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Hydatid cyst of parotid gland mimicking as Branchial cyst in a 7 year child: A cytological impression

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Abstract

Hydatid disease also known as Echinococcosis is a parasitic infection caused by the cestodes of *Echinococcus granulosus*. Hydatid cyst of the head and neck region is uncommon and the involvement of parotid gland is very rare and few cases have been reported in literature. We report a rare case of hydatid cyst in a 7 year old female child who presented to the otorhinolaryngology outpatient with gradually progressive swelling on left parotid region for 3 years. The probable clinical and radiological differentials were 1st branchial cleft cyst, lymphangioma and retention cyst. However aspiration cytology revealed multiple scolices, hooklets, and granular necrotic debris which confirmed the diagnosis of hydatid cyst. CT scan of the abdomen and thorax did not reveal any other organ involvement by the disease process. Chemotherapy with albendazole was started and surgery was performed after 1 month. This case is reported here owing to its rare and unusual presentation.

Keywords: Fine needle aspiration cytology, Hydatid disease, Parotid gland

Introduction

Echinococcosis is a zoonotic disease caused by parasites of genus *Echinococcus*. Cystic echinococcosis and alveolar echinococcosis are the two main forms of this disease in humans caused by *Echinococcus granulosus* and *Echinococcus multilocularis* respectively. Herbivores and omnivores serve as intermediate hosts of *Echinococcus* by ingesting the parasite eggs in contaminated food and water and harbouring the larval stage of the parasite in their viscera. Humans are often the accidental hosts as they get infected by ingesting the contaminated food and water similar to intermediate hosts but are dead end to the cycle of transmission.

Echinococcosis is distributed globally and is endemic in parts of Argentina, Peru, East Africa, China and South Asia including India [1]. Rural and farm raising areas have highest prevalence rate making it a serious medical and public health problem. Human infection with *E. granulosus* leads to development of one or more hydatid cysts in various organs of the body. Most commonly it is seen in liver (65-70%), lung (5-15%) and rarely other parts of the body. Hydatid cyst of maxillofacial region is very rare and the cysts occurring in salivary gland is extremely rare even in endemic areas and only few case reports have been found in the literature [2]. We are reporting this case to emphasize the role of fine needle aspiration cytology (FNAC) in the early diagnosis of primary hydatid cyst in parotid gland.

Case report

A 7-year-old female child born in a farmer family presented to surgical outpatient clinic with the complaint of gradually progressive left parotid swelling for 3 years. The swelling was painful at onset with no history of fever and discharge. Local examination revealed a 2 x2 cm cystic swelling, fluctuant and non-tender with no regional lymph node involvement. Rest of the systemic examination was normal. On ultrasound a well-defined round to oval anechoic lesion with posterior acoustic enhancement was seen with provisional diagnosis of 1st branchial cleft cyst, lymphangioma and parotid retention cyst were given.

Fine needle aspiration of the swelling by a 22 gauge needle yielded 10 ml of watery fluid which was later centrifuged and stained. Hematoxylin and eosin and pap stained cyto-smears demonstrated multiple scolices with radially arranged hooklets, foamy macrophages and lymphocytes in a necrotic background consistent with the diagnosis of hydatid cyst. [Fig 1, Fig 2] The patient was further investigated by ultrasonography (USG) of the abdomen and

chest radiograph, which were normal. Preoperative chemotherapy with albendazole was started at a dose of 10 mg/kg/day for 1 month, after which the size of the cyst decreased.

The patient was operated after 1 month. Aspiration of the cyst was performed after injecting 20% saline and the surrounding tissue was packed with mops soaked in 3%. Complete enucleation of the cystic mass was performed with no spillage of fluid and was sent to histopathology lab.

Microscopically multiple sections showed chitinous laminated wall (ectocyst) and fibrocollagenous tissue. [Fig 3] The postoperative course was uneventful and she was discharged in a satisfactory condition after 7 days with a 4-week course of postoperative albendazole to prevent recurrences. The progress of patient follow-up was smooth. Owing to the rarity of the site and such rare clinical presentation of hydatid disease, this case is being reported here.

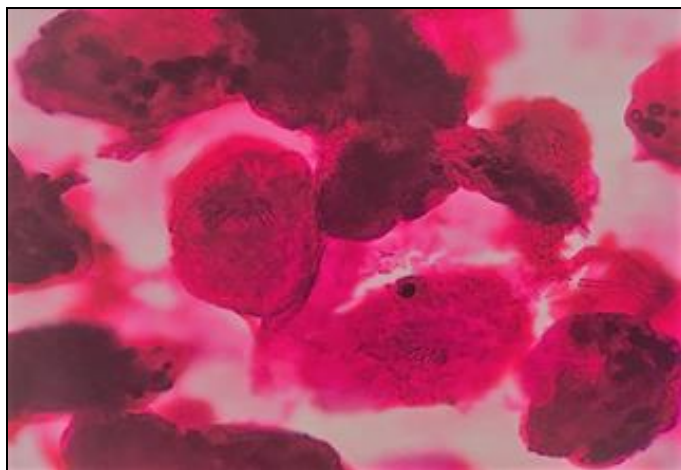


Fig 1: Smear shows multiple Scolices with radially arranged Hooklets. (H&E, 40X)

