

Submandibular and thyroid gland involvement of hydatid cysts: a very rare association with percutaneous treatment

Serhat Avcu, Özkan Ünal, Çetin Kotan, Mustafa Öztürk, Özkan Özen

ABSTRACT

A 48-year-old man who had a complaint of swelling in the left submandibular region and right side of the neck was referred to our radiology clinic for sonographic examination that revealed a type 1 hydatid cyst with a diameter of 32 mm in the left submandibular gland, and a type 3 hydatid cyst with a diameter of 33 mm in the right thyroid gland. The diagnosis was verified by computed tomography, magnetic resonance imaging, and hemagglutination tests. To the best of our knowledge, this is the second case in the literature of hydatid cysts involving both submandibular and thyroid glands. There was no abdominal, thoracic or brain involvement. The patient was a singer and did not accept surgical treatment, and percutaneous drainage with the PAIR (Puncture-Aspiration of cyst contents-Injection of hypertonic saline solution-Reaspiration) technique was performed for both cysts. There were no complications during or after the intervention. During 4 months of follow up the sizes of both cysts were decreased and the germinative membranes were shrunken, having a pseudotumor appearance. To the best of our knowledge, this is the first case in the literature that the PAIR method was applied for thyroid and submandibular gland hydatid cysts. Percutaneous treatment of thyroid and submandibular gland hydatid cysts seems to be a safe and effective procedure as a possible alternative to surgery.

Key words: • hydatid cyst • submandibular gland • thyroid gland • interventional radiology

H ydatid disease is an important infestation caused by the tape-worm parasite *Echinococcus granulosus* and is common in the temperate zones, including the Mediterranean countries, the Middle East, South America, New Zealand, Australia, and Southeast Asia. Hydatid cysts affecting human beings involve mostly the liver (65%) and lungs (25%). Hydatidosis of head and neck is rare even in the regions where the disease is endemic (1). It is claimed that the overall incidence of thyroid hydatid cysts in Turkey is 0.51% (2). To the best of our knowledge, there is only one case in the literature involving the submandibular and thyroid glands together (3).

The traditional treatment of choice for a hydatid cyst in the thyroid and submandibular glands is surgery as for the cysts located elsewhere. Therefore, all the reported cases were treated by surgical means. However, intraoperative and postoperative complications, and high recurrence rates are not rare. Successful percutaneous treatment of liver, kidney, pulmonary, orbital, and parotid gland hydatid cysts has been reported in past decade (4).

We present the findings of 4 months of follow-up in a case with a type 3 hydatid disease of the right thyroid gland and a type 1 hydatid disease of the left submandibular gland treated percutaneously by using the PAIR (Puncture-Aspiration of cyst contents-Injection of hypertonic saline solution-Reaspiration) technique.

Case report

A 48-year-old man who had a complaint of swelling in the left submandibular region and in the right side of the neck for two years applied to our hospital. On physical examination, moderate-firm masses were palpated at left submandibular and right thyroid regions that caused no skin warmth or redness. On ultrasonography (US), a cystic lesion measuring 32 x 23 mm with a regular contour and double membranes was observed in the left submandibular gland, and a cystic lesion containing a separated germinative membrane and two daughter vesicles was detected in the right thyroid gland measuring 33 x 25 mm. With these sonographic findings, the diagnosis of type 1 and type 3 hydatid cysts was made for the left submandibular gland and right thyroid gland lesions, respectively. The diagnoses were supported with computed tomography (CT) and magnetic resonance imaging (MRI) (Figs. 1, 2). The hydatid cyst in the right thyroid gland was displacing the common carotid artery and jugular vein mildly to the right, and the trachea and esophagus mildly to the left. No more cystic lesions were detected in the abdominal, thoracic and brain examinations of the patient. The hemagglutination test for hydatid cyst was positive. Other physical examination findings and history were unremarkable. Blood tests (complete blood count, blood chemistry,

From the Departments of Radiology (S.A. ✉ serhatavcu@hotmail.com, Ö.Ü., Ö.Ö.), General Surgery (Ç.K.), and Endocrinology (M.Ö.), Yüzüncü Yıl University Faculty of Medicine, Van, Turkey.

