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# Hydatid cyst of the pterygopalatine-infratemporal fossa

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**Abstract**
Hydatid disease is caused by the parasitic tapeworm *Echinococcus*. This parasite in larval stage can thrive in many parts of the body, most commonly in the liver and the lung. Hydatid disease in the head and neck region is rare. An unusual location for hydatid disease in the pterygopalatine fossa-infratemporal fossa is presented. The patient did not have evidence of any other cyst on a ten-year follow-up.

**Key words: Echinococcosis; Head and neck**

## Introduction

Hydatid disease is caused by the smallest tapeworms of medical importance, echinococci. The larval forms of all four currently recognised species of *Echinococcus* are recognised as causing disease (echinococcosis or hydatid disease) in humans: *Echinococcus granulosus, E. multiocularis, E. vogeli, E. oligarthrus* (Kammerer and Schantz, 1993). *E. granulosus* is the most common cause of hydatid disease in man. *E. granulosus*, whose life cycles involve dogs and other canids as final hosts and a variety of domestic livestock species and wild ungulates as intermediate hosts, is of cosmopolitan distribution but concentrated in major sheep raising and pastoral areas of the world (Williams *et al.*, 1971; Matossian *et al.*, 1977). The final or definitive host harbours the adult worm in its intestinal tract, and the intermediate host carries the larval stage in its organs. Man becomes accidentally involved in the life cycle of the parasite by ingesting the eggs shed in the faeces of the infested definitive host but does not participate in the complete cycle since infested human organs are not eaten by the definitive host. The sheep-dog cycle is dominant in Europe, parts of the Soviet Union, Western USA, Mexico and the sheep-rearing areas of South America and Austraila. The camel-dog associations are implicated in the life-cycle in Eastern Mediterranean regions, North Africa and the Middle-East (Morris and Richards, 1992).

Typically, cystic echinococcosis consists of a single, unilocular cyst; however, 20 per cent to 30 per cent of cases may have multiple cysts in the same or multiple organs (Kammerer and Schantz, 1993). The clinical course of this disease depends on the site of involvement and size of the cyst. The majority of hydatid cysts appear in the liver (65 per cent), lungs (25 per cent), and, less frequently, in the spleen, kidneys, heart, bone and central nervous system (Akyildiz *et al.*, 1991). Hydatid disease in the head and neck region is rare and only a few case reports are found in the literature.

A hydatid cyst will typically have a trilaminar wall. The outermost layer is called the ectocyst and is comprised of compressed host tissue. The intermediate layer is called the laminar layer, this is derived from the parasite. The intermediate layer, the germinal layer, is live cellular parasite tissue which is responsible for cyst growth and production of protoscolices, brood capsules (budded off pieces of germinal layer with attached protoscolices) and daughter cysts. The fluid contained in an uncomplicated hydatid cyst is crystal clear and odourless (Morris and Richards, 1992).

## Case report

A 23-year-old female patient attended the Head and Neck Clinic of the King Faisal Specialist Hospital and Research Centre, Riyadh, Saudi Arabia, with a history of progressive swelling over the left zygomatic region for two years. On examination she had a $5 \times 7$ cm, cystic, nontender swelling situated over the left zygomatic arch and above it. She also had mild trismus. The posterior teeth of the left upper jaw were displaced medially and there was a bulge in the left retromolar trigone. The rest of the ear, nose and throat examination were normal. Computed tomography (CT) scan demonstrated a cystic swelling in the left pterygopalatine-infratemporal fossa (Figure 1). The lesion displaced the lateral pterygoid process medially, the zygomatic process laterally and the posterior wall of the maxillary antrum anteriorly. The ascending ramus of the mandible was eroded. Ultrasound examination of the abdomen and chest X-ray were normal. An echinococcal antibody titre (Indirect haemagglutination) was 1 in 512.

The cyst was excised utilising a subtemporal approach. Approximately 150 ml of clear fluid was aspirated from the cyst. Frozen section diagnosis was suggestive of a non-malignant lesion. The temporalis muscle was used to fill the defect. The excised cyst was $7 \times 5 \times 3$ cm in size. On histological examination laminated, acellular cyst membrane was seen which was compatible with a hydatid cyst (Figure 2).

The patient was treated with 400 mg of mebendazole three times a day for three months post-operatively. At the end of mebendazole therapy the echinococcal antibody

