PRIMARY CERVICO – AXILLARY HYDATID DISEASE.

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Mr. Editor:

Uruguay is one of the countries with the highest prevalence rates of *Echinococcus granulosus* infection. The organs most frequently affected by echinococcal cysts are the liver and the lung (80%). However, in 15% to 20% of the cases, the embryo passes through the capillary systems in the liver and in the lungs, reaches the general circulation, and determines the formation of hidatid cysts (HC) in atypical sites. This latter pathway is so rare that subcutaneous tissue accounts for only 0.5% of the cases.

The current paper presents an unusual case of primary hydatid disease of the soft tissues in the cervico-axillary region, stressing the importance of adequate diagnosis and surgical treatment.

OB is an 84-year-old female who presented with a small painful inflammatory lump in the base of the right axilla. The physical examination also showed a large soft swelling on the right side of her neck, which had progressively developed during the last six years, without any symptoms.

In the last six months, she had begun to experience progressive impairment of the arm’s movements and occasional fulgurant paresthesias of the right hand with paresia of the muscles of the hand and forearm, predominantly in the hand flexors. She would occasionally present pain and fleeting paresthesias on the medial aspect of hand and forearm, with no impairment of reflexes. Normal muscular tone, with no atrophies. Normal sensitivity on the physical examination.

The inflammatory mass in the axilla was diagnosed as suppurative hidradenitis and on drainage it produced pus and several hydatid vesicles.

Abdominal ultrasound showed no liver or spleen hydatid involvement. No evidence of intrathoracic hidatid disease was found found either through chest X-ray or computed tomography (CT). However the CT scan of the chest and neck showed multiple hidatid cysts in the soft tissue of the right side of the neck (Figure 1), extending through the cervico-axillary passage behind the clavicle, inside the axilla (Figure 2).

No evidence of muscle or bone involvement. The serological test by the ELISA method was positive for hidatid disease.

She was then operated on with the diagnosis of primary cervico-axillary echinococcosis. Two independent approaches were used, cervicotomy along the sternocleidomastoid muscle, and axillary-transverse incision at the base of the axilla. The brachial plexus was found to be displaced forward and elongated by the hidatid cysts, but *in toto* excision of the hidatid tissue was possible without injuring any vascular or nervous structures. A suction drain was left in place. The post-operative course was uneventful and the drain was removed on the third day.

Following surgery, the patient has experienced no pain or paresthesias, although she remained with a slight motor deficit limited to the flexor muscles of the hand and forearm in spite of post-operative physical therapy.

Five years after surgery, there is no clinical evidence of recurrence.
The pathologist reported multiple HC, with a fibrous adventitia with small calcifications and a foreign body granulomatous reaction, surrounded by fatty tissue. The overall mass weighed 935 grs.
Hydatid disease of the subcutaneous tissue can be diagnosed by its anatomical location. To be defined as such there should be no muscular tissue in the adventitia. As consequence, this presentation should not be confused with primary muscular hidatid disease extending to the subcutaneous tissue. Primary HC are the result of a haematogenous dissemination of the embryos of *E. granulosus*, which go through the capillary bed of the liver and the lung, finally reaching more distant sites such as the subcutaneous tissue. In our case, however, the finding of multiple HC surrounded by a common fibrous adventitial capsule, reasonably leads us to assume that the primary cyst ruptured in the surrounding subcutaneous tissue and originated a secondary hidatid disease. The presence of calcifications in the adventitia support this hypothesis, since that has never been found in primary HC of the soft tissues.

It is difficult to establish the real incidence of these conditions in the published series (1,2,3). However, HC of the soft tissues account for 3% of the total number of HC reported, with only 59% of these corresponding to HC of the subcutaneous tissue (1). Schaefer (2) which reported the experience on 59 cases, showed that only 3 of these had occurred in the subcutaneous tissue.

Clinical presentation of such cases usually occurs as an asymptomatic subcutaneous tumor developing over a long period of time. Complications are uncommon, but sometimes symptoms develop as a consequence of compression of neighbouring structures or pericystic infection and external cyst rupture. In our patient, the long asymptomatic growth of the hidatid mass during 6 years led to an anterior and downward displacement of the brachial plexus (with a late onset of symptoms) and even to deformation to the thoracic wall, which is evident in the CT scan. Compression of bones by neighbouring cysts determine bone sclerosis, secondary to impairment of periosteal vascularization; it is totally different from the multiple osteolytic bone lesions characteristic of primary hidatid location in the bone (4). External fistulization is reportedly common (1) but suppuration is mentioned only in 20% - 30% of the cases.

Diagnosis is confirmed by ultrasound or CT. Both imaging methods show one or more cystic tumors, with an internal tomographic density between 0 and 6 Hounsfield units. (5) It is highly convenient to use intravascular contrast to adequately show the relationships with vascular structures and allow a safe surgical approach to the disease. In countries with a high prevalence of Echinococcus infection such as Uruguay, the "honeycomb image" is highly suggestive of echinococcosis, (one cyst with multiple daughter vesicles or multiple HC)

The treatment of hidatid disease of the soft tissue is surgical, and it is aimed at preventing compression of surrounding structures and secondary rupture of the cysts. Total pericystectomy with excision of all the HC and the fibrous adventitia is the ideal treatment, and it could be done easily in our observation in spite of anatomical distortion of the cervico-axillary region. Sometimes,
however, there are very dense adhesions of the adventitia to major vessels or nerves and in such cases, our view is that all cysts should be excised with a partial removal of the adventitia, leaving the area near the vessels or nerves in order to prevent their surgical damage. This case serves to illustrate the successful outcome of complex and unusual form of HC which is fortunately not frequently found in surgical practice.

REFERENCES.


TEXT FIGURES.

Figure 1. View of the neck, showing only one HC. with three hydatid
vesicles inside it.

Figure 2 - Right axilla is completely occupied by hydatid cysts with the typical "honeycomb image". Chest wall is deformed without hydatid invasion.