Case Report

JRAD Journal of Research and Advancement in Dentistry

J Res Adv Dent 2016;5:1:365-368.

Unusual Presentation of Hydatid Cyst in Masseter Muscle: A Case Report

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ABSTRACT

Background: Hydatid cyst or Echinococcosis is a zoonotic parasitic disease caused by tapeworm that occurs primarily in sheep grazing areas. It has serious impact on health and economy in the endemic countries. Hydatid disease is most frequently caused by E. granulosus and commonly affected organs are liver and lung. It is a chronic disease and cyst can be localized in different organs. A high index of suspicion is required to diagnose it in the unusual region and non endemic areas. Incidence of hydatid cyst in head and neck region is extremely rare. This case report describes a benign swelling (Hydatid Cyst) in right cheek region of a 45 years old female patient.

Keywords: Hydatic disease, Echinococcus granulosus, Masseter muscle.

INTRODUCTION

Hydatidosis is a zoonotic disease that occurs worldwide and is most common in agricultural regions. Human echinococcosis, also known as hydatid disease, most frequently results from infestation with the tapeworm *E. Granulosus*[1]. Infection with *E granulosus* results in the formation of a unilocular hydatid cyst, whereas infection with *E multilocularis* produces multilocular disease [1]. The disease is endemic in many Mediterranean countries, the Middle East and Far East, South America, South and East Africa. Liver (60-75%) and lungs (15-25%) are most commonly involved organs. The involvement of all the other organs including brain, heart, kidney, bone, skeletal muscle, breast, thyroid gland comprises only about 10 % and are listed under unusual localization classification [2-4]. Involvement of head and neck region by Hydatid cyst is very rare, and one of the unusal presentation of hydatid cyst in Facial muscle (Masseter muscle) has been discussed here in article.

CASE REPORT

History

A 45 years old female presented with slowly progressing non tender swelling in the right cheek of 1month duration. The swelling is not associated with pain or trismus. Patient's occupation was agriculture and she gave history of close association with cow, buffalo, sheep and dog.

Clinical features

On examination the lesion was presented with the swelling in the right side of the face on the middle third of anterior border of masseter. On intra oral examination swelling was appreciated in right buccal mucosa in relation to 1st molar region in the maxillary plane of muscles. The mass was oval in shape deeply seated. It was bimanually

Received: Dec. 12, 2015: **Accepted:** Feb. 1, 2016
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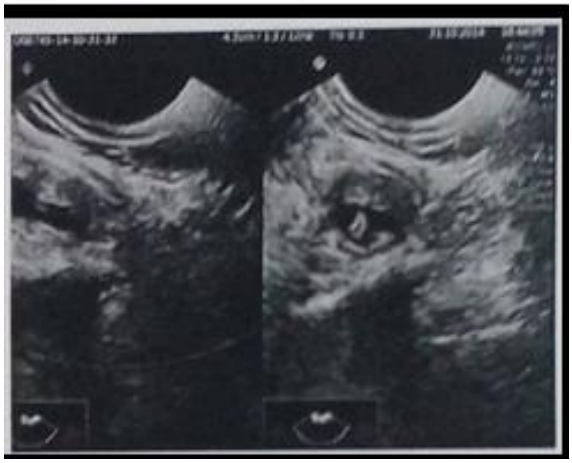
pISSN 2278-0076
eISSN 2321-9270

Fig 1: Firm cystic swelling in the right cheek region.



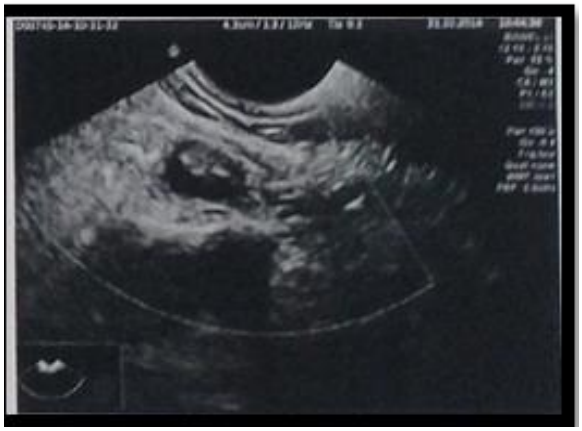


Fig 2 a & b: USG of the masseter showing well defined hypoechoic lesion.

palpable, was non-tender on palpation, and was movable and firm in consistency. Fig.1.The patient was sent for an ultrasonography.

The ultrasonographic report described a 16x16mm well defined cyst with pericyst hypoechoic thicking noted in right cheek region muscle bulk. On colour Doppler mild peripheral vascularity was noted. The impression was of benign cystic lesion in right buccal region (muscle plane) and a differential diagnosis of muscle paracytic cyst and muscle cyst was given (Fig 2). After complete examination of the patient and evaluation, the patient was further sent for a sialographic examination for further confirmation and to rule out sialocele of parotid duct which revealed no abnormality of parotid gland and duct. Fig.3

We surgically explored the site with the patient under general anesthesia through intra oral vestibular approach and blunt dissection through the masseter muscle, a cystic lesion was exposed. Accidental breakage of cystic lining revealed soft creamy yellowish membranous cystic content. Fig.4.

