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RESEARCH ARTICLE

PRIMARY HYDATID CYST OF THE THIGH - ABOUT A CASE AND REVIEW OF THE LITERATURE

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Abstract

Hydatid disease is a parasitic infestation of humans and herbivorous animals, caused by *Echinococcus granulosus*. Dogs and some wild carnivores, such as foxes, are definitive hosts, harboring worms in their intestines. Musculoskeletal cysts represent 0.7 to 3% of the total number of cases of hydatidosis. Primary muscular hydatidosis without involvement of the thoracic or abdominal organs is extremely rare. Intramuscular infestation can mimic a soft tissue tumor resulting in inappropriate rupture of the cyst with the associated risks of anaphylaxis and dissemination to other organs. Preoperative evaluation is therefore essential to avoid life-threatening complications. We report the case of a 64-year-old patient presenting with a cystic swelling of the thigh admitted to the emergency department for a ruptured hydatid cyst. Surgical excision with complementary pharmacological treatment is necessary to obtain complete cure.

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Introduction:-

Echinococcosis (hydatid cyst disease) is a zoonotic infection most commonly caused by the larvae of *E. granulosus*, and the larvae of *Echinococcus multilocularis*, *Echinococcus vogeli*, and *Echinococcus oligarthrus* that inhabit the small intestine of carnivores [1-2]. The adult worms produce eggs which are released with the feces and spread in various ways, such as by wind, water or flies [3]. After ingestion by the host, the embryos migrate through the intestinal wall and are either arrested in the capillary bed of the liver evolving into hepatic cysts, or manage to enter the systemic circulation thus ending up in distant organs. The lungs, brain and muscles or bones are the most frequently affected distant organs.

Other manifestations are found in 15% of patients, with the skeletal system accounting for 1–4% of all cases [4]. Voluntary muscles are a very rare site of infection, accounting for < 1% of the total [5]. Muscle hydatid cyst is uncommon because the presence of lactic acid creates an unfavorable environment for growth [6]

We report here a case of primary subcutaneous hydatid cyst of the left thigh in a 63-year-old patient presenting for 2 years with a painless cyst-like mass without inflammatory signs, complicated by a cystic rupture.

Observation:-

This is a 64-year-old patient, operated for valvular heart disease 10 years ago under Sintrom and having the notion of contact with dogs. Admitted to emergency for ruptured hydatid cyst.

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The clinical examination objectifying a stable patient on the hemodynamic and respiratory level with the demonstration of a painful and diffuse cystic mass attached to the muscle with signs of local inflammation located on the posterior face of the left thigh evolving in a febrile context 2 days ago. (Figure 1)

The analysis of biological data (NFS, blood ionogram, liver test) did not show any particularity, the hemostasis test showing a TP at 89% and INR at 1.25. Hydatid serology is positive. A soft tissue ultrasound indicating a voluminous cystic mass containing a floating membrane and an MRI of the thigh showing a voluminous intracompartmental cystic formation on the posteromedial aspect of the thigh in favor of a remodeled cyst. A thoraco-abdomino-pelvic CT scan showing bilateral pulmonary micronodules of infectious origin with hepatic lesion of segment VII of the liver. A recent cardiac evaluation was made objectifying a mechanical mitral prosthesis of good functioning, the cardiac chambers of size and function preserved with non-dilated right cardiac chambers.

The preoperative preparation of the patient consisted in stopping anticoagulants 5 days before the procedure with a relay by curative dose LMWHs stopped 24 hours before the procedure.

The procedure is performed under general anesthesia balanced with fentanyl (4µg/kg), etomidate (0.3mg/kg), and rocuronium (0.6mg/kg). Anesthesia is maintained by sevoflurane and fentanyl with good hemodynamic tolerance. The surgical position was in the right lateral decubitus view of the patient's terrain. The intraoperative went without incident. The operative gesture consisted of gentle aspiration of the cystic contents with abundant washing of the muscle compartment and the wound was closed with a suction drain on site. (Figure 4)

After 1h30 of the gesture, the patient was extubated without incident, then sent to the surgery department with resumption of anticoagulation within 24 hours of the postoperative period according to the hemorrhagic risk evaluated by the surgeons with a cardiological opinion for therapeutic adjustment until adaptation of the INR level.

