SOLITARY SUBCUTANEOUS HYDATID CYST: A CASE REPORT

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Abstract. In the absence of hepatic and pulmonary involvement, hydatid disease of other organs is extremely rare. In this paper, we report on a patient who had a solitary subcutaneous hydatid cyst in the submandibular region.

INTRODUCTION

Hydatid disease is an infestation caused by the parasite *Echinococcus granulosus*. The definite hosts are animals such as dogs, wolves, and foxes. Intermediate hosts are herbivores (sheep, goats, cattle) and humans. Seventy percent of the ingested ova are trapped in the liver and those which pass through the hepatic filter disseminate to the lungs, heart, and other organs.

Hydatidosis of the head-neck region is extremely rare even in geographic areas in which echinococcal infestation is frequent.1,2 In this paper, we present a case in which a patient had a hydatid cyst in an uncommon site.

Case report. A 12-year-old girl presented with a cystic mass in the right submandibular region. On physical examination, a 4 × 3 cm mass was palpated under the middle one-third of the right mandible; it was cystic, fluctuant, mobile, and painless. The overlying skin was normal. The preoperative examinations (chest radiograph, complete blood count, urine analysis, and blood biochemistry) revealed no abnormality.

Upon surgical exploration, the mass was found to be attached to the subcutaneous tissue but was not associated with any major structure in the neck. The macroscopic appearance suggested a hydatid cyst; perforation was avoided by means of meticulous dissection. Because the preoperative diagnosis had not been hydatid disease, investigations for other organ involvement (including liver ultrasonography) was performed postoperatively and no other focus was found. Histopathologic examination of the specimen revealed a hydatid cyst.

The patient has been followed for 4 years and no recurrence of hydatidosis has been detected. During the control period, follow-up examinations were performed—chest radiography, complete blood count, blood routine biochemistry (glucose, ALT, AST, alkaline phosphatase, blood urea nitrogen, serum creatinine, total protein, and albumine), and abdominal ultrasonography. Indirect hemagglutination tests were done once every six months in the first year, and once every year in the later follow-up period.

DISCUSSION

Hydatid disease is caused by *Echinococcus granulosus*. The adult parasite (0.5–1 cm) resides in the intestines of animals such as dogs, foxes, wolves, and jackals. The ova, which are resistant to many environmental conditions, are excreted in the feces and ingested by the intermediate hosts, herbivores and humans. The ova hatch in the small intestine, where the parasites penetrate the mucosal wall and reach the liver via the portal vein. They are trapped in the sinusoids; therefore, the liver is the most frequently involved organ (70%).3 The larvae which pass through this first filter, reach the lung via the right heart; the lung is the second most frequently involved site (10–15%). The larvae, which pass through this second filter cause hydatid disease in other organs (5–15%).3 In a large series from Greece, the frequency of extrapleural and extrapulmonary hydatidosis was 9%.4 In addition to systemic dissemination via the portal vein, there are preliminary data on the possibility of dissemination through lymphatic channels, whereas the role of the bronchial route is dubious.3

In our case, the cyst was in the subcutaneous tissue in the submandibular region. In a different series, the frequency of subcutaneous tissue involvement was approximately 2%.5 The interesting aspect of cases with solitary cysts in uncommon sites is the absence of disease in the liver and lungs.2,3,5 It is very difficult to explain how the larvae pass through two filter sites and form solitary cysts. Although no route other then the portal route has been proven in humans, it is strongly possible that systemic dissemination via the lymphatic route accounts for cases with solitary cysts in uncommon sites.

Our case has been under follow-up for 4 years and hydatidosis has not been detected in control examinations. In endemic regions it may be difficult to differentiate a recurrence from a reinfection. However, since our case has had a 4-year disease-free period, if hydatidosis is detected in the future, it will be considered a reinfection. Such cases are communicated in case reports and naturally large series with cysts in uncommon sites are lacking. The final outcome of such patients is usually known only to the authors. However, according to a study by Beard,6 the interval between infection and diagnosis does not exceed several years and contrary to the previously accepted view, the age of the cyst is not equal to the age of the patient.

The most important issue in cases with cysts in uncommon sites is whether hydatidosis will develop in the primary sites. These patients should be followed, paying close attention to this possibility.

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