



Case Report

Dumb-Bell Shaped Hydatid Cyst in the Parotid Gland – A Rare Presentation

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Abstract

Human echinococcosis is a zoonotic infection caused by the tapeworm of the genus *Echinococcus* and transmitted by dog and other canine animals. It is a serious problem in tropical areas and is seen in many parts of Bangladesh. In children lungs are the most common site of infection, whereas in adults liver is infected most frequently. Primary hydatid cyst of the parotid gland is extremely rare, even in the endemic areas and very few cases are reported in parotid gland. The disease is prevalent in most part of the world, though it is most extensive in the sheep and cattle raising areas. FNAC is contraindicated in suspected hydatid cyst cases due to risk of hypersensitivity reaction and additional precaution of intact cyst excision is mandatory in such cases to prevent recurrence and anaphylaxis reaction. A 30 year old female patient presented with gradually increasing painless large smooth cystic swelling of the right parotid region with trismus. CT scan showed a huge dumb bell shaped cystic lesion in the superficial and deep lobe of the parotid gland that is compatible with hydatid cyst. The parotid tumor was surgically removed by midline mandibulotomy approach. Pathological study of the cystic lesion confirmed hydatid disease of the parotid gland.

Keywords: *Echinococcus granulosus*, Hydatid cyst, Parotid

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Introduction:

Echinococcosis, or Hydatid disease, is an infection caused by tapeworms of the genus *Echinococcus*, a tiny tapeworm just a few millimeters long. Five species of *Echinococcus* have been identified which infect a wide range of domestic and wild animals. Echinococcosis is a zoonosis, a disease of animals that affects humans. Like all tapeworms

the life cycle involves two animals. A carnivore is the definitive host— where the adult worms live in the intestines— and almost any mammal, including humans, can be the intermediate host - where the worms form cysts in various organs. The disease symptoms are caused by the cysts, which are slow growing fluid filled structures that contain the larvae and are most often located in the liver or lungs.

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Adult worms live in the small intestine of the definitive host. They reproduce releasing eggs into the environment in the faeces of the host animal. The eggs are well adapted to survive in the environment for as long as a year in cool moist conditions, but are susceptible to desiccation. Fresh eggs are sticky and may adhere to the fur of definitive hosts facilitating their spread. The intermediate host ingests the eggs incidentally while grazing, foraging or drinking. The eggs hatch in the small intestine, become larvae which penetrate the gut wall, and are carried in the circulatory system to various organs. There the cysts, called hydatid cysts or metacestodes, are formed. The cysts, which contain larvae, either comprise fluid filled bladders, which contain larval pre-tapeworms (protoscoleces), and cause the disease cystic echinococcosis due to *E. granulosus* or alternatively, for *E. multilocularis* a multivesiculated lesion or mass containing protoscoleces that grows rapidly by exogenous budding and causes alveolar echinococcosis in rodents and other small mammals. Though slow growing in humans and long-lived animals (e.g. camels or horses) cysts of *E. granulosus* can reach a size of 10-20 centimetres, but in sheep the size is usually 2-6 cm. The life cycle is completed when the cysts are ingested by a carnivore definitive host (e.g. dog, fox, or wolf), the larvae (protoscoleces) are released from the cyst into the small intestine, and develop into adult tapeworms that produce eggs which are released into the environment in the faeces of the host animal within 25-80 days depending on the species and strain of Echinococcus.

Case report:

A 28-year-old lady presented to the OPD of ENT and Head-Neck Surgery of Rajshahi Medical College Hospital with a large swelling in the right side of the face for 3 years. The swelling is gradually increasing in size from the beginning but there was no pain. Patient also complained of difficulty to open the mouth but food intake is not compromised. Examination reveals a large non-tender cystic swelling in the right parotid area with smooth surface and negative transillumination.

Patient has no sign of facial nerve palsy but trismus allowing only 2 fingers in between the jaws. Examination of the oral cavity and oropharynx revealed no abnormality. Systemic examination is normal.

USG examination of the swelling finds that the lesion comprises of a large complex cyst with definitive echogenic wall and almost completely replacing the parotid gland. FNAC of the lesion reveals turbid fluid and microscopic examination reveals inflammatory picture.

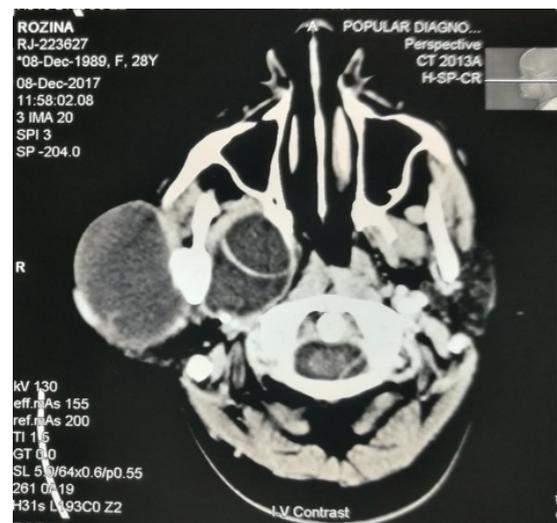




Fig 1: CT findings of the patient showing the cysts in both sides of the mandible.

