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Review

Solitary subcutaneous hydatid cyst
Review of the literature and report of a new case in the deltoid region

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ABSTRACT

Background: Solitary subcutaneous hydatid cyst is not frequent and the only symptom is generally a silent growing mass. Total excision remains the mainstay of treatment. Aim of the study was to present a case surgically treated and perform a statistical analysis reviewing previous published works in order to define a correct approach to diagnosis and treatment.
Methods: 264 documents from Medline database were considered for primary subcutaneous hydatid cyst cases. Data concerning geographic region, gender, age, job, location, evolving time, history and physical, mobility, diameter, laboratory, imaging, locularity (uni- or multilocular cyst), fine-needle aspiration, preoperative diagnosis, neoadjuvant chemotherapy, treatment, spillage, adjuvant therapy, follow-up and recurrences were ordered in a database and analysed performing t-test, Fisher's test and Pearson's test.
Results: 23 cases, included ours, resulted suitable for our study. Lower extremities were involved in most cases (60.9%) and the thigh represented the most common site (34.8%), whereas upper extremities were the rarest location (8.7%). Patients with head and neck located cysts were younger than those with upper extremities cysts ($P = 0.037$). Patients who underwent multiple imaging approach received a significantly correct first diagnosis ($P = 0.001$) and ultrasonography, unlike other techniques, appeared to be essential ($P = 0.013$).
Case report: A 68-year-old man who lived and worked in his farm in Sicily (Italy) presented with a 30-year-growing mass in the deltoid region measuring 10 cm. Ultrasonography and magnetic resonance imaging strongly suggested hydatid cyst. Therefore the cyst was excised and pathology confirmed the diagnosis.
Conclusion: Solitary subcutaneous hydatid cyst must always be considered in the differential diagnosis of silent growing mass in soft tissues. History and physical associated with ultrasound and magnetic resonance imaging are sufficient to achieve a correct preoperative diagnosis.

Contents

1. Introduction

Hydatid disease is a tapeworm zoonotic infestation caused by *Echinococcus* species which leads to parasitic cysts formation. *Echinococcus granulosus* is, by far, the most common species responsible in the Mediterranean area and, because in this case the disease develops as single cyst, whence the name “cystic echinococcosis”, it

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is the least aggressive and the most treatable form of hydatidosis [1–3].

Even if life cycle of the parasite does not include human beings, they can occasionally ingest eggs becoming intermediate hosts [1,4]. Liver and lung hydatids together account for 90% of all echinococcal cysts [5–7]. Subcutaneous localization of a solitary hydatid cyst on the other hand is not frequent accounting for barely 1.6% of all locations. The way of transmission and development in these cases remains not completely clear, although different hypotheses have been postulated [6,8,9].

The only symptom is usually a painless growing mass [6,10,11] even if inflammatory signs have occasionally been reported [12–14]. Ultrasonography (US) and Magnetic Resonance Imaging (MRI) are the most preferred tools to achieve a correct preoperative diagnosis [15]. However, biohumoral and radiologic diagnostic work-up of the disease, when located in the subcutaneous tissue, is still not standardised.

Total resection is the mainstay of treatment when a single parasitic cyst is located in the subcutis [1,2]. Anthelmintic drugs are often administered especially after surgery [2,16,17]. Risk related to spillage, such as anaphylactic reactions, and recurrence after surgery needs to be addressed.

In this study the Authors, in order to define a correct approach to diagnosis and treatment of primary subcutaneous hydatid disease, collected data from different previous works of the literature and performed a detailed statistical analysis. Moreover, we report our single experience of hydatid cyst in the deltoid region of the left upper arm. Transmission ways of subcutaneous location of the disease and results of the review are discussed.

2. Literature review

2.1. Materials and methods

In January 2013 the Authors searched at Medline/PubMed the exact string: [(echinococc* OR hydati*) AND ("soft tissue" OR subcutaneous)]. It resulted with 264 documents.

Considering that "soft tissue" is not only subcutaneous tissue, the exclusive subcutaneous involvement, which was the target of this study, had to be mentioned and specified, with or without muscular extension. Reported cases with muscle–skeletal, axillary or visceral involvement were ruled out and only "primary/solitary" hydatid cysts were considered. Only papers written in English, French or Italian, available for the Authors, were considered. Our case, presented here, was considered in this analysis of the literature.

Specific collected data were: article's reference, geographic area, patient's gender, age, job, cyst location, evolving time from first observation, history and physical elements associated with the enlarging mass, mobility, maximum diameter in cm, laboratory findings, imaging techniques, uni- or multilocular type of cyst, fine-needle aspiration (FNA), preoperative probable diagnosis, neoadjuvant chemotherapy, type of treatment, intraoperative spillage, postoperative adjuvant medical therapy, follow-up period and recurrences. Every single record, whenever available, was inserted into the database in Tables 1 and 2.

