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Submandibular hydatid cyst caused by *Echinococcus oligarthrus*

J. K. Sahni, M.S., M.N.A.M.S., M. Jain, M.D.*, Yogesh Bajaj, M.S., D.N.B., Virender Kumar, M.S., Anubhav Jain

Abstract

An unusual case of *Echinococcus oligarthrus* infestation of the submandibular salivary gland is reported. *Echinococcus oligarthrus* is a rare variant of the *Echinococcus* species affecting humans. To the best of our knowledge only one case of submandibular hydatid cyst caused by *Echinococcus oligarthrus* has been reported.

A 28-year-old female patient was admitted with a progressively increasing swelling in the left submandibular region of four years' duration. There was no pulmonary or hepatic involvement. The present case of submandibular hydatid cyst caused by *Echinococcus oligarthrus* is of interest because of the unusual site of the disease.

Key words: Submandibular gland; Hydatid cyst; Echinococcus

Introduction

Hydatidosis of the head and neck region is uncommon even in countries where *Echinococcus* infestation is high.[1] The present case of submandibular hydatid cyst is of interest because of the unusual site of the disease and the unusual causative organism i.e. *Echinococcus oligarthrus*, a rare variant of the *Echinococcus* species. This appears to be the second case report of submandibular hydatid cyst caused by *Echinococcus oligarthrus* in the world literature. Three other cases of hydatid cyst of the submandibular gland caused by *Echinococcus granulosus* have been reported.

The dog is the primary host in echinococcal infestation and intermediate hosts are sheep, cattle, horses and occasionally man. The adult worm is provided with a scolex and four suckers and has 30–50 hooklets. When the eggs are swallowed by a suitable intermediate host they hatch in the duodenum, then migrate through the intestinal capillary filter to various organs and tissues. The overwhelming majority of the embryos are retained in the capillary systems of the liver and lungs, but a small minority escape to pass to other organs.

Case report

A 28-year-old female presented with swelling in the left submandibular region of four years' duration. To begin with the patient had pain in the left submandibular region along with high grade fever for a few days. Immediately after this episode, the patient noticed a small swelling at the same site which progressively increased in size over the next four years. There was no history of abdominal pain, urticaria, cough, chest pain, haemoptysis or any swelling elsewhere in the body. She had no contact with any animals.

Examination revealed an afebrile patient with a firm swelling in the left submandibular region, 4 cm × 5 cm in size. The overlying skin was normal and showed no

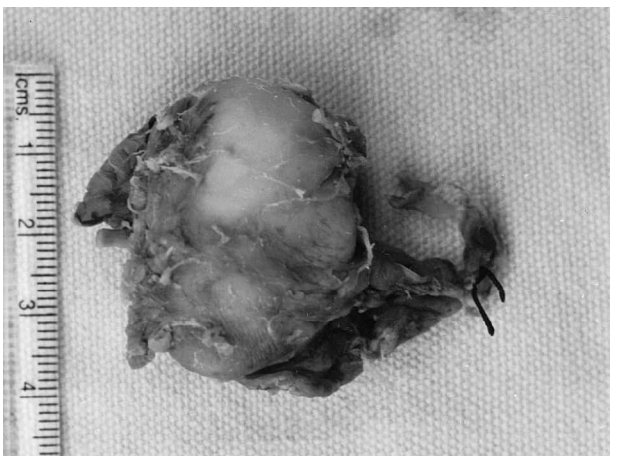

Fig. 1
Specimen of submandibular gland with hydatid cyst.

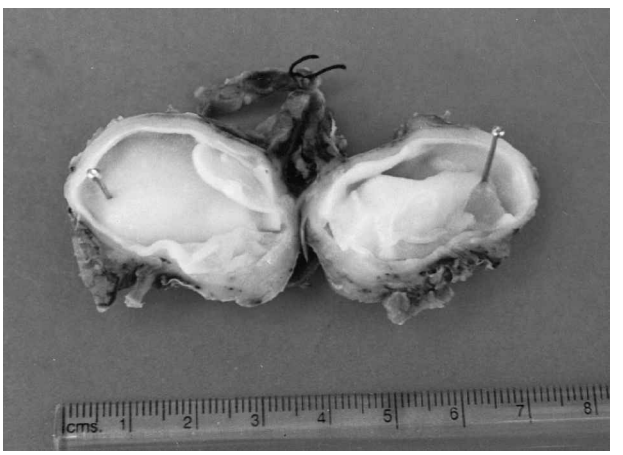

Fig. 2
Submandibular gland cut open, showing hydatid cyst.

