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#### **Research Article**,

### Vertebro-Medullaire Hydatidosis: About Three Cases Report And Review Of Literature

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#### Abstract:

Echinococcosis is an anthropozonosis that occurs in underprivileged areas near sheep and dog farms. Vertebral hydatidosis is rare but dramatic, with a poor prognosis, in spite of an essentially surgical treatment, the recurrences are very frequent and aggravates the prognosis. We report three cases of vertebromedullary hydatidosis involving a 51-year-old woman and two men aged 25 and 48 with an average age of 41 years. The assessment includes standard radiographs, CT and MRI, hydatid serology, abdominal ultrasound for other locations. The clinical picture is dominated by a syndrome of spinal cord compression: parapaesthesia, paraplegia and sphincter disorders, back pain or cervicalgia depending on the location. The seat is dorsal for two cases (D1-D2 and D3-D7) and cervico-dorsal for one case (C7-D3) The MRI is the exam of choice. Treatment is Surgical consists of a cystic vesicle evacuation with wide resection of infiltrated tissue associated with the antiparasitic medical treatment (albendazole) .The diagnosis of hydatidosis was confirmed on histopathological examination. Recidivism frequents two of the three cases. The medullary involvement is due to the development of cysts inside the canal or para vertebral. To think of vertebral hydatidosis in front of a medullary compression table in an endemic zone, Algeria is one of them. Imaging makes it possible to affirm the diagnosis, to assess the lesions and to follow the evolution. Radical surgery followed by medical treatment with albendazole represents the best therapeutic strategy however the frequency of recurrences darken the prognosis.

Key words: Medullary echinococcosis, compression, recurrence, therapeutic difficulties.

#### **Introduction :**

Hydatidosis is a parasitic disease that is endemic in many traditional livestock producing countries, including Algeria (14,39). It can reach any organ with a predilection for the liver and lung. Vertebromedullary hydatidosis is rare. representing less than 1% of all localizations of the hydatid cyst. It remains the most frequent and most serious manifestation of bone hydatidosis. The objectif of our work is to establish a well codified action plan to lead to a better management of our patients for a better life and to limit the economic effects of this affection, to discuss the therapeutic difficulties and the bad prognosis of this disease which is serious because

of the frequency of recurrences and neurological complications.

#### Epidemiology of hydatidosis in the maghreb :

Hydatidosis is an endemic disease in the Maghreb (Morocco, Algeria, Tunisia and Libya) (39). The publications concerning hydatidosis in North Africa concerned 93% Tunisia, Morocco and Egypt, the other countries (Algeria, Libya, Sudan) only concerned 7% of the indexed publications. In Algeria, the prevalence rate of hydatidosis reported by Dar and Alkarmi (14) is 3.4 to 4.6 cases per 100 000 inhabitants, the rate reported by Seimenis in 2003 (32) being 1.8 to 2, 3 cases per 100,000 inhabitants. In Algeria, the ovine strain of

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E. granulosus appears the most incriminated in human infection in 2003 (5), although a camelina strain has also been demonstrated, with possible crosses between strains (28).

#### Materials and methods:

We conducted a retrospective study, spread over 11 years, on the medical records of 03 cases of medullary vertebro hydatid cysts operated at the neurosurgery department of Mustapha PACHA Hospital in Algiers between January 2007 and December 2018. Various parameters were exploited from the medical records of the department.

#### **Case report 1:**

The patient Mr. KH, 34 year old, plasterer, who lives in the rural area of CHLEF, is hospitalized in September 2007 in the neurosurgery department of Mustapha PACHA Hospital in Algiers for the exploration of a posterior cervical mass. The history of the disease dates back to 5 years marked by the appearance of a subcutaneous nodule 1 cm long which has increased the volume for a year. The physical examination revealed a large posterior soft, multi-nodular cervical mass measuring 10 cm long. There are no neurological disorders apart from neck pain, the rest of the somatic examination is normal. There is no inflammatory syndrome or eosinophilia. Leukocytes are at 6500 / mm3. Calcemia, phosphoremia and electrophoresis of blood proteins are normal.

Cervical computed tomography (CT) scan reveals fluid formation of the posterior cervical soft tissues, located at the level of the retro-spinous greasy lodge, which comes into contact with the muscular structures namely the muscles, large coplexus, transverse neck and splenius. of the head. There is no bone lesion in contact with this cystic formation, which is multilocular, well limited by a regular wall, taking the peripheral contrast and measuring 80x40 mm (Figure 1a). To confirm the non-invasiveness of the spinal cord or duct, magnetic resonance imaging (MRI) was performed, confirming the diagnosis of cervical echinococcosis, which is in the form of a large multi-cystic formation of 98x55 mm wide since C1 to C6 without intracanal extension. (Figure 1b)



Figure 1a: Axial CT scan with injection of contrast agents, multiple hypodense vesicles enhancing on the periphery



Figures 1b: Spinal MRI: T1 sagittal sections on the left and T2 on the right (images of expanded hydatid vesicles from C1 to C7) axial T1 cuts in the middle: post cervical hydatid vesicles. Abdominal and cardiac ultrasonography and chest x-ray are normal. The hydatid serology (enzyme immunoassay by the Elisa technique