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CASE STUDY

HYDATID CYST OF SUBMANDIBULAR GLAND

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ABSTRACT

Hydatid cyst is a parasitic infection of the human caused by the larval stage of Echinococcus. Hydatid cyst of the head and neck region is uncommon and the involvement of salivary glands, especially the submandibular gland is very rare. We report a case of isolated hydatid cyst of left submandibular gland. An 11-year-old female presented with a slowly growing painless swelling of left submandibular region since 4 months. Fine needle aspiration cytology yielded fluid aspirate and smear showed fragment of laminated membrane, suggestive of HC. FNAC was suggestive of lymphatic cyst of left submandibular region .Ultrasonography of neck was suggestive of a cystic mass with internal debris. The cyst was excised as a whole along with submandibular gland and it was sent for histopathological examination. Pathological examination confirmed the swelling to be a hydatid cyst of the left submandibular gland.

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INTRODUCTION

Echinococcosis or hydatid disease is a zoonotic infection caused by the tapeworm parasite Echinococcus granulosus. It is common in the temperate zones, including the Mediterranean countries, the Middle East, India, Africa, South America, New Zealand, Australia, Turkey and Southeast Asia. It has been recognized by humans for centuries, as it was described by Hippocrates more than two thousand years as a "fluid-filled liver", and the famous Arabian physician Al-Rahzes described it, but it took till the 17th century when Francesco Redi illustrated that the hydatid cysts or echinococcosis were of animal origin. The primary hosts are dogs. Intermediate hosts are sheep, cattle, horses and occasionally man. Occurrence of hydatid cyst is extremely rare in the head and neck region even in geographical areas where echinococcal infestation is frequent. Here we report a rare case of hydatid cyst localised to the submandibular region.

Case report

An 11-year-old female presented to our ENT Outpatient department with chief complaints of slowly enlarging painless swelling of left submandibular region of 4 months duration. The general condition of the patient was good; he had no history of fever or weight loss. No history of cough, chest pain, haemoptysis or jaundice was reported, and examination revealed the patient to be afebrile. Clinical examination revealed a well-defined, soft, non-tender, mobile mass measuring 6 × 5 cm approx.in left submandibular region with positive fluctuation on bimanual palpation. (Fig.1, 2) The skin overlying the mass showed no signs of redness, tenderness or local rise of temperature. Intraoral examination revealed no stones in the left submandibular gland duct. Complete blood count and sedimentation rate were normal and showed no eosinophilia. Liver function tests and coagulation profile was normal. The tuberculin test (PPD) was also normal. FNAC of the submandibular swelling was performed but it did not reveal any features of hydatid cyst. Ultrasonography of the left submandibular area revealed a well-defined fluid filled cystic mass of size 4.6-5 cm with a solid component. No daughter cysts or calcification of the wall was noticed with mild enlargement of left submandibular gland. Chest radiograph and abdominal ultrasonography results were normal. After ultrasound examinations, patient was evaluated on a computed tomography (CT). CT scan of the neck showed a well-defined large cystic lesion with sharp and thin margins involving the left submandibular space, of about 4.5–5 cm in size. The mass was displacing the left submandibular gland supero-medially.
With the impression of a cystic mass of the submandibular gland we planned for excisional biopsy and consent of the patient was taken. Surgery was performed by a horizontal incision about 4 cm below the lower border of angle of mandible. After dissection of the surrounding tissue, the submandibular gland along with the cyst was completely

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exposed. A whitish colour cyst of size 5 x 4 cm was dissected from the superficial lobe of submandibular gland and was removed without rupturing the wall of the cyst. (Fig. 3) On gross examination, submandibular gland was enlarged with features of chronic sialoadenitis. Hence, we excised the entire gland (superficial and deep lobes) and both cyst and the submandibular tissue was sent for histopathological examination. The postoperative period was uneventful. The patient was discharged from the hospital without any complications. The pathological report showed a hydatid cyst in the left submandibular region which revealed portions of laminated membrane. (Fig.4, Fig.5) Albendazole (10 mg/kg day) was administered orally to the patient during discharge from the hospital for a duration of 2 months. Periodic check-ups, including abdominal ultrasonography and chest x-ray examination, were performed during the first 6 months follow up which revealed no pathological findings.