From the Department of Otolaryngology and Pathology*, Lady Hardinge Medical College, New Delhi, India.
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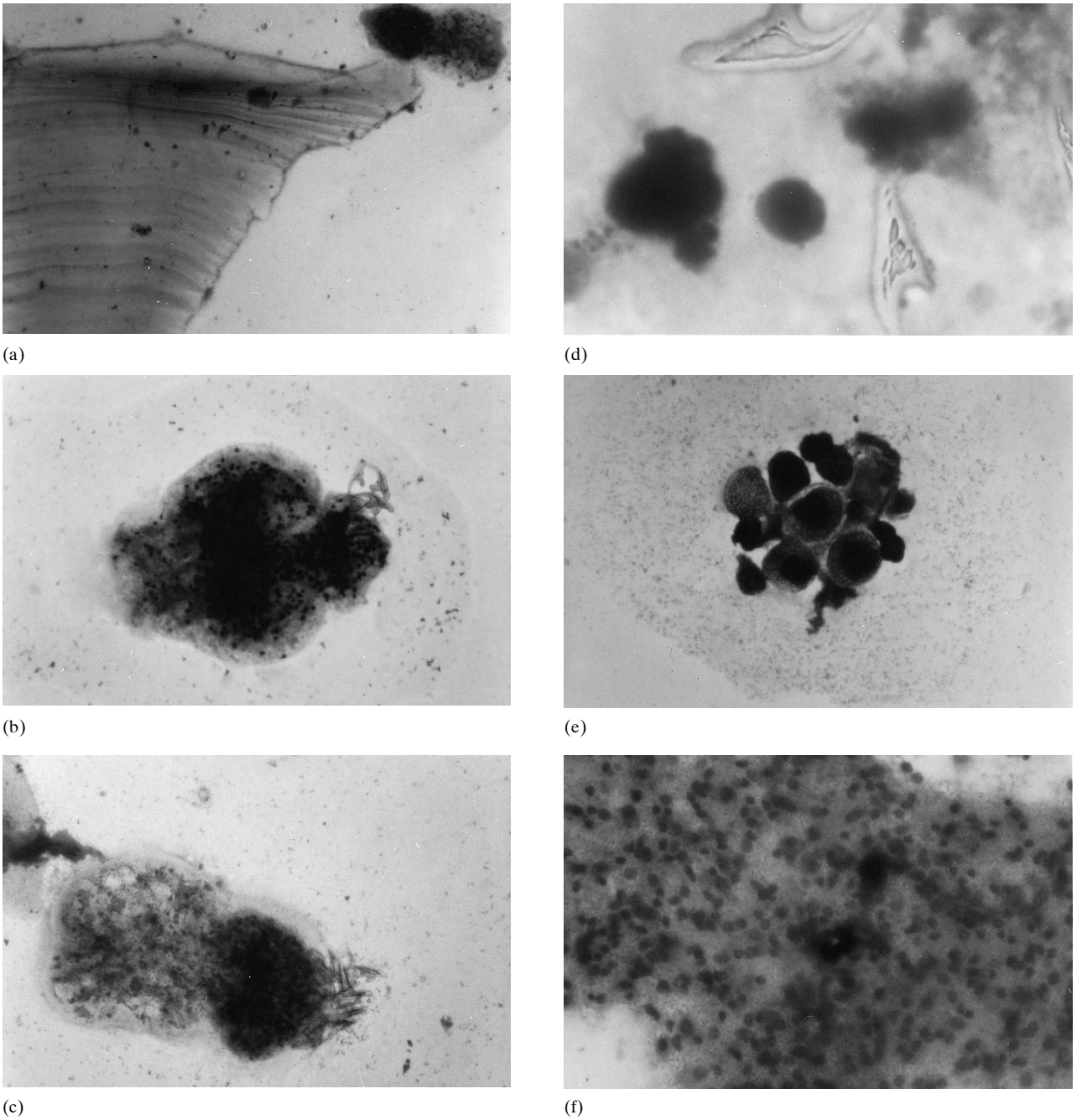

