women are affected equally. After the ova enter and hatch as embryo in the intestinal mucosa, migrate through the intestinal mucosa and enter the portal circulation. After portal circulation, they reach the pulmonary circulation and finally the systemic circulation. Hence, hydatidosis of bone is uncommon.<sup>[10]</sup> These lodge within capillaries of various organs and invoke an inflammatory response of mononuclear cells and eosinophils. While many of them are destroyed, a few survive by forming a slow-growing, thick-walled cyst inside which they divide. However, the life cycle, that is, formation of the adult tapeworm, cannot be completed unless one of the canine species, its primary host, ingests these cysts.

The diagnosis of *Echinococcus* infection can be based on the clinical history, physical examination, diagnostic imaging, aspiration, and serologic test. Regardless of location, the sensitivity of serologic tests is inversely related to the degree of sequestration of the echinococcal antigens inside cysts; for example, healthy, intact cysts can elicit a minimally detectable response, whereas previously ruptured or leaking cysts are associated with strong responses. The indirect hemagglutination test is sensitive, but has now been replaced by the enzyme immunoassay (ELISA) for initial screening of sera. In the present case, the diagnosis of the disease was not considered before surgery, and a definitive diagnosis was made only by postoperative histopathology.

Until the 1980s, surgery was the only option for treatment of echinococcal cysts; however, chemotherapy with benzimidazole compounds and, more recently, treatment with cyst puncture, aspiration, injection of chemicals and re-aspiration (PAIR) have been introduced and increasingly, have supplemented or even replaced surgery as the preferred treatment. However, surgical removal of intact echinococcal cysts, whenever possible, remains the treatment with the best potential to remove cysts and lead immediately to complete cure. Surgical technique is also relevant to the further clinical course of the patient, as spillage of cyst content can lead to dissemination or local recurrence of disease and, therefore, an excision of the cyst in toto is highly desirable.<sup>[9,10]</sup> The intraoperative use of scolicedal compounds is questionable. There is no ideal agent that is both effective and safe. For optimum efficacy, compounds require a 15 min “dwell time” within the cavity. Compounds that seem to be fairly safe and effective include 70–95% ethanol, 15–20% saline, and 0.5% cetrimide solution. Surgery is contraindicated in patients who refuse it, are pregnant, have preexisting medical conditions that put them at risk, or have multiple cysts that are difficult to access.

Another feasible option can be image-guided PAIR of scolecicidal method for percutaneous treatment of hydatid cyst of parotid gland,<sup>[11]</sup> have been reported as effective procedure and as an alternative to surgery, however, controversy exists regarding aspiration of cystic contents, as there is possibility

of leak of cystic fluid, precipitating acute and fatal anaphylaxis and risk of recurrences and dissemination due to spillage of daughter cyst.

The benzimidazole compounds – albendazole and mebendazole – have been the cornerstone of chemotherapy for cystic echinococcosis. However, it has a proven adjunctive role to surgery in cases of management of patients with recurrence and high-risk contamination. Sporadic indications of pharmacotherapy as a sole modality of treatment in cases of poor general condition of patients who cannot tolerate surgical procedures, multi organ involvement or cysts located in some inaccessible areas are noted.

Although no route other than the portal route has been implicated in humans, it is likely that systemic diffusion through the lymphatic system may play a role in cases with solitary cysts at uncommon sites.

## CONCLUSIONS

Although hydatid cyst rarely appears in the maxillofacial region, it should be considered in the differential diagnosis of benign growths especially in areas where it is endemic. In the surgical treatment of cysts of the maxillofacial region in which definite diagnosis cannot be made before the operation, great care must be taken to avoid spilling of the cystic contents. Maintenance of a high index of suspicion for this disease offers the best opportunity for a rapid and accurate diagnosis and prompt treatment.

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