

## Hydatid Cyst of Infratemporal Region – A Rare Case Report

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**Abstract:** Hydatid disease, caused by larval stage of parasite echinococcus, is considerable health problem world wide. Hydatid cyst of infratemporal region is extremely rare, even in endemic areas. A 35 year old woman presented with slowly progressive swelling in infratemporal region. At operation the cystic mass was demonstrated and histopathologic examination confirmed the diagnosis of hydatid cyst. A high index of suspicion is necessary to diagnose the hydatid cyst in an unusual location and in non-endemic areas. When incidence in maxillofacial region is rare, hydatid cyst should be considered in differential diagnosis of benign swellings of orofacial region.

### Introduction

Hydatid disease, hydatidosis, cystic echinococcus are all terms describing infections which are caused by cestodes of genus echinococcus usually echinococcus granulosus [1,2]. It is endemic in some parts of world as Mediterranean, Middle East & South American countries. Although the most commonly involved organs are liver & lung respectively any organ or tissue can be primary site of hydatid cyst. Hydatid cysts are very rarely located in head & neck region. However very few cases of hydatid cyst in infratemporal region have been reported in literature [3]. The word echinococcus originates from greek meaning “hedgehog berry” a term descriptive of gross pathology of lesion. Hydatid is also a greek word meaning “a drop of water”. This disease process probably was known to Hippocrates who described “liver.... Filled with water”. The etiological agents & its character were delineated during 17<sup>th</sup> & 18<sup>th</sup> centuries, but the complete life cycle was documented not until 19<sup>th</sup> century [4]. We report the unusual case of hydatid cyst in right infratemporal region in 35 year old woman and remind the clinician that in a slow growing cystic benign lesion of orofacial region, hydatid cyst should included in the differential diagnosis.

### Case History

A 35 year old woman from the country side was referred to the dental hospital of Al-Ameen Medical Institute, Bijapur, India with a very slow progressive swelling in right check region below zygomatic process for the past 12-15 years. According to patient the swelling was very slowly progressive & was not associated with pain or pressure sensation. Patients past personnel history revealed that she lived in close proximity with sheep & cow with agriculture being an occupation.

On examination there was moderately hard non tender swelling below the right zygomatic process causing asymmetry of face. The general health of the patient was otherwise normal. There was no cervical lymphadenopathy. Chest radiograph, blood & urine analysis were normal expect slight increase in eosinophilic count. Fine needle aspiration cytology was done and it was consistent with that the mass being benign and cystic. The mass being a benign soft tissue tumor with some degeneration, an excision of lesion was planned. Pre operative contrast C.T finding on an axial section revealed a well circumscribed calcified lesion measuring 41mm X 25mm in its greatest dimension in the infratemporal fossa region with a midline shift impinging on the surrounding structure (Fig.1).



Fig.1 Photograph of the CT scan showing well circumscribed lesion with calcifications

Calcification indicates the chronicity of the lesion. Hemifacial degloving incision was taken with combination of hemicoronal & weber-Fergusson incision under general anesthesia. Later zygomatic bone was osteotomized & rotated to explore the lesion, a cystic lesion was found & the entire cyst was carefully removed in toto. No difficulty was encountered during the surgery and recovery was uneventful. Histopathological examination: H & E stain revealed lamellated calcified capsule (Fig.2) and also exocyst, endocyst and pericyst (Fig.3) being a characteristic feature of hydatid cyst.

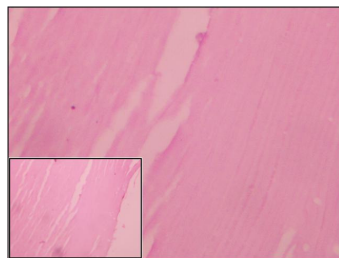


Fig.2 Photomicrograph showing Lamellated calcified capsule

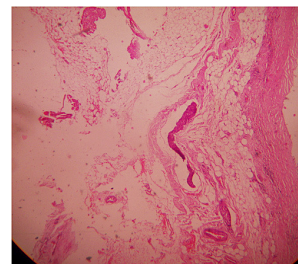


Fig.3 Photomicrograph showing exocyst, endocyst and pericyst

Part of the section also reveals calcified head of scolex along with calcific foci in the luminal side of the cyst (Fig4).

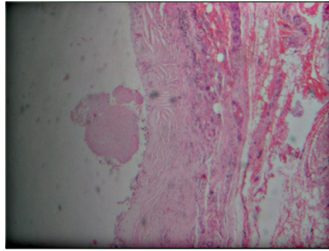


Fig.4 Photomicrograph showing calcified head of scolex

Ultrasonography of liver and an abdominal CT were done to rule out the any visceral involvement. Patient received albendazole 800 mg/day for 4 weeks. Patient has been disease free for 12 months.