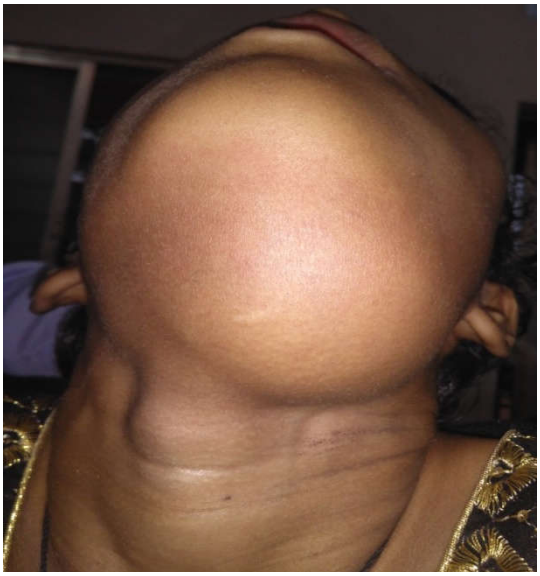

Fig. 1.

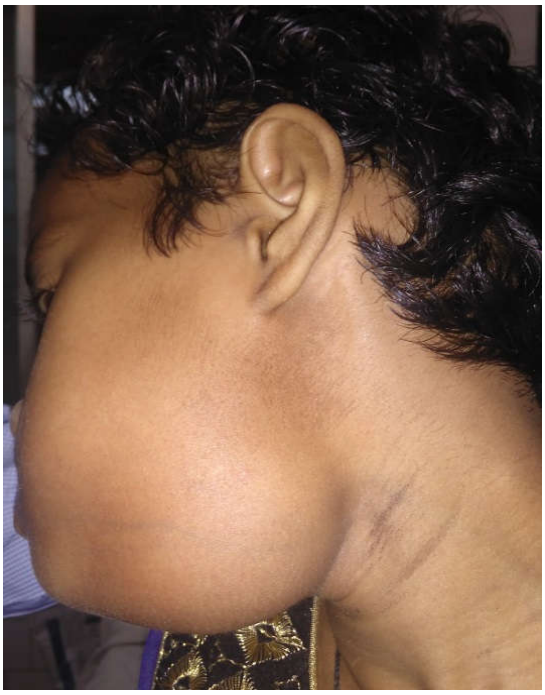

Fig.1. and Fig. 2. Cystic swelling of left submandibular gland

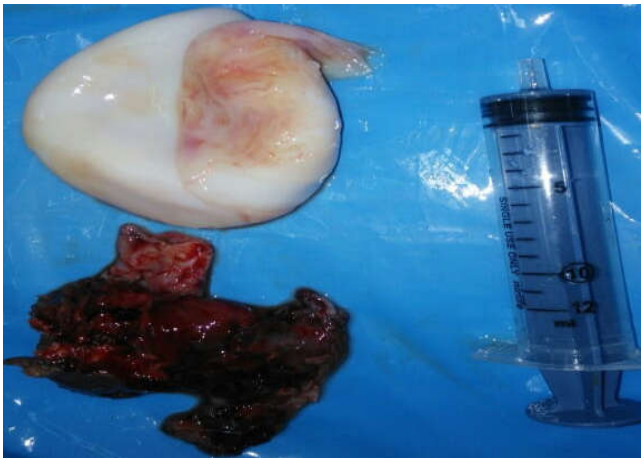

Fig. 3. Whitish colour cystic mass (gross specimen)