Discussion:-

Hydatid disease is an anthrozoosis caused by *Echinococcus* species; humans represent intermediate hosts in the life cycle of the parasite when they occasionally ingest eggs via contaminated food or water [7,8]. The shell of parasite eggs opens in the presence of acidity of the upper gastrointestinal tract. The oncospheres, thus released in the small intestine, penetrate the intestinal wall and are transported by the portal circulation into the liver. After passing through the portal filter, they could potentially reach every organ with the lungs serving as a secondary filter. Possible dissemination via the lymphatic route has also been reported [9,10]. The majority of hydatid cysts occur in the liver, lungs, or both.

Thus, primary soft tissue involvement is very rare; resulting in a diagnostic challenge [11]. It is widely postulated that the low prevalence of this form of the disease is potentially due to physical barriers, hematogenous dissemination of cysts created by hepatic sinusoids and pulmonary capillaries. Furthermore, it is widely believed that the higher concentration of lactic acid in skeletal muscle and mechanical factors, such as contractions, make encystment less likely. The most common musculoskeletal sites include the pelvic, thigh, and paraspinal muscles. Another compatible hypothesis is that of

Cysts can present in many ways, including a slowly growing mass with variable pain or a sudden onset of symptoms due to the rupture of the cyst. Rupture of the cysts releases antigens into the muscles causing an inflammatory response which may be complicated by secondary bacterial infection [13].

Diagnosis is based on clinical evidence such as anamnestic data regarding origin and history of exposure to livestock, presentation as well as radiological support. Hydatid serology is often negative in 1 out of 2 cases of extra hepatic hydatidosis, in our case the serology was positive. Ultrasound is a very useful diagnostic imaging tool for hydatid disease. Not only does it detect hepatic cysts, but it also allows classification and staging of liver hydatid disease [14, 15]. Nevertheless, we strongly believe that the sensitivity of ultrasound for floating membranes, daughter cysts and hydatid sand in purely cystic lesions makes it very ideal in the initial workup of even rare localizations. In general, MRI has higher specificity and sensitivity than ultrasound for hepatic hydatid cyst [16]. It also allows a better characterization of the anatomical relationships and facilitates the surgical management of the cyst, as was the case in our patient.

It is essential to establish a definitive preoperative diagnosis of skeletal muscle hydatid cysts. This contraindicates certain treatment options like marginal excision or incisional biopsy due to the likelihood of dissemination and anaphylactic shock in the event of a spill [17]

Intraoperative leaks or accidental ruptures of hydatid cysts of the liver may cause anaphylactic reactions secondary to systemic absorption of hydatid fluid. The mechanism of these reactions is complex. In some cases, it is typically a type I hypersensitivity reaction linked to immunoglobulin E in response to the high plasma concentration of Ag of *Echinococcus*. The anaphylactic or anaphylactoid reaction can also be secondary to complement activation with release of anaphylatoxins. In vitro, hydatid fluid activates the alternative pathway of complement with the formation of C3a. Anaphylactic manifestations can take on very different aspects, such as bronchospasm and/or anaphylactic shock [17]. In our case, the gesture had passed without incident.

Conclusion:-

Muscle infestation by *Echinococcus* is a rare but important entity, more frequent in rural areas. This case of primary skeletal muscle hydatidosis illustrates the significant morbidity associated with hydatid disease. Preoperative diagnosis is important in the management of the hydatid cyst. Surgical excision is the main modality of treatment. During surgery, antiscolicide solutions and meticulous surgical technique go a long way in preventing the recurrence of this disease.

Figures



Figure 1:- Hydatid cyst of the posterior thigh preoperatively.



Figure 2:- Bulky intra-compartmental cystic formations on the posteromedial aspect of the thigh.



Figure3:- MRI image showing multiple hydatid cysts in the muscle mass of the thigh.



Figure 4:- Postoperative specimen with aspiration material from ruptured hydatid cysts.

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