Hydatid cyst in submandibular salivary gland

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Abstract This case of 52-year male with swelling in the left submandibular area for 1 year came to ENT OPD of Patna medical college, Patna. Clinical examination showed swelling to be multiple nontender cysts with enlarged submandibular salivary gland. CT-scan revealed large well-defined encapsulated multicystic lesions in the left submandibular gland. FNAC was suggestive of aspirate to be ectopic to that area. Excision biopsy was done. HPE showed features suggestive of parasitic infestation along with chronic sialadenitis. Gross picture and investigations were highly suggestive of parasitic infestation to be primary hydatid cyst of the gland. Scanning for other areas like liver and lungs done, no lesion was found.

Keywords Submandibular gland · Hydatid cysts · Echinococcus

Key message So far only one case of primary hydatid cyst in submandibular gland is reported by Onercei M, et al [1] making this case worth reporting.

Introduction

Hydatid cyst in submandibular salivary gland is very uncommon finding. Hydatid cyst is parasitic infestation by tape-worm of genus *Echinococcus*. It is endemic in Asia. Theoretically, hydatid cyst can occur anywhere in the body. But the most common sites remain liver and lungs [1]. Primary hydatid disease in submandibular salivary gland is a very rare entity [1, 2]. It is a disease transmitted to humans from dogs by exposure to food and water contaminated by the feces of the latter or by hand to mouth by close contacts of dogs and leads to Echinococcosis. In humans, worm is in larval form. It presents as cystic swelling. Cyst contains clear fluid, which is highly anaphylactic, if it leaks inside the body.

A 52-year-male patient came to the ENT OPD of Patna Medical College, Patna, with complaints of swelling in the left submandibular area for 1 year. For the last 6 months, swelling was increasing in size, which alarmed the patient. He tried some medication but of no value. He came to the OPD for the management of the same.

History was taken, which revealed that the mass was painless and was gradually increasing in size, unnoticeable until 6 months back, when it increased in size little rapidly. He also had some pricking sensation, which would aggravate when he scratched or had shaving done in that part.

Examination findings

There was a swelling 6×3 cm in size, ovoid in shape having well defined margin with normal appearance of overlying skin and beard in the left submandibular area. Temperature was not raised. Mass was nontender. On deeper palpation multiple cystic lesions were found. Fluctuation, translucency, reducibility all were negative.

Investigations

Complete Hemogram, ESR, Blood sugar (F) and (PP), ELISA for Koch's IgM and IgG, ELISA for HIV 1 & 2,

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R/E of Urine, FNAC of the mass, CT scans of the submandibular area, USG of the liver, X-RAY chest PA-view.

Positive findings of investigations were:

- Eosinophil count – 21%
- FNAC - aspirated material shown to be ectopic to that area. It did not match with the parent tissue contents.
- CT –scan revealed, 6×3 cm large well-defined encapsulated multicystic lesion in the left submandibular area. Margin of lesion appeared hyper dense but did not show contrast enhancement. (Resemblance with daughter cysts). There was no lymph node enlargement on either side of the neck. No evidence of bony destruction. Strap muscles appeared normal (Fig. 1).

At that point of time, picture was not very clear. So to confirm the diagnosis, excision and biopsy of the mass was planned under general anesthesia.

Operative findings

Skin incision given and with dissection, surface of gland was reached. While the gland was being freed from the surrounding structures and dissected, cheesy material started coming out from the gland. With further manipulation it burst open and pale, gelatinous shell like structure came out from within the gland. When incised and examined, it was found to be filled with small glistening white cysts with pearl like appearance. Cysts were intact and did not

burst. Submandibular gland was dissected out. Hemostasis was secured, drainage was given and wound was closed in layers.

The gland was examined thoroughly outside the body. On posterior surface similar shell like structure was found. When it was split opened, two clear fluid filled cysts of bigger size about 2×2 cm dimension were found (Fig. 2).

Gross appearance resembled very much with hydatid cyst and suspicion raised in our minds towards that. Biopsy material was sent for gross and microscopic histopathological examination. Postoperative period was uneventful. Patient was discharged after removal of stitches.

Much to our surprise and satisfaction his biopsy report revealed lesion to be chronic sialoadenitis with parasitic infestation. We discussed the case with pathologist keeping before them the operative findings and gross appearance of the lesion. They too gave opinion that parasitic infestation to be none other than hydatid cyst. Patient's subsequent follow-ups are normal.

Discussion

Hydatid cyst is caused by the larval tapeworms *Echinococcus granulosus*, *E. multilocularis* etc. *E. granulosus* is the most common cause.

For diagnosis, different tests like FNAC showing protoscolex in aspirate, serum immunoelectrophoresis that gives positive results for *Echinococcus* and HPE showing scolices and protoscolices of parasite. CT scan consistent with cystic lesions is important. But at the same time history, clinical picture, preoperative findings and gross appearance of lesion also contribute a great deal in making the diagnosis. That is emphasized by the facts that no serological

Fig. 1 CT scan of submandibular area

Fig. 2 Showing clear fluid filled hydatid cysts

test is highly specific for hydatid cyst in humans. Lack of protoscolex in aspirated fluid does not rule out the diagnosis [4].

The sensitivity of serological test is low especially in the cases of extrahepatic hydatidosis (only 50%) [5]. Eosinophil count is increased in only 25% of all infected cases, while hypogammaglobulinemia is present in only 30% of cases [3].

In plain X-ray, a thin rim of calcification delineating a cyst is suggestive of hydatid cyst [3]. CT-scan has an accuracy of 98% to demonstrate the daughter cysts [3].

Results

In our case the points in favour of the diagnosis were raised eosinophil count, CT scan findings, gross appearance of mass and FNAC and Histopathological picture. Low socioeconomic group and alcoholic habit of the patient were suggestive of poor personal hygiene, which in turn favoured the diagnosis. Screening of other areas like liver and lung, showed the lesion to be primary for submandibular region.

Conclusion

To conclude, hydatid cyst is not the common differential diagnosis of extrahepatic and extrapulmonary cystic lesions, but high degree of suspicion can lead to its diagnosis.

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