CYSTIC ECHINOCOCCOSIS OF THE TONGUE LEADING TO DIAGNOSIS OF MULTIPLE LOCALIZATIONS

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Abstract. The tongue is a rare site of localization of cystic echinococcosis. We report a 3-year-old patient with cystic echinococcosis of the tongue demonstrated by histopathology. The cyst of the tongue was surgically removed. The tongue lesion led us to find additional liver and lung cystic lesions that were successfully treated with albendazole therapy.

INTRODUCTION

Cystic echinococcosis is a parasitic zoonotic disease endemic in cattle-raising regions worldwide. In Latin America, it is most frequent in Argentina, Uruguay, Brazil (Rio Grande do Sul), Chile, and Peru. The annual incidence of human cystic echinococcosis in Chile is 3.6 per 100,000 inhabitants and is more prevalent in the southern XI Region, Aysen, where 16% of all cases in the country are concentrated.1 Although cystic echinococcosis can affect any organ, the most frequent localizations are the liver and lung (80%). Cysts can also be found in the peritoneum (20%), spleen (0.7–8%), kidney (7%), skin and muscles (4%), nervous system (0.2–3%), bones (2%), and heart (0.2–2%).2

This report describes a patient from a sheep-raising region who presented with a cystic echinococcosis lesion of the tongue. The tongue is a rare site of localization for cystic echinococcosis; the clinician, warned by this finding, was able to detect the presence of cysts in the liver and the lung of the patient and initiate treatment.

CASE REPORT

A 3-year-old girl presented at the Dentistry Service of Coyhaique Hospital in Aysen, Chile, on June 14, 1999, with a presumptive physician’s diagnosis of ranula. Physical examination disclosed a single circumscribed globular lesion 3 cm in diameter located in the inferior lateral aspect of the tongue (Figure 1). On July 5, surgery was performed; presumptive diagnosis was that of mucocele of the tongue. During the operation, the swollen part of the tongue was completely removed. However, the lesion ruptured and released a white membrane and crystal-clear, watery fluid, leading the physician to suspect cystic equinococcosis. The surgical wound was carefully washed several times with hydrogen

FIGURE 1. Single globular circumscribed lesion, 3 cm in diameter, in the lateral inferior aspect of the tongue, as observed at the time of first medical consultation.
peroxide and then sutured. The specimen was sent to the Pathology Service of Coyhaique Hospital and to the Oral Pathology Reference Center of the University of Chile in Santiago, where the diagnosis of cystic echinococcosis of the tongue was confirmed (Figure 2).

On July 6, 1999, chest radiography demonstrated 3 pulmonary cystic lesions of 4 cm; 2 were on the right lung and 1 was on the left lung. An abdominal sonogram revealed an additional 5 cysts in the liver measuring 2 to 4 cm in diameter. Four were located in the right lobe, and one was on the left liver lobe. The patient received 3 cycles of albendazole therapy 10 mg/kg per day for 30 days separated by 15 days for each cycle. Treatment was completed on October 8, 1999. Fifteen days after the third cycle of therapy, she presented with fever, dyspnea, and cough. Chest radiographs revealed a diffusely dense zone in the base of the left lower lung lobe. She was admitted to hospital for 2 weeks and was treated with the antibiotics cloxacillin and ceftriaxone. The pulmonary lesions improved, and the pulmonary cysts disappeared. Analysis of chest films revealed nothing abnormal 2 months after the patient was discharged from the hospital. The hepatic cysts were followed up by means of ultrasound. The cysts decreased in size from type I to IV, according to Gharbi's classification, 3 6 months after the completion of albendazole therapy. On January 27 and March 1, 2000, the patient was readmitted for observation of scant hemoptysis, which improved in 2 days. She remains periodically under the care of a physician and is currently leading a normal life.

It is exceptional that a patient only 3 years old could present with a multiple cystic echinococcosis disease that involved the liver, lung, and tongue. This can be explained by early acquisition of echinococcosis, which is highly prevalent in dogs and other mammals in the XI Region of Chile. 4 Soil contamination with eggs of the cestode Echinococcus granulosus is an important problem in this re-
The low socioeconomic level of the family of this patient may have also favored the acquisition of the disease. The tongue lesion prompted the medical consultation. This is an infrequent site for localization of cystic echinococcosis. When the oncosphere trespasses the liver and lung barrier, it can localize in any organ. A literature search resulted in 5 additional reports published between 1963 and 1976,6–10 Cystic echinococcosis should be included in the differential diagnosis of single nodules of the tongue. Such findings may signal the possibility of disseminated disease. Surgery is the preferred treatment for single and accessible nodules, such as the tongue lesion of our patient. Albendazole therapy provides a good alternative to surgery when several lesions are present. The patient we studied was treated with albendazole therapy in recommended doses, and her improvement was consistent with the 60 to 70% regression described in the medical literature.11

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