FIG. 3

Histopathology of hydatid cyst (*Echinococcus oligarthrus*). (a) Thick laminated ectocyst wall with attached protoscloex showing presence of calcareous corpuscles (Giemsa stain; ×200). (b) Protoscolex. (c) Scolex with hooklets. (d) Straight back hooklets of *E. oligarthrus* (Giemsa stain; ×1000). (e) Cluster of scolices with germinative layer. (f) Germinative layer showing calcareous corpuscles (Giemsa stain; ×400).

redness nor warmth. The swelling was bimanually palpable. There was no discharge from the submandibular duct opening. The oral cavity floor was within normal limits. The chest was clear and the liver was not enlarged.

Fine needle aspiration was requested. Two to three drops of clear fluid were aspirated. This showed numerous scolices with suckers and hooklets. Fragments of a laminated membrane (ectocyst) and a germinative layer showing the presence of calcareous corpuscles were also observed. Features were consistent with *E. oligarthrus*.

Blood investigations revealed eosinophilia (eosinophil count – 21 per cent, absolute eosinophil count 1250 cells/μl) and raised ESR. Liver function tests were normal. X-rays of the submandibular region showed a soft tissue mass with no radio-opaque shadow or erosion of the mandible. X-rays of the chest were normal. Ultrasonography of the abdomen was within normal limits. Ultrasonography of the submandibular region revealed a cystic lesion within the left submandibular gland.

The swelling was explored surgically through the submandibular approach. No difficulty was encountered in finding a plane of separation. The submandibular gland along with the enclosed hydatid cyst was removed in toto and sent for histopathology. Post-operative recovery was uneventful.

Histopathology confirmed the diagnosis of submandibular hydatid cyst caused by *Echinococcus oligarthrus*.

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Discussion

Parasitic diseases are common in tropical India but head and neck region manifestations of parasitic diseases are rare. Echinococcus has also been called hydatid disease ('hydatid' in Greek means 'a drop of water').

In the natural cycle the hydatid occurs in the organs of sheep (intermediate host), which like man are infected by ova disseminated in the faeces of dogs. Dogs are infected when they eat hydatids in the tissues of sheep or other domesticated herbivores. Man is infected by ova from his hunting dogs.

When the ova of *Echinococcus* excreted in the faeces of dogs are ingested accidentally by man; they hatch in the gastrointestinal tract where the embryos pass through the wall and enter the portal circulation. They usually settle in the liver and lungs, but in rare instances also in the bones, brain, eyes, heart, spleen or kidney. Six to 10 days later, the embryo becomes vacuolated and transforms into an hydatid cyst. These cysts consists of three layers. One from the host and two of parasitic origin. The outer layer consists of more or less dense fibrous tissue with an infiltrate of inflammatory and giant cells. The parasitic intermediate layer is non-nucleated and laminated. The inner layer is formed by germinal epithelial that produces the hydatid sand consisting of brood capsules in which the scolices are being formed. When these scolices are liberated due to rupture of the cyst wall, especially during surgical removal, they may form new hydatid cysts. Table I shows the subtle similarities and differences between *E. granulosus*, *E. oligarthrus* and *C. cellulosae*. Thus, the structure of *E. oligarthrus* was composed of a laminated wall (ectocyst) thicker than that of *E. granulosus*; a very thick, undulated germinal layer (endocyst) unlike that in *E. granulosus* (which if aspirated in a needle, would simulate the structure of *Cysticercus*) and large calcareous corpuscles in the germinal layer (similar to those seen in *Cysticercus*) and several brood capsules with numerous protoscolices.