The histological findings revealed a cystic lesion containing degenerated parasitic larvae. The cystic cavity was surrounded by a collagenous capsule that comprised of multinucleated foreign body giant cells with admixture of lymphocytes, plasma cells and histiocytes. In histopathology, a typical hydatid hyaline ectocyst and germinative layer were confirmed. Fig 5

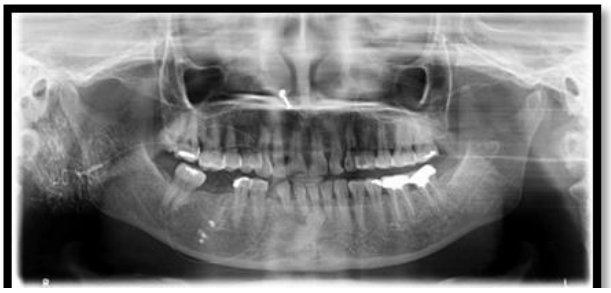


Fig 3: Sialograghy of right parotid showing no abnormality.

The patient was treated with Albendazole after the established diagnosis. Patient was recalled for regular follows up for 6 months and no nodule elsewhere has been noticed thereafter.

DISCUSSION

Hydatid cyst in the maxillofacial region is rare accounting for only about 2% of hydatid

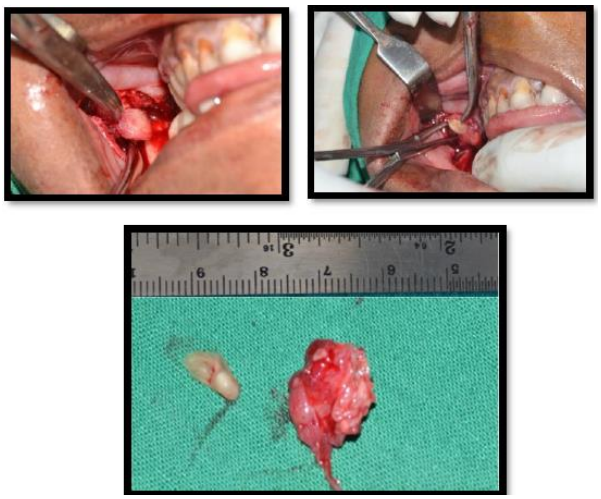


Fig 4: Clinical picture showing the location of the lesion and surgical exploration revealed a shiny capsule and yellowish cystic content.

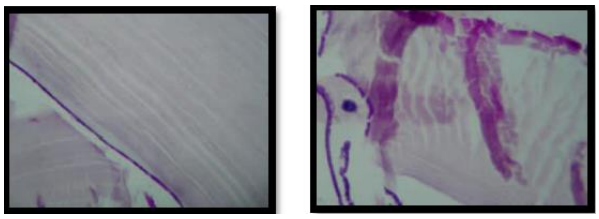


Fig 5: Hyaline ectocyst and Germinative layer.

infections of the body. Humans are accidental host and do not play a role in the biological cycle of *Ecchinococcus Granulosus*[5]. *E granulosus,* a small (3-5mmlong) tapeworm 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. These worms produce eggs that are released to environment and are infective to intermediate hosts like sheep, and accidentally, to humans. Once the eggs are ingested with 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 filters out most of the larvae, most cysts occur in the liver [5]. Later on fluid filled cyst develops, that differentiates into three 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 or daughter cysts in which scolices are formed [1]. Hydatid disease is more prevalent in individuals living under poor socioeconomic conditions and has a poor health status [1]. Georgopaulus et al for the first time reported a hydatid cyst in submandibular gland. It has been observed that a solitary organ involvement without evidence of hepatic or lung involvement occurs eventhough embryos must have passed through the organs [2]. According to literature, rural women and children are more closely associated with domestic farming duties and hence are more susceptible as in the present case.

There have been previous case reports of hydatid cysts in a variety of different parts of the head and neck area including the neck[6-9], nasopharynx[9], skull base[9,10], maxillary region[11], pterygopalatine fossa[12], and infratemporal fossa[13]. However, even in areas where the parasite is endemic, a hydatid 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[6,7]. In our case, fine-needle aspiration cytology was used to confirm that the mass was benign and cystic, and an ultrasonograghy was used to confirm its location. A diagnosis of a hydatid cyst was not considered before surgery, and a definitive diagnosis was made only by postoperative histopathology. Although we used fine-needle aspiration cytology, its use is controversial due to the risk of acute anaphylaxis or spread of daughter cysts [8]. However, several reports have indicated that it is a safe diagnostic technique if performed properly [14].

In terms of treatment, surgery is the most effective way to manage a hydatid cyst in the maxillofacial region, and particular efforts should be made to prevent breakage of the cyst during the procedure, which could lead to anaphylaxis and/or cyst recurrence [6,7,11,13]. Inactivation of daughter cysts before surgery can be achieved by injecting 20% saline, formalin, or 0.5% silver nitrate into the cyst [11]. The area around the cyst can also be protected from contamination with surgical pads soaked with a scolicidal agent (1.5% cetrimide, 1.5% chlorhexidine gluconate)[8]. Following surgery, imidazole derivatives such as albendazole should be administrated to manage any potential risk of contamination or recurrence [6,7,11] as was done in present case.

CONCLUSION

Hydatid cyst should be kept as differential diagnosis of cyst in head and neck region, especially in countries where Echinococcus infestation is endemic. A high index of suspicion is required to diagnose it in the unusual region and non endemic areas. Fine needle aspiration cytology is a good tool for diagnosis but risk of anaphylaxis is a major concern. During surgical removal of cysts great care must be taken to avoid spilling of the cystic contents. Histopathological examination of surgical specimen and patient follow up seems critical in all cases in order to offer accurate diagnosis and definitive treatment and prevent recurrence.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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