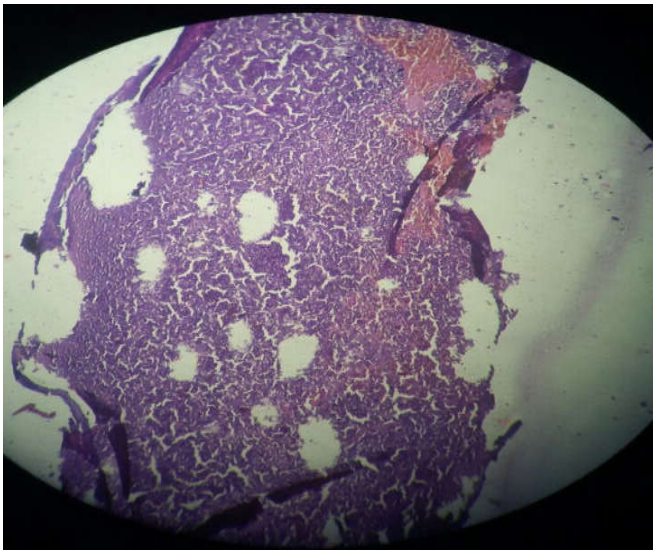

Fig. 4. Histologic section of submandibular gland showing hypertrophied glandular tissue with scattered inflammatory cells

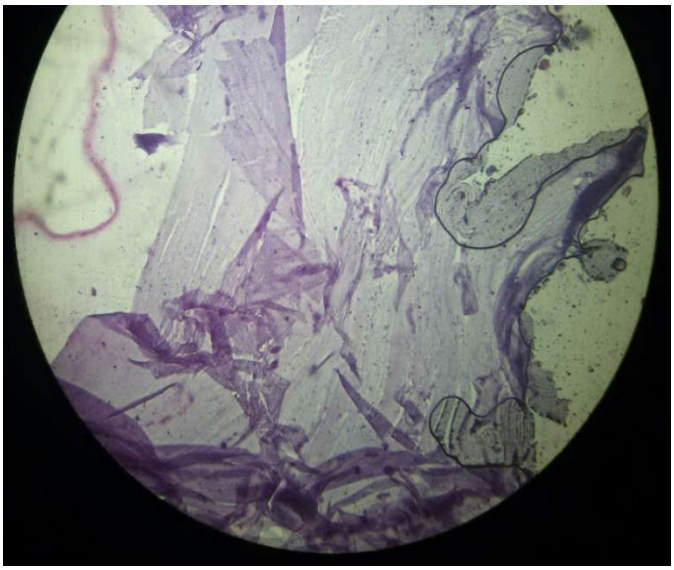

Fig. 5. Histologic section of Hydatid cyst showing bands of eosinophillic laminated membrane

DISCUSSION

Hydatid cyst (Hydatosis or Echinococcosis) is a zoonotic infection caused by the cestodes (tape worm) of genus