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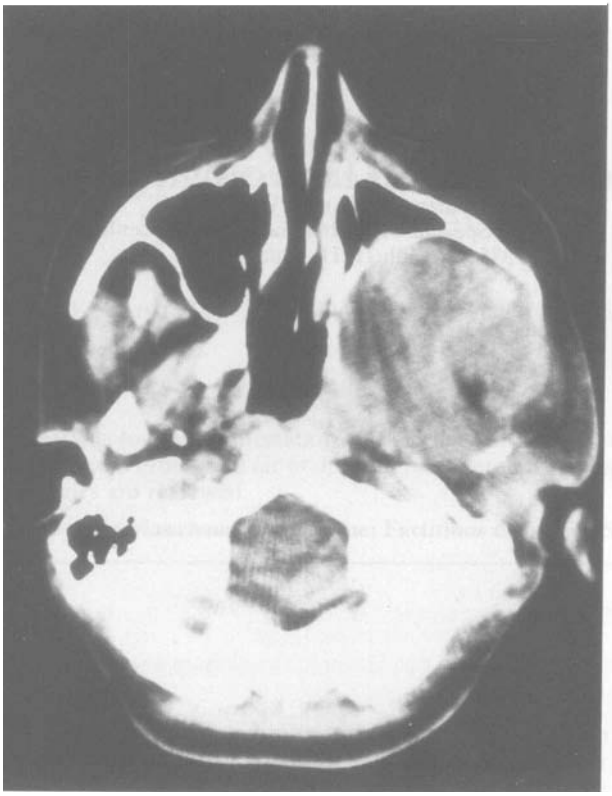

FIG. 1

Contrast CT scan of the head showing the lesion in the left pterygopalatine-infratemporal fossa. The posterior wall of the left maxilla is displaced anteriorly.

titre dropped to 1 in 16. Ten years since the surgical removal of the cyst she remains well without any evidence of any other cyst anywhere in the body.

## Discussion

Mass lesions in the infratemporal fossa include primary and secondary malignant tumours, benign tumours such as haemangiomas, schwannomas, and, paragangliomas, and cystic lesions such as encephalocoeles and retention cysts (Sener, 1994). Hydatid cysts being so rare in the head and neck, particularly at this site; are not usually considered in the differential diagnoses, especially in absence of cysts elsewhere. Hydatid cysts in the head and neck are infrequent even in countries where *Echinococcus* infestation is high (Akyldiz *et al.*, 1991).

Shuker (1982) described a infratemporal hydatid cyst in a 10-year-old boy, the cyst was removed surgically. The patient also had a hepatic hydatid cyst.

Akyildiz *et al.* (1991) described a hydatid cyst in the pterygopalatine fossa in a 15-year-old boy. The cyst was removed surgically. No other site was found to be involved on a four-year follow-up.

Sener (1994) described a magnetic resonance imaging (MRI) finding of a hydatid cyst in the infratemporal fossa. The patient was a 60-year-old female who had multiple pulmonary and hepatic cysts. She died due to an unrelated myocardial infarction.

Sennaroglu *et al.* (1994) described hydatid cyst of the infratemporal fossa in a 26-year-old female. The patient had had hydatid cyst of the liver removed previously. The infratemporal hydatid cyst was removed surgically.

Our patient did not have any evidence of another cyst at the time of first presentation. Even on ten-year follow-up she did not have cysts anywhere else in her body.

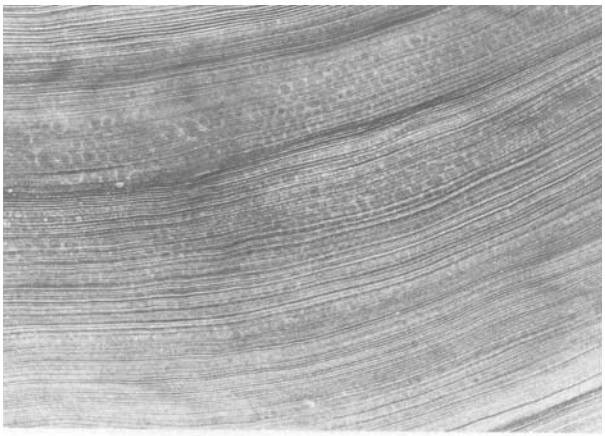

FIG. 2

Photomicrograph showing the characteristic laminated wall of the hydatid cyst (PAS stain, × 200).

Surgical removal of the hydatid cyst is the most effective treatment. The surgeon must be very careful to remove the cyst totally avoiding the adverse consequences of spilling its contents since fatal anaphylaxis on spilling the contents of the cyst has been reported. Prior to manipulation of the cyst the protoscolices can be inactivated by hypertonic saline (20 per cent) or silver nitrate (0.5 per cent) (Sennaroglu *et al.*, 1994).

*In vitro* and *in vivo* studies in animal models have demonstrated the safety, efficacy, and limitations of benzimidazole compounds (mainly mebendazole and albendazole). Clinical trials have been promising, but shown variable results. Both albendazole and mebendazole have demonstrated efficacy. Investigators who have compared both drugs concluded, however, that slightly greater efficacy, in terms of rates of complete cure and improvement has been obtained with albendazole (Kammerer and Schantz, 1993). In case of spillage, post-operative chemotherapy should start immediately after the operation because albendazole is found to be less effective when used 15 days after the operation (Morris and Taylor, 1988).

Serological tests are often used to confirm or rule out a presumptive diagnosis of echinococcosis; however there are many difficulties (Kammerer and Schantz, 1993). Both false-positive and false-negative tests are common. However, serology has a clear role in the follow-up of patients after surgery, titres should fall after resection and any subsequent rise is likely to indicate recurrence (Morris and Richards, 1992). In our case the initial echinococcal antibody titre (Indirect haemagglutnation) was 1 in 512 at the time of surgery and the titre dropped to 1 in 16 by the time a three-month course of mebendazole was completed.

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