If, according to geographical distribution of *E. granulosus* [3], a country was split in different endemic regions, the town was indicated. Cyst sizes were measured at physical exam or by means of instrumental imaging techniques. If both were discordant the larger imaging-assessed measurement was reported. Laboratory tests were also recorded. In this review, with the term routine blood investigation (RBI) the Authors considered complete blood count (CBC) associated to biochemistry tests. If not specified we considered neither laboratory tests nor imaging scans nor FNA was performed, and no anthelminthic drugs were administered before and/or after surgery.

Mean values and percentages were calculated. Statistical analysis (SPSS 20.0.0) was performed using a two-tailed Student's t-test, a two-sided Fisher's exact test and a two-tailed Pearson's correlation test. A *P*-value of 0.05 was considered statistically significant.

3. Results

Our search criteria resulted in 23 cases [7–27]. All cases were from endemic areas: 15 (65.2%) from highly endemic regions, 4 (17.4%) from endemic countries and 4 patients from sporadic endemic areas. Male patients were 8 (36.4%) against 14 females (63.6%), out of 22 patients with available gender record. Male/female ratio was 0.57. Age, at the time of presentation, ranged from 12 to 73 years with a mean age of 46 years. In 5 out of 6 cases with mentioned employment data, the patients worked as farmers (83.3%).

The most common location was subcutaneous tissue of the thigh in 8 cases (34.8%) followed by gluteal site in 3 patients (13%). Lower extremities were involved in most cases: 14 out of 23 (60.9%), head and neck in 4 (17.4%), trunk in 3 (13%), whereas upper extremities were the rarest location accounting for only 2 patients (8.7%). Patients with upper extremities cysts were older than those with head and neck located cysts (mean age: 67.5 ± 0.7 vs. 32.8 ± 19.4 years; $P = 0.037$) (Fig. 1). There was no relationship between patients' gender and cysts' anatomical distribution ($P > 0.1$). Lesions evolved over a period between 2 days and 30 years from patients' first discovery (mean evolving time: 43.5 ± 88.3 months). A significant correlation between age and evolving time was not found ($P = 0.153$). The anamnestic data referred by all patients was a gradually-growing mass. In most cases, 17 patients (73.9%), this increasing swelling was described as silent and painless. Nevertheless, some patients presented with one or two of the following: pain in 4 cases (17.4%), skin redness in 3 patients (13%), surrounding-lesion dilated veins with positive cough impulse in one case (4.3%), and wasp sting in a single patient. No relationship was found between the presence or absence of signs and symptoms associated with the evolving mass and the anatomical site involved ($P > 0.1$).

Nine cysts out of 14 (64.3%) were mobile on palpation, 2 (14.3%) were semi-mobile and other 3 (21.4%) fixed upon lower planes. Maximum diameter of the cysts ranged from 3 to 20 cm (mean max diameter: 8.7 ± 4.5 cm). Cysts' size was not statistically different in relation to the four body regions ($P > 0.1$). Moreover, the diameter was correlated neither to age of the patients ($P = 0.611$) nor to evolving time from first observation ($P = 0.941$), even when in both cases single locations where separately considered ($P > 0.1$).

RBI was performed in 15 patients (65.2%) but resulted within normal range in 11 (73.3%). An increased erythrocyte sedimentation rate (ESR) of 40 or 60 mm/h was found in 4 patients (26.7%) out of 15 in which biochemistry tests were performed. CBC, performed in 18 patients (78.3%), was positive only in a single case (5.6%) showing eosinophilia with 10% of eosinophils. Urinalysis (UA), performed in 5 patients (21.7%) was always negative. Parasitological serologic tests, including enzyme-linked immunosorbent assay (ELISA), indirect hemagglutination antibody test (IHAT), complement fixation test (CFT), western blotting (WB), indirect immunofluorescence antibody test (IFAT) and immunodiffusion test (IDT), were performed in 14 patients (60.9%). Positive results were observed only in 4 cases (28.6%).

US of the lesion was performed in 16 patients (69.6%), X-ray in 7 (30.4%), CT scan of the cyst in 11 (47.8%) and MRI in 9 (39.1%). The cysts were described as multilocular in 16 cases out of 19 (84.2%) and unilocular in 3 patients (15.8%). Maximum diameter of unilocular cysts was similar to that of multilocular cysts ($P = 0.212$). FNA was performed in 4 cases (17.4%). Preoperative diagnosis revealed correct in 11 out of 16 patients (68.8%) and incorrect in 5 (31.3%). Considering the number of imaging techniques the first probable diagnosis revealed significantly correct in patients who underwent multiple imaging approach. Mean number of imaging techniques was 2.27 ± 0.786 when preoperative diagnosis was correct vs. 0.60 ± 0.548 when revealed otherwise wrong ($P = 0.001$) (Fig. 2). Furthermore, ultrasonography,