Hydatid cysts develop most frequently in the liver (60 per cent) and lungs (20 per cent), rarely in the bones, brain, eye, heart, kidney or spleen. The involvement of salivary glands including the submandibular gland is a rarity.

Placitelli,[2] Singh[3] and Onerci[4] reported cases of hydatid cysts in the submandibular gland. In these three cases the causative organism was *E. granulosus*. Kini *et al.*[5] reported the first case of submandibular hydatid cyst caused by *E. oligarthrus*. Ours is the second case report of submandibular hydatid cyst, where the caustive organism is *E. oligarthrus*.

The majority of hydatid cysts cause no symptoms. Symptoms depend on the location and size of the cyst and are characteristic of slow-growing, benign tumours. Patients with echinococcosis must undergo a thorough systemic investigation, since the disease is generally located in other parts of the body and hepatic or pulmonary lesions are generally noted. However, in our case there was no hepatic nor pulmonary involvement.

Closed aspiration of cysts should not be carried out, because spillage may cause anaphylaxis or secondary lesions. Surgical removal of the hydatid cyst remains the most effective treatment, since there is no effective medical treatment. The aim of surgery is total removal of the cyst while avoiding spilling of its contents. Silver nitrate 0.5 per cent, hypertonic saline 20 per cent and other chemicals are often injected into hydatid cysts at the time of surgery to inactivate the protoscolices. If surgery is not possible because of the general condition of the patient, treatment with mebendazole (50 to 150 mg/kg per day orally as a single dose for three months) or albendazole (10 to 15 mg/kg per day orally in two equal doses every 12 hours for 12 weeks) may be tried although the results are unpredictable and adverse reactions have been reported. Prolonged follow-up is required to determine the eventual outcome.

TABLE I
DIFFERENTIAL DIAGNOSIS OF E. GRANULOSUS, E. OLIGARTHRUS AND C. CELLULOSAE ON CYTOLOGY AND HISTOLOGY[5]

Parameter	*E. granulosus*	*E. oligarthrus*	*C. cellulosae*
Site	Viscera Muscle (uncommon)	Extrahepatic	Any tissue
Lesion	Large (>5 cm) Unilocular Filled with fluid	Large (>5cm) Polycystic Filled with fluid	Relatively small (1–2cm) Unilocular Filled with scanty fluid
Aspirate volume	Voluminous	Voluminous	Scanty
Colour	Clear or straw-coloured	Clear to straw-coloured	Clear
Scolices	Multiple	Multiple	Single
Hooklet	Numerous (20–30 μm)	Numerous (20–30μm)	Large (130–150μm)
Ectocyst	Fragments	Fragments	Fragments of nucleated spiral wall
Endocyst	Usually not aspirated	Fragments thick, with nuclei in fibrillar matrix	Nuclei in fibrillar matrix
Calcareous corpuscles	Absent	Present	Present
Histology (other features) Germinative layer	10–25 μm Ill-defined brood capsules	500–700μm Well-defined brood capsules with peripheral undulations to germinal layer	Absent
Protoscolices	Number variable	1–7 in each brood capsule loculus	Single scolex
Calcareous corpuscles	Absent	In both germinative layer + protoscolices	In spiral body wall

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References

1 Altman MM, Gutman D. Echinococcus of the parotid gland. *J Laryngol Otol* 1966;**80**:409–11

2 Placitelli G. Sulle cisti de echinococco primitive delle ghiandde salivari. *Riv Chir* 1938;**4**:109

3 Singh S. Submandibular hydatid cyst. *J Laryngol Otol* 1972;**86**:647–50

4 Onerci M, Tuen E, Ruacan S. Submandibular hydatid cyst. *J Craniomaxillofac Surg* 1991;**19**:359–61

5 Kini U, Shariff S, Nirmala V. Aspiration cytology of *Echinococcus oligarthrus*. *Acta Cytol* 1997;**41**:544–8

Address for correspondence:
Professor J. K. Sahni,
LD-155,
Sardar Patel Apts,
Pitampura,
Delhi-110034
India.

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