Echinococcus granulosus, E. multilocularis and E. oligarthus. The unilocular cystic form is caused by E. granulosus is far more common than the rare multilocular form caused by E. multilocularis. (Dahniya *et al*., 2001) Hydatid disease affecting human beings is a parasitic infestation caused by Echinococcus granulosus. (Dahniya *et al*., 2001; Engin *et al*., 2000) The definitive hosts are dogs, wolves and foxes, while intermediate hosts are sheep and horses. Humans are accidental intermediate hosts and do not play any role in the biological cycle. (Nandy Manab *et al*., 2012) This disease is endemic in cattle and sheep rearing regions of the world. Food contamination by canine feces containing ova of Taenia echinococcus is more likely to occur among people living in close association with animals. (Nandy Manab *et al*., 2012) On ingestion of such contaminated food by humans leads to the hatching of ova in the gastrointestinal tract. Most of the ova pass into the portal venous system or lymphatic system and form hydatid cyst in the liver or lung. Some ova can cross the hepatic sinusoids and pulmonary capillary barrier and escapes into the general circulation to involve the brain, kidneys, bones and other tissues. (Engin *et al*., 2000; Nandy Manab *et al*., 2012) It develops most frequently in liver (65%), lungs (25%) and 10% occurs in muscle, spleen, bones, kidneys, brain, eye, heart, and pancreas. (Engin *et al*., 2000; Nandy Manab *et al*., 2012; Önerci *et al*., 1991) Hydatid cyst of head and neck is uncommon even in the areas where the disease is endemic. (Nandy Manab *et al*., 2012) Multi-organ involvement is seen in 20–30% of the cases with involvement of the liver in all cases. (Beggs, 1985) There are very few such cases reported in the literature involving the submandibular glands. (Beggs, 1985; Iynen *et al*., 2011) The clinical signs and symptoms depend on the anatomic location, size and mechanical effect of growing cysts. They usually presents as a slowly growing, painless, fluctuant, cystic mass. (Iynen *et al*., 2011) Symptoms develop when the cyst enlarges into a space-occupying lesion and after allergic reactions to the cyst fluid. (Darabi *et al*., 2009) Majority of HCs of neck are misdiagnosed on clinical examination as the signs and symptoms are variable like in our case. HCs may be single or multiple, unilocular or multilocular, and thin or thick walled. (Sultana *et al*., 2012)

The diagnosis can be confirmed after serological tests, imaging techniques and pathological examinations. Different serological investigations like ELISA, latex agglutination, direct hemagglutination, skin test (Casoni's test) may be of use in diagnosis. These tests are highly sensitive (80–100 %) and specific (88–96 %) for liver HC, it is less sensitive for lung (50–56 %) or other organ (25–56 %) involvement.[9,10] Ultrasonography, CT scan, and magnetic resonance imaging are useful for delineating the location of the cyst, but the findings are nonspecific. Daughter cysts, vesicle, internal septae, membrane detachment and wall calcifications may be seen during imaging. (Polat *et al*., 2003) Fine needle aspiration cytology (FNAC) is a useful diagnostic tool for evaluation of a neck mass. But it is not recommended in suspected case of HC due to possibility of acute anaphylactic reaction. In the present case, FNAC and USG finding was suggestive of unilocular cystic lesion of left submandibular region and exact aetiology could not be ascertained. In our case, hydatid cyst involved the submandibular gland and this possibility should be borne in mind when a submandibular mass is seen. Differential diagnosis like lymphatic cyst, retention cyst, abscesses, empyema's, tubercular mass, hematoma, lipoma, benign or malignant cystic tumours should be considered. (Önerci *et al*., 1991; Iynen *et al*., 2011) Paraclinical work up of our case did not confirm hydatid cyst. Final diagnosis of hydatid cyst was made after histopathological examination. (Tekin *et al*., 2004; Dziri *et al*., 2009) The microscopy shows three layers of the cyst wall. The outer layer, known as pericyst, is a rigid protective layer which represents the host response to the parasite. The middle layer is white acellular laminated membrane. The inner germinal layer is thin and translucent. (Dziri *et al*., 2009) The cystic space consists of jelly-like matrix, mostly it is sterile but occasionally it may contain protoscolices. Multilocular HC consists of numerous small spaces or cavities, separated from each other by connective tissue. Surgical excision remains the standard treatment of choice for a hydatid cyst in submandibular glands since they do not respond to oral or systemic medications. (Iynen *et al*., 2011; Darabi *et al*., 2009; Sultana *et al*., 2012; Chevalier *et al*., 1994) However, intraoperative and postoperative complications, and high recurrence rates are not rare. Successful percutaneous treatment (PAIR Technique) of liver, kidney, pulmonary, orbital, and parotid gland hydatid cysts has been reported in the past. (Aletras and Symbas, 2000; Akhan *et al*., 1998) The cyst should be excised as a whole without being ruptured to prevent recurrence or anaphylaxis. Inactivation of daughter cysts and scolices before surgery is achieved by injecting 20 % hypertonic saline, 5 % silver nitrate or formalin into cyst. Postoperative medical treatment with benzimidazole derivatives (albendazole, mebendazole) is frequently combined with surgery to prevent recurrence and high risk contamination. (Gossios *et al*., 1997)

Conclusion

Prolonged follow-up is required to determine the eventual outcome following surgical excision. We report this case for its unique presentation and unusual location. Imaging modalities, although sensitive, cannot make the exact diagnosis of HC. Thus, clinicians as well as radiologists should consider HC in differential diagnosis of cystic swellings in the head and neck regions.