Received 26 August 2008; revision requested 6 October 2008; revision received 7 October 2008; accepted 20 October 2008.

Published online 9 October 2009
DOI 10.4261/1305-3825.DIR.1967-08.2

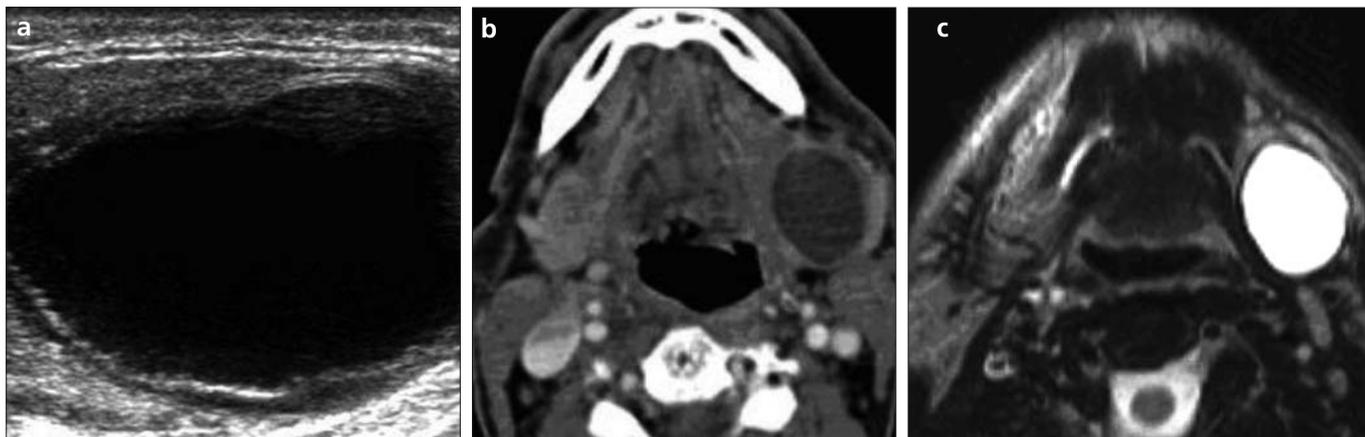


Figure 1. a–c. US image (a) showing left submandibular gland cystic lesion with double membrane appearance. Axial CT (b) and T2-weighted MR (c) images show the thick-walled cystic lesion in the left submandibular gland (type 1 hydatid cyst).

