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doi:10.1016/j.aller.2010.10.001

Recurrent angioedema and urticaria in patient with severe osteoporosis☆

To the Editor,

Hydatidosis is a parasitic disease caused by encysted larvae of *Echinococcus granulosus* which have the dog as the definitive host.

The presence of the cyst may be an incidental finding in many cases, often already present from childhood[1].

The determination of antibodies to Echinococcus is usually indicated in the study of chronic urticaria and angioedema because the hydatid cyst can cause skin symptoms (rash, urticaria and angioedema), anaphylaxis, pain (if there is an organ compression), haemoptysis...

We present the case of a patient with recurrent urticaria and angioedema who after the surgical removal of a hydatid cyst in liver has an improvement in her skin symptoms.

We present a female aged 69 with no toxic habits or drug allergies known controlled in the allergy service since 2005 for moderate persistent bronchial asthma, mild intermittent allergic rhinitis and food allergy (urticaria after the ingestion of peach and shrimp).

Pathological history

- Severe osteoporosis with loss of 12 cm of her height in recent years, for this reason she is being treated with subcutaneous teriparatide.
- Mild decrease of immunoglobulins without clinical relevance.
- No other relevant history.

Clinical Evolution

The patient remains asymptomatic of respiratory disease and she avoids eating peaches and shrimp. Although the clinical evolution of her asthma and food allergy was good, she began to present recurrent episodes of facial angioedema with lingual involvement that were treated in the emergency service with oral corticosteroids. She also referred generalised urticarial lesions almost daily. There are no dietary transgressions or relation to other specific triggers that might explain the appearance of skin symptoms. Approximately one month after initiating these episodes, the patient suggests that it could be caused by an adverse reaction to subcutaneous administration of teriparatide that she was receiving for osteoporosis. If she suspends treatment for a few days, the skin lesions do not appear.

Allergy studies were performed and we raise the possibility of undertaking a study of drug allergy to assess the involvement of teriparatide in this cutaneous reaction.

Allergy study

- *Prick test with inhalant allergens*: positive to grass pollen, hazel, birch, and *Chenopodium*. Negative for cat and dog dander, latex, dust mites, and fungi.
- *Prick test with food allergens*: positive to peach and shrimp. Negative for anisakis, vegetables, nuts, legumes, fish, meat, milk, eggs, and other fruits and seafood
- *Simple spirometry*: mixed ventilatory disorder (secondary to important kyphosis).
- *Analytical*:
  - normal haemogram, calcium 8 mg / dl,
  - alpha-1-antitrypsin 122 mg / dl, normal thyroid function, antithyroid antibody negative, basal tryptase 1.8 mcg / L.
  - Total IgE 64 IU / ml, IgA 642 mg / dl, IgG 684 mg / dl, IGM 35 mg / dl, IgG1 359 mg / dl, IgG4 5 mg / dl, normal other subclasses, specific IgE to *Phleum* 2.9 IU / ml, shrimp 1.1 IU / ml, peach 3.4 IU / ml, *Anisakis*, *Ascaris*, *Echinococcus* <0.35 IU / ml.
  - ANAS, antiDNAs, rheumatoid factor negative.
  - Complement: normal.
  - Serology HBV, HCV, toxocara and echinococcus: negative.
- *Parasites in stool* (three serial samples): negative.

☆ The case described was presented in January 2009 at the annual meeting of the Catalan Society of Allergy and Clinical Immunology (SCAIC), having received an award for best communication.

- *Thorax radiology*: impingement right costophrenic sinus, no other significant alterations.

Given the radiological alteration it was decided to request a chest TAC.

- *Thoracic TAC*: we observed a calcified mass (6x6 mm diameter) suggesting a possible gallbladder neoplasm versus chronic cholecystitis or xanthogranulomatous. No lung alterations have been observed.

Subsequently, abdominal ultrasound and nuclear magnetic resonance (NMR) were performed without conclusive results and it was decided to operate on the patient. A cholecystectomy with resection of a liver hydatid cyst (with a well-preserved wall structure) was made. Afterwards a preventive treatment with mebendazole was performed.

After cyst surgical removal, at present (after 24 months), the patient remains asymptomatic without submitting any episodes of angioedema or urticaria. She has continued, as previously, the same treatment for osteoporosis without problems. The patient was diagnosed of urticaria and angioedema due to hydatid cyst.

Hydatidosis is a parasitic disease caused by encysted larvae of *Echinococcus granulosus* and dogs are the definitive host[1]. The infection in man (aberrant host) occurs by ingestion of contaminated plants by the larvae or by contact with infected dogs. The larvae can migrate through the bloodstream to any organ[2]. The most frequently infected organs are liver and lungs.

Most cysts could be asymptomatic for long periods of time, in many cases the infection is acquired in childhood[2]. They can also produce variable symptoms depending on the location of the cyst: right upper quadrant pain, vomiting, diarrhoea, haemoptysis... If the cyst breaks, the liquid inside may sensitise the infected individual and this can lead to episodes of urticaria, rash, pruritus, and anaphylaxis[3–5]. In these cases, determination of specific IgE to *Echinococcus* can be useful and can be a tool for monitoring patients who receive surgery or medical treatment for hydatidosis. However, it is not a definitive proof and in our case was not a useful tool for diagnosis (specific IgE to *Equinococcus* was < 0.35 UI/ml)[2,6]. On the other hand, the existence of cross-reactivity with other parasitic infections can cause false positives[6].

In our patient *Equinococcus* serology was negative before and after surgery and the cyst was found through an imaging test. The immune response to infection by *Echinococcus granulosus* in an immunocompetent host usually includes production of IgG, IgM and IgA[6].

Some studies have described skin testing with hydatid cyst fluid obtained after the exeresis of a cyst in an animal, but it is not usually done in routine practice. In our case it was impossible to delay the operation because of the suspicion of a tumour as a more specific study was not indicated.

The germinative layer surrounding the hydatid cyst acts as a barrier that prevents contact of antigens with the bloodstream. Micro-cracks or breaks in this layer must exist to produce antigenic stimulation and generate antibodies that can subsequently be detected.

It has been observed that when the cyst is in the lungs there is a lower percentage of breakage compared to other locations, such as the liver. When hydatid cysts are hyaline and small there is less risk of leaking fluid[1]. This could explain some cases with negative immunological study.

In our case, the cyst was perfectly encapsulated in the liver so it is unclear if the cyst could be related to the skin clinic that prompted the study in this patient.

We considered the possibility that treatment with teriparatide may have provided the encapsulation of the cyst because of calcium retention. Another possibility would be that the hydatid cyst had been just a chance finding and the urticaria and angioedema have been limited spontaneously and that this would have coincided with surgical removal.

Some cases with presence of hydatid cyst at the kidney, liver and adrenal with a negative serology have been reported. Therefore the negativity of the in vitro tests does not exclude the diagnosis.

In clinical practice, other complementary investigations are not usually made to study a hydatid disease (intradermal Casoni, indirect haemagglutination, immunoelectrophoresis...). These specific tests may have allowed a more accurate diagnosis in this patient.

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doi:10.1016/j.aller.2010.10.005