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Declaration

- Ethical committee approval and consent of the patient has been taken.
- No Conflict of interests regarding publication of this article.
- No financial support from any organisations.

REFERENCES

Akhan O, Üstünsöz B, Somuncu İ *et al*. 1998. Percutaneous renal hydatid cyst treatment: long-term results. Abdo Imaging, 3:209-213

Aletras H, Symbas N. Hydatid disease of the lung. In: Shields TW, LoCicero J, Ponn RB, editors. 2000. General thoracic surgery. 5. Philadelphia: Lippincott Williams and Wilkins; pp. 1113–1122.

Beggs I. 1985. The radiology of hydatid disease. *AJR*, 145:639-648.

Chevalier X, Rhamouni A, Bretagne S, Martigny J, Larget-Piet B. 1994. Hydatid cyst of the subcutaneous *tissue* without

other involvement: MR imaging features. AJR, 163:645-646.

Dahniya MH, Manna RM, Ashebu S *et al*. 2001. The imaging of hydatid disease at some unusual sites. *Br J Radiol*., 74; 283-289.

Darabi M, Varedi P, Mohebi AR, Mahmoodi S, Varedi P, Nabavizadeh SA, Erfan A, Ostadali Makhmalbaf A, Saedi D, Saadat Mostafavi SR, Mousavi SM. 2009. Hydatid cyst of the parotid gland. *Oral Maxillofac Surg*., 13:33–35. doi: 10.1007/s10006-008-0138-0.

Dziri C, Haouet K, Fingerhut A, Zaouche A. 2009. Management of cystic echinococcosis complications and dissemination: where is the evidence? *World J Surg*., 33:1266–1273. doi: 10.1007/s00268-009-9982-9.

Engin G, Acunaş B, Rozanes İ, Acunaş G. 2000. Hydatid disease with unusual localization. *Eur Radiol*., 10:1904-1912.

Gossios KJ, Kontoiyannis DS, Dascalogiannaki M, Gourtsoyiannis NC. 1997. Uncommon locations of hydatid disease: CT appearances. *Eur Radiol*., 7:1303-1308.

Iynen I, Sogut O, Guldur ME, Kose R, Kaya H, Bozkus F. 2011. Primary hydatid cyst: an unusual cause of a mass in the supraclavicular region of the neck. *J Clin Med Res*., 3:52–54.

Nandy Manab, Chakrabarti Abhiram, Mallik Sudesna. 2012. Hydatid disease presenting as multiple cystic swelling in the right supraclavicular region. *Asian Pac J Trop Dis*., 2:490–491. doi: 10.1016/S2222-1808(12)60108-X.

Önerci M, Turan E, Ruacan S. Submandibular hydatid cyst. J Cranio-Max-Fac Surg 1991; 19: 359–361.

Polat P, Kantarci M, Alper F, Suma S, Koruyucu MB, Okur A. 2003. Hydatid disease from head to toe. Radio Graphics. 23:475–494.

Sultana N, Hashim TK, Jan SY, Khan Z, Malik T, Shah W. 2012. Primary cervical hydatid cyst: a rare occurrence. *Diagn Pathol*., 7:157. doi: 10.1186/1746-1596-7-157.

Tekin M, Osma U, Yaldiz M, Topcu I. 2004. Preauricular hydatid cyst: an unusual location for echinococcosis. *Eur Arch Otorhinolaryngol*., 261:87–89. doi: 10.1007/s00405-003-0650-7.

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