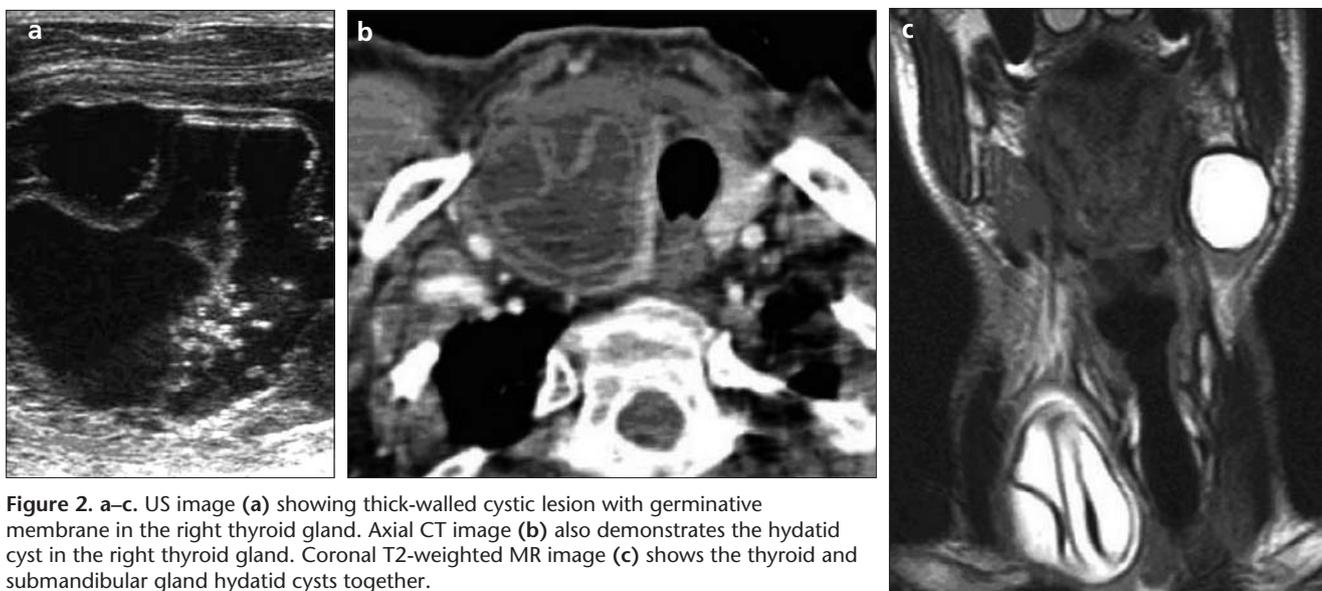


Figure 2. a–c. US image (a) showing thick-walled cystic lesion with germinative membrane in the right thyroid gland. Axial CT image (b) also demonstrates the hydatid cyst in the right thyroid gland. Coronal T2-weighted MR image (c) shows the thyroid and submandibular gland hydatid cysts together.

and thyroid function tests) were also unremarkable.

As the patient was a singer and did not accept surgical treatment, we performed percutaneous intervention with the PAIR technique for both of the cysts after getting informed consent from the patient. Albendazole (10 mg/kg day) was administered orally to the patient for 1 week before the procedure and 2 weeks after the procedure as a prophylactic measure to prevent spillage of the disease. Because of the risk of anaphylaxis, antiallergic prophylaxis with oral diphenhydramine and corticosteroid was applied 15 min before the intervention. Before the procedure, liver function tests were performed and blood counts and coagulation functions were determined. Necessary medicines and equipment were kept

ready in the procedure unit in case of anaphylaxis. An anesthesiology team was available during the procedure. The percutaneous intervention was performed under sonographic guidance. After maintaining standard sterile conditions, cyst puncture was carried out under local anesthesia. The cyst was treated by the PAIR technique. A Seldinger needle was used for the puncture of the cyst. When the needle entered the cavity, 35% of the estimated volume of cystic fluid was aspirated. The cavity was then filled with hypertonic saline (20% NaCl) to an amount 10% lesser than the aspirated volume. We waited for 10 minutes after the injection of hypertonic saline and then reaspirated the cyst content. Sonographically we monitored the complete separation of the endocyst (ger-

minative and laminated membranes) from the pericyst. We performed the same technique in both submandibular and thyroid gland hydatid cysts. The diagnosis of hydatid disease was confirmed by microscopic examination which revealed the fragments of membranes and protosolices. At the end of the procedure there were still 2 daughter vesicles in the cyst cavity of the right thyroid gland. There were no complications during or after the intervention.

Follow-up sonography 4 months after the procedure revealed a 32 x 19 mm mass in the right thyroid gland and a 23 x 15 mm mass in the left submandibular gland, both giving a “pseudotumor appearance” without any fluid component or daughter vesicles (Figs. 3, 4).



Figure 3. US image of cyst cavity in the left submandibular gland filled with germinative membranes giving a solid “pseudotumor” appearance with decrease in size at 4 months of follow-up.

