SHORT COMMUNICATION

Isolated nasolabial hydatid cyst: an unusual location

Although human cystic echinococcosis (CE) may be a common health problem in areas where the causative cestode (*Echinococcus granulosus*) is endemic, such as southern Europe and the Middle East (Pasaoglu et al., 1998; Akhan et al., 2001), it is also being seen increasingly in northern Europe, and other non-endemic areas, because of the migration of labour and international tourism. The liver is the most common site of involvement, the portal transport of the parasite’s oncospheres from the gut, and the entrapment of most of those oncospheres in the liver sinusoids, usually leading to one or more hepatic cysts. Cysts can, however, develop almost anywhere in the human body, from oncospheres carried past the liver into the general circulation, including, albeit rarely, the head or neck (Pasaoglu et al., 1998; Eroğlu et al., 1999; Akhan et al., 2001; Adaletli et al., 2005; Benhammou et al., 2007). Cysts have been reported, for example, in the neck, parotid gland, infratemporal fossae and parapharyngeal space (Sennaroglu et al., 1994; El Kohen et al., 2003; Katılmıs et al., 2007). Even in such rare cases, there is usually hepatic or pulmonary involvement. Solitary hydatid cysts in the head or neck are very rare but important because, in the absence of liver or lung cysts, they may be excised before being correctly identified as hydatid, with the risk of intra-operative anaphylaxis and secondary echinococcosis if protoscoleces are spilled during surgery. An Iranian patient who presented with an isolated facial hydatid cyst in the anterior aspect of his maxilla is described below.

CASE REPORT

A 41-year-old man was referred to the Imam Khomeini Hospital (which is affiliated to Tehran University of Medical Sciences) in Tehran, with a 1-month history of a progressive, painful swelling on the right side of his nose. The lesion was tender and erythematous, with inflammatory signs, swelling of the gingivobuccal sulcus and obstruction of the right nostril. A computed tomographic (CT) scan showed a cystic lesion extending to the lacrimal crest, with some pressure effect on the sinus wall (Fig. 1). A clinical examination revealed no other abnormalities, and routine laboratory tests all gave normal results.

The cyst was completely resected using a sublabial approach. No early post-operative complication was seen and the patient was discharged after 2 days. Subsequent histological examination of the excised cyst, which measured $2.5 \times 2.0 \times 0.5$ cm, revealed chronic inflammation around the cyst and the laminated layers that are characteristic of the wall of a hydatid cyst. Four months after the surgery, the patient returned with another swelling on the right side of his face (Fig. 2). A CT scan revealed a mass that occupied the infratemporal fossae and the lateral aspect of the right orbit, with communications between these two components (Fig. 3).

Since the first cyst had been identified as hydatid, the patient was given albendazole (400 mg twice daily) for 2 weeks before the surgery to remove the second cyst. The patient was found positive in an ELISA for anti-*E. granulosus* antibodies but a thorough check for other cysts, including abdominal, chest and brain CT scans, gave a negative result.

In the lateral removal of the second cyst, the superior branch of the facial nerve was carefully preserved. The exposed cyst was injected with methanol, to render any
protoscoleces non-viable, before the whole mass, including the germinative layer, was carefully excised, being careful to avoid the cyst’s rupture. The orbital portion was extracted through the inferior orbital fissure. In the 6 months since the removal of the second cyst, the patient has made a good recovery, there has been no further recurrence of the echinococcosis, and the patient’s titres of anti-\textit{Echinococcus} antibodies have gradually fallen.

\textbf{DISCUSSION}

The main site affected by human CE is the liver, the result of the onchospheres that hatch from ingested ova, in the host's gut, being carried in the portal vein to the hepatic sinusoids. Most onchospheres that manage to pass through the liver are caught in the pulmonary capillaries, making the lungs the second most common site of cyst development. Very few onchospheres pass through the liver, heart and lungs, then entering other organs by means of the general circulation.
who lives, or has lived, in an area where *E. granulosus* is endemic (Rena et al., 2004). Hydatid cysts, particularly if isolated in a rare site, may be mistaken for slow-growing benign tumours (Fradis et al., 1989). The pre-operative identification of hydatid cysts is important because it allows the surgeon to take precautions to reduce the risk of the anaphylactic reaction and/or secondary CE that can result if a hydatid cyst is ruptured during surgery (El Gbouri et al., 1997). The second cyst seen in the present, Iranian case probably represented secondary CE caused by the accidental rupture of the first cyst. Unfortunately, at the time the first mass was excised, the (remote) possibility that it was a hydatid cyst had not been considered and, in consequence, there was no pre-operative treatment with a scolicide such as albendazole and no intra-operative scolicidal injection.

Serological tests (e.g. direct haemagglutination, latex agglutination, immunoelectrophoresis, skin tests, and ELISA) and CT scans or other imaging techniques can be useful in the differential diagnosis of CE. None of the serological methods has 100% specificity but, in recent years, computed tomography, ultrasonography and/or magnetic resonance imaging have greatly facilitated the accurate diagnosis of CE, with ultrasound currently the method of choice for searching for the relevant pathognomonic criteria (Pasaoglu et al., 1998; Rena et al., 2004).

Although some cases of human CE appear to have been treated successfully using only chemotherapy (Guven et al., 2004), most hydatid cysts, including most of those that have been found in the head and neck (Katılmış et al., 2007), are still surgically excised. Even with surgical excision, however, pre-operative and, ideally, post-operative treatment with a scolicide such as albendazole is recommended. Percutaneous treatment of cysts, as used against a parotid cyst by Akhan et al. (2001), can reduce the risks of post-operative morbidity and complications but the efficacy of such treatment, compared with excision, has still to be fully explored.
In conclusion, the present case, of an isolated nasolabial hydatid cyst, represents an unusual case of CE. It emphasises the importance of considering CE in the differential diagnosis of a cystic mass, even in the absence of hepatic and/or pulmonary cysts, especially in patients who have been in areas where *E. granulosus* is endemic. Pre-operative imaging and serology should permit hydatid cysts to be identified and the appropriate precautions against intra-operative anaphylaxis and secondary CE to be taken.

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