The majority of the cases reported in English literature and found usually in the liver and lungs but can also develop anywhere in the body, with frequent reports in the spleen, kidney, pancreas, peritoneum, central nervous system, soft tissues, and the breast. Differentiation of the cystic mass from any malignancy poses some difficulties. In addition to routine serologic tests, various radiologic methods are used for identification of the cysts in the unusual localizations. In a recent study, the relation of the carbohydrate antigen (CA 19-9) levels with liver hydatid cysts has been investigated. We report a case of hydatid disease of the breast associated with an elevated CA 19-9 level that was decreased after excision.

INTRODUCTION

Cystic hydatid disease is caused by the parasite Echinococcus granulosus and found usually in the liver and lungs but can also develop anywhere in the body, with frequent reports in the spleen, kidney, pancreas, peritoneum, central nervous system, soft tissues, and the breast. Differentiation of the cystic mass from any malignancy poses some difficulties. In addition to routine serologic tests, various radiologic methods are used for identification of the cysts in the unusual localizations. In a recent study, the relation of the carbohydrate antigen (CA 19-9) levels with liver hydatid cysts has been investigated. We report a case of hydatid disease of the breast associated with an elevated CA 19-9 level that was decreased after excision.

CASE REPORT

A 69-year-old woman presented with a palpable mass in the lower inner quadrant of the left breast, which had been detected by a physician on a routine examination. The patient reported that the mass had existed and had been growing slowly for 6 years. She had no history of pain, injury, discharge from the nipple, fever, or drug misuse and no family history of breast cancer. On clinical examination, there was a dense, slightly mobile, painless mass about 3.5 × 2 × 1.5 cm in the lower inner quadrant of the left breast, close to the areola. Both nipples were normal and there were no enlarged lymph glands in the neck or axilla. Bilateral mammography showed a homogenous round mass containing environmental calcification with regular contour (Figure 1), and this finding was confirmed by ultrasonography. An antero-posterior radiograph of the chest and the abdominal ultrasound showed no abnormalities. Laboratory findings were normal, except that the echinococcal hemagglutination test was positive 1/100 (a titer > 1/100 is considered a positive result) and CA 19-9 had an increased level of 41 kU/L (value range 0.0–39.0 kU/L). In light of the image scanning result and laboratory findings, we suspected a hydatid cyst because the patient came to our department from an epidemic area. Thus, the mass was excised completely in the operation. In macroscopic examination, the wall thickness of the cyst was about 0.1 cm with a dirty-white appearance. Mural calcification was observed on the cyst wall. Laminated membrane was not observed. Histologic examination confirmed the diagnosis of hydatid disease. For main differential diagnosis, postoperative abdominal ultrasonography, chest radiograph, thoracoabdominal CT, and bilateral mammography were carried out, but they did not reveal other parasitic or nonparasitic cysts anywhere. After an uneventful recovery, she was treated with an average dose of 10 mg kg⁻¹ day⁻¹ of albendazole for a short course of 3 months, which might have contributed to the decreasing recurrence rate. Three months after the operation, CA 19-9 value returned to 12 kU/L (value range 0.0–39.0 kU/L).

DISCUSSION

The breast is a very rare primary site for hydatid cyst’s lesion. Reported incidence of hydatid cyst in the breast is 0.27–0.37%, and the majority of the cases reported in English or non-English literature are single case reports. Hydatid cysts of the breast rarely take part in great patient series of systemic hydatid disease.

Breast hydatid generally occurs in females. Age range is wide, but most of the patients are within the third to fifth decades. Typically, the patient presents with a long history of a breast lump. Mammography reveals opacity with calcification, which must be differentiated from that of a calcified fibroadenoma, simple cyst, or well-circumscribed carcinomas. Sonography does not easily distinguish hydatid cysts from simple or complicated cysts. In the absence of breast abscess, magnetic resonance imaging may suggest a hydatid cyst of the breast. Indirect hemaggulination tests are rarely performed and vary in degree of reliability. However, serologic tests may help to confirm the diagnosis even in the absence of associated liver and lung involvement as in the reported case.

Actually, elevated serum values of the CA 19-9 have been found in the sera of patients with pancreatic cancers; however, elevated CA 19-9 was not shown in breast cancers. It is also elevated in patients with benign cysts diseases such as retroperitoneal, thymic pancreatic, hepatic, and bronchogenic cysts. A putative explanation for this elevated CA 19-9 could be provided by differences in the cellular and humoral immune responses to hydatidosis. In a recent study by Ewing and others, increased CA 19-9 levels in patients with liver hydatidosis have been reported. The theories about this finding include substances that may be originated from the parasite or substances that may be synthesized by the host in response to infection as observed in viral hepatitis. Elevated levels of CA 19-9 in extrahepatic hydatid disease, as in the reported case, supports the former theory about its mechanism.
With the help of serologic tests and radiologic investigations, it should not be difficult to differentiate other benign tumors of the breast from hydatid cyst. Complete excision of the hydatid cyst is generally recommended as a rational therapy. Accidental implantation may be prevented by irrigation of the cyst bed with 3% saline solution. Decreasing CA 19-9 levels after curative resection was observed in the current case, as discussed previously by Pfister, but there is still a small and perhaps insignificant question due to this case. This issue should be confirmed with other studies.

In conclusion, hydatid cyst in the breast is a rare condition but should be differentiated from breast carcinoma and other lesions observed in the breast tissue. Imaging modalities and serologic investigations facilitate diagnosis by clinicians in the presence of competent pathologic examination. Moreover, the possibility of a hydatid should be born in mind in the differential diagnosis of palpable mass and elevated CA 19-9 levels in the breast, especially in endemic areas.

Received May 20, 2004. Accepted for publication March 20, 2005.

Note: A preliminary account of some data contained in this case report will be presented in abstract form to the 40th Annual Meeting of The European Society for Surgical Research Congress, which will be held on May 25–28, 2005 in Konya, Turkey.

Authors’ addresses: Bulent C. Yuksel, Hakan Ozel, Tezcan Akin, Fatih M. Avsar, and Suleyman Hengirmen, First Department of Surgery, Ankara Numune Training and Research Hospital, 06100, Sihhiye, Ankara, Turkey.

Reprint requests: Bulent C. Yuksel, Eryaman 2 capacité, Demirer bloklari, A2-2, Açıcalı Apt. No:5/50, 06930, Ankara, Turkey, Telephone: +90 0 312 280 70 38, Fax: + 90 0 312 418 27 60, E-mail: bulent.yuksel@turktelekom.com.tr.

REFERENCES

10. Safi F, Berger HG, Bittner R, Buchler M, Krautzberger W,