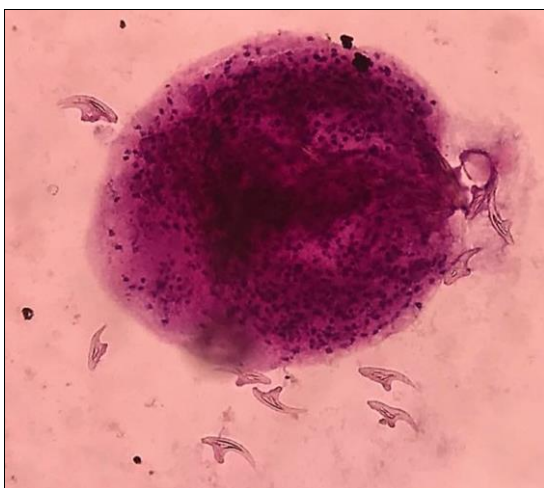


Fig 2: Pap smear shows a single scolex with Refractile dagger shaped hooklets (Pap, 40X)

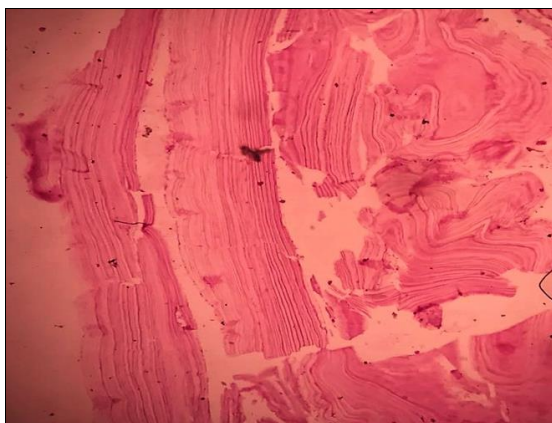


Fig 3: Histopathology shows the characteristic laminated membrane (Ectocyst). (H&E×10X)

Discussion

Echinococcosis is a unique parasitic disease that is endemic in many parts of the world and is an important public health problem in India, especially in rural and cattle raising areas. The most common locations of the hydatid cyst, after the lung and liver, are the central nervous system, orbit, musculoskeletal system, cardiovascular system, kidney, and urinary tract. The hydatid cyst of the parotid and submandibular gland is very rarely reported [3, 4]. Hydatid cysts are usually not considered in the differential diagnosis of head and neck cystic swellings, especially in non-endemic areas in the absence of hydatid disease elsewhere in the body. The rarity of the disease in this anatomical location presents a diagnostic difficulty for the physician if he or she is not familiar with the disease [5]. Fine needle aspiration cytology (FNAC) is a useful technique for identification of these organisms, including in cases where there is a low index of suspicion for this lesion. The patients must undergo complete systemic examination, especially to check for any hepatic or pulmonary lesions and other sites of involvement [6].

Imaging studies such as ultrasound (USG) is the method of choice for searching for the pathognomonic criteria of the hydatid cysts [7]. Serological tests such as enzyme-linked immunosorbent assay for Echinococcus can be used for the preoperative diagnosis of hydatid disease [8]. CT and MRI greatly facilitate the diagnosis and are more sensitive than serological studies [9]. But the diagnosis was missed in our case. Therefore, FNAC of the swelling was done. Performing aspiration on the cyst for diagnosis has often been discouraged because of its potential for acute anaphylaxis or dissemination of the disease. However, some argue that no sequelae were observed that could be attributable to aspiration if done for the diagnosis of hydatid cyst [10]. No urticarial or anaphylactic reactions occurred in

our case and the H and E stained smear on unraveled spectrum of cytomorphological features of hydatidosis, including presence of scolices, hooklets and granular necrotic debris consistent with the diagnosis of hydatid cyst.

Traditionally, the treatment of choice for hydatid disease is surgery combined with chemotherapy using albendazole and/or mebendazole before and after surgery, albendazole is preferred twice a day for 1–5 months^[11]. However in the recent literature, there are several reports on percutaneous treatment of abdominal and pulmonary hydatid cyst as an alternative to surgical treatment^[12]. They reported lower rates of morbidity and mortality and less recurrence^[12].

To sum up, this case reinforces value of FNAC as an extremely useful, cost effective and safe technique in diagnosing hydatidosis, especially at unusual location like parotid, where there is a low index of suspicion by physician. Identification of spectrum of cytological features on aspirates, as noted in the present case can obviate need for a biopsy confirmation.

Conclusions

Hydatid disease should be included in the differential diagnosis of slow growing cystic lesions of parotid gland. A high index of suspicion is needed to avoid any complication and prompt management. To sum up this case reinforces the value of FNAC as cost-effective, safe and useful technique in the diagnosis of hydatidosis, thus obviating the need for biopsy confirmation.

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