### Discussion

Intestinal parasitic infections in human are rare as they are accidental host, but the occurrence of parasitic infections in the maxillofacial region are rare, accounting for only about 2% of hydatid infections of the body. Maxillofacial hydatid cysts are not usually considered in the differential diagnosis of head and neck cystic swelling, especially in non endemic area and in absence of hydatid disease elsewhere in the body. Even in endemic areas maxillofacial hydatid cysts accounts for only 1% [5]. The life cycle of *Echinococcus* involves definitive and intermediate host. Humans are accidental host and do not play a role in the biological cycle [6]. *E. granulosus* a small (3-5mm long) tapeworm that resides in the jejunum of dog (definitive host) and other canines and produces eggs that are trapped in stool. Definitive host may be infected with thousands of worms. The life cycle begins when worms in the intestine of dog produces eggs, which are then expelled in faeces of definitive host and are released to environment. These are infective to intermediate host like sheeps accidentally humans. After ingestion of eggs by the intermediate hosts through the ingestion of contaminated fruits or vegetables, digestive enzyme liberates an embryo in duodenum that passes through intestinal mucosa to portal circulation and migrates to visceral organs. As liver filter out most of larvae, most cysts occur in liver [6, 7]. Later on a fluid filled cyst develops, that differentiates into 3 layers to form hydatid cysts [6]. The wall of the cyst is composed of outer layer formed by host; inner two are formed by parasite. The inner most parasitic lining gives rise to "hydatid sands," which are brood capsules for daughter cysts in which scolices are formed [5]. Of the reported cases, 3 involved the tongue, 2 involved the cheek and very few involved the infratemporal fossa [7]. Hydatid disease is more prevalent in individual who live under poor socioeconomic conditions and have a poor health status [5, 7]. In 1938, Placitelli may have been first to report a hydatid cyst in submandibular gland, whereas Singh and Onerci et al reported submandibular gland hydatid cyst in English

literature [5]. Patients with echinococcus must undergo thorough systemic investigations because 20-30% has multiorgan involvement. As case presented with solitary occurrence of hydatid cyst has been confirmed by other authors, it is intriguing that the cyst can occur in the maxillofacial region without evidence of hepatic or lung involvement, though embryos must have passed through the organs [5]. Literature shows rural women and children are more closely associated with domestic forming duties and are more liable to be affected as in our case. Prevalence is highest in second to fourth decades of life, even though humans usually become affected during childhood. The period between the initial infection and clinical symptoms are variable. These lesions are characteristically slow growing and well tolerated and manifestation of symptom depend on location, size and pressure caused by enlarging cyst [5, 8]. Past medical history, family history, patient occupation and patient residence may suggest diagnosis of hydatid cysts in differential diagnosis, but unless suspected or demonstrated histopathological findings or radiological findings, preoperative diagnosis may be missed [8, 9]. Fine needle aspiration cytology has minimal complications and is efficient for diagnosis of hydatid cysts of soft tissues but there is controversy over use of Fine needle aspiration biopsy and is not advised because of potential to precipitate acute anaphylaxis and spread of daughter cysts [8]. In our case Fine needle aspiration cytology result was reported as benign and cystic lesions and much information about nature of mass needed further investigation. Therefore diagnosis of hydatid cyst was not considered before surgery and definitive diagnosis was made only by the histopathologic examination after surgery. The extent of calcified lamellated capsule, calcified scolex and multiple foci of calcifications may be considered as evidence of necrosis of parasite, which may explain the fact that mass was very slow growing, stable and inactive for many years. Diagnostic imaging has been greatly facilitated with Ultrasonography, CT, and MRI, serological tests like ELISA, latex agglutination, and indirect haemagglutination [1, 10]. Both false positive and false negative results after serological test are common and have low sensitivity and specificity and their use is controversial. The paramount role of serologic tests is in the follow up of treated patient for whom a drop in titre indicates resolution and rise is likely to indicate recurrence of cysts. Surgery is most effective way to treat hydatid cyst in maxillofacial region and an additional effort should be made in order to prevent life threatening anaphylaxis and recurrence [6, 7]. Inactivation of daughter cysts and scolices before surgery can be achieved by injecting 20% hypertonic saline or 5% silver nitrate into cyst [5]. Combination of medical therapy with imidazole derivatives has been used for the management of patients with recurrence and high risk contamination [1,8,10]. Although there were no signs of any other organ involvement in our case, we treated the case with albendazole (800mg/day) for 4 weeks postoperatively.

### Conclusion

In the nonendemic areas echinococcus of head and neck region is too rare to be included in the differential diagnosis of benign tumor in everyday clinical practice,

histopathological examination of surgical specimen and patient follow up seems critical in all cases in order to offer accurate diagnosis and definitive treatment.

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