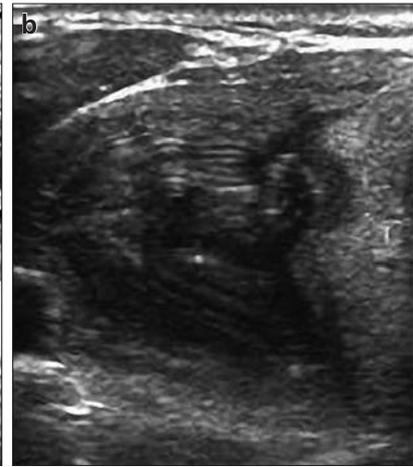
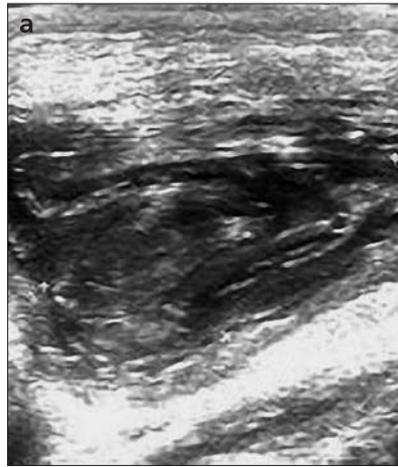


Figure 4. a, b. US images of cyst cavity in the right thyroid gland filled with germinative membranes giving a solid “pseudotumor” appearance with decrease in size at 4 months of follow-up.

Discussion

Echinococcosis, although eradicated in many countries, is still widespread in communities where agriculture is dominant, and cystic hydatidosis is a significant public health problem in regions where echinococcosis is endemic. In the hydatid disease which is caused mostly by *E. granulosus* and rarely by *E. multilocularis* the dogs are the primary hosts, and cows, sheep, horses, pigs, and human beings are the intermediate hosts. The parasite eggs entering the organism open in the small intestine of the intermediate host and reach the liver and lung through the portal venous or lymphatic systems; they then form the hydatid cyst lesions. The embryos that pass over the hepatic sinusoid or pulmonary capillar barriers enter the systemic circulation and may lodge within any organ in the body (5).

Hydatid cysts develop most frequently in the liver and lungs, and rarely in the bones, brain, eye, heart, kidney or spleen. Patients with echinococcosis should undergo a thorough systemic investigation, since the disease is usually also located in other parts of the body, mostly in the liver and lungs. The involvement of salivary glands including the submandibular gland is a rarity (6). The hydatid disease of the thyroid gland is also a very rare condition (7). Thyroid involvement is seen only in 1–2% of patients who have liver or lung hydatid disease (8). In our case there was no abdominal, thoracic or brain involvement.

The majority of hydatid cysts cause no symptoms. Although the disease is generally asymptomatic, it may show clinical signs due to the dimension and localization of the cyst, as well as due to the pressure of the growing cyst (9). In our case there were no symptoms apart from painless, mobile swelling in the left submandibular and right thyroid regions.

In cases of echinococcosis, the diagnosis is made primarily by patient history, physical examination, radiological imaging modalities, serological tests, and aspiration (9). Radiological diagnostic modalities are usually US, CT, and MRI (10, 11). Serological tests are indirect hemagglutination, latex agglutination, ELISA, and immune electrophoresis. Indirect hemagglutination test was done in our patient and the result was positive. The serologic tests are widely used to confirm the diagnosis, although they are associated with false-negative and false-positive results (9). Therefore, imaging modalities remain more sensitive than serodiagnosis, especially with unusual cyst locations, and a characteristic scan demonstrating germinative membrane and daughter vesicles in the presence of negative serologic results should still suggest the diagnosis of echinococcosis.

Surgical removal of the hydatid cyst remains the most effective treatment, since there is no effective medical treatment. The aim of the surgery is total removal of the cyst while avoid-

ing spilling of its contents. If surgery is not possible because of the general condition of the patient, treatment with mebendazole or albendazole may be tried although the results are unpredictable and adverse reactions have been reported. Prolonged follow-up is required to determine the eventual outcome (6). However, it seems that antiparasitic agents may have a role in killing the alive parasites and preventing a possible contamination caused by the spillage of cyst contents during the operation, rather than provide a complete cure.

Although it is claimed that it should be risky to make needle aspiration in hydatid cyst because of possible contamination by leakage, and cyst leakage or rupture may even be associated with severe allergic reaction to parasite antigens, and may cause anaphylactoid reactions, it is well known that the PAIR technique is successfully used for the percutaneous treatment of liver hydatidosis (12) and of soft tissue hydatid cysts (13). Also, Akhan et al. (4) reported a case of parotid gland hydatid cyst which underwent percutaneous treatment as an alternative to surgery. But as far as we know, the PAIR method is not applied to thyroid and submandibular hydatid cysts until now.