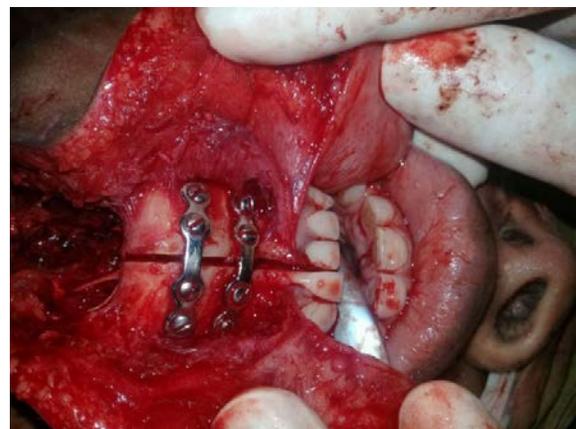


Fig 2: Per-operative pic showing cyst and midline mandibulotomy approach.

CT scan of the lesion found two different but similar cystic lesions in the right parotid gland and the parapharyngeal space separated by the angle of the mandible. The cysts are heterodense in nature

and have thick capsule. The lateral one is 12X10 cm and the medial one about 8X6 cm in size. The adjacent part of the mandible and masseter muscle is thinned but there is no definitive erosion around either the lesion though the parapharyngeal part reached to the skull base. Suspicion of hydatid cyst is raised by the radiologist.

The hematological parameter and relevant investigation were normal. ICT for *E. granulosus* was not available. Keeping the possibility of hydatid cyst in mind all the precautions were taken. Maxillofacial surgeon was consulted and included in the surgical team and hypertonic solution of sodium chloride was prepared. On exploration of the mass the cyst was found under the facial nerve and compressed the gland. Facial nerve was closely related to the capsule and hard to dissect apart from it. PAIR (puncture, aspiration, injection & re-aspiration) was done with hypertonic saline to prevent recurrence and reduction of the size. The upper buccal branch was sacrificed and the cyst was delivered intact squeezing between the two main divisions. Midline mandibulotomy was performed by the maxillofacial surgeon to approach the parapharyngeal part. This part was difficult to get access but finally dissected out with finger keeping the cyst intact. The wound was washed with hypertonic solution and closed in layers keeping drain and NG tube in situ. Postoperative recovery was uneventful and stitch was removed at 7th POD.

Histological examination of the cyst reveals hydatid cyst. Multiple section from cyst was taken, processed and stain with hematoxyline and eosin and examined under microscope. Histopathological findings confirm the diagnosis. Section shows parotid gland parenchyma with hydatidcyst composed of chitinous laminated wall (ectocyst), endocyst having brood capsule and scolices. The outer pericyst consists of parotid gland parenchyma and fibrous tissue.

Discussion:

The parotid gland hydatid cysts are always primary. Only a few cases are reported in English

making it difficult to understand. Preoperative clinical diagnosis of parotid swelling as a hydatid cyst is challenging due to its rarity and mimicking imaging with any benign cystic lesion like Warthins tumor or pleomorphic adenoma and the management of both conditions is different. Ultrasound, the diagnostic tool of hydatid disease may be less informative in unusual locations .FNAB is to be avoided on suspicion of hydatid disease due to risk of hypersensitivity or recurrence & spread by seeding daughter cysts. Other immunologic test like IgG ELISA, IFAT also recommended for confirmation of hydatid disease with 90% specificity and sensitivity for liver & 85% for lungs. Unusual sites data are not available. We did not perform this test because of highly suggestive diagnostic CT images & not availability of immunological tests. During surgery, it is mandatory to excise the cyst intact for same reasons. Spillage of cyst content may stimulate hypersensitivity chain reactions and anaphylaxis. PAIR (puncture, aspiration, injection & re-aspiration) also recommended for huge unilocular cyst to reduce risk of recurrence after surgery as was done in our case. In small to moderate lesion, surgery is optimal management with intact total excision. Prophylactic anti-parasitic treatment is also recommended in such cases before surgery though not mandatory. Because the route of drug to cross & reach the inside the cyst is not specified but it may be useful to eradicate the organism in circulation, if any. Albendazole course for next 3 months in all cases is recommended.

Conclusion:

Though cystic lesion of the parotid gland is a common presentation, the possibility of hydatid cyst is a rarity. The only precaution during surgery of any undiagnosed cystic lesion of the parotid gland should be to remove the tumour intact to avoid spillage which is a well practiced surgical habit.

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