We performed a successful percutaneous treatment of both thyroid and submandibular gland hydatid cysts with no complications during or after the procedure and with an outcome of decrease in cyst-size and shrinkage of

cyst germinative membranes giving a solid "pseudotumor" appearance on US at the 4-month follow-up. Successful percutaneous treatment of the liver, kidney, pulmonary, and orbital hydatid cysts has been reported in past decade. Healing criteria after percutaneous procedure, such as reduction in size and volume of the cyst, thickening, and irregularity of the cyst wall, decreased fluid content, and solid appearance of the remnant on US examination, have already been described (4). All these healing criteria were observed in our case after percutaneous treatment. Although the cysts were successfully treated, long-term follow-up is necessary due to the risk of recurrence.

In conclusion, hydatid disease should be considered in the differential diagnosis in patients with cystic masses in the head and neck regions, and percutaneous treatment of thyroid and submandibular gland hydatid cysts seems to be a safe and effective procedure as a possible alternative to surgery.

References

1. Önerci M, Turan E, Ruacan S. Submandibular hydatid cyst. *J Cranio-Max-Fac Surg* 1991; 19:359–361.
2. Özerkan E, Gürçınar M, Sarıoğlu B, Aydınlioğlu H. A case of cystic echinococcosis in thyroid gland: a very rare localization of echinococcosis infection. *Turk J Endocrinol Metabol* 1999; 4:181–183.
3. Ulku CH, Uyar Y, Gungor S, Koroglu D, Arbag H. Hydatid cyst of the submandibular salivary gland in a child. *Int J Ped Otorhinolaryngol Extra* 2007; 2:23–25.
4. Akhan O, Ensari S, Ozmen M. Percutaneous treatment of a parotid gland hydatid cyst: a possible alternative to surgery. *Eur Radiol* 2002; 12:597–599.
5. Koşar U, Kacar M, Şirin F, Erdoğan A. Hidatid kistlerin ender lokalizasyonları ve BT görünüşleri. *Tanı Girişim Radyol* 1998; 4:457–460.
6. Sahni JK, Jain M, Bajaj Y, Kumar V, Jain A. Submandibular hydatid cyst caused by *Echinococcus oligarthrus*. *J Laryngol Otol* 2000; 114:473–476.
7. Amrani M, Zouaidia F, Belabbas MA, Labrousse F, Catanzano G, Elhachimi A. [Hydatidosis: apropos of several uncommon locations]. *Med Trop (Mars)* 2000; 60:271–272.
8. Versaci A, Scuderi G, Rosato A, Angiò LG, Oliva G, Sfuncia G, et al. Rare localizations of echinococcosis: personal experience. *ANZ J Surg* 2005; 75:986.
9. Bouckaert NM, Raubenheimer EJ, Jacobs FJ. Maxillofacial hydatid cysts. *Oral Surg Med Oral Pathol Radiol Endod* 2000; 89:338–342.
10. Turgut S, Ensari S, Katirci H, Çelikkanat S. Rare otolaryngologic presentation of hydatid cyst. *Otolaryngol Head Neck Surg* 1997; 117:418–421.
11. Jain SKT, Jamdade PT, Muneshwar SS, Ambulgekar VK, Panzade SM. Hydatid cyst of thyroid: an unusual cause of stridor. *Ind J Otolaryngol Head Neck Surg* 2005; 57:80–81.
12. Paksoy Y, Ödev K, Şahin M, Arslan A, Koç O. Percutaneous treatment of liver hydatid cysts: comparison of direct injection of albendazole and hypertonic saline solution. *AJR Am J Roentgenol* 2005; 185:727–734.
13. Akhan O, Gumus B, Akinci D, Karcaaltincaba M, Ozmen M. Diagnosis and percutaneous treatment of soft-tissue hydatid cysts. *Cardiovasc Intervent Radiol* 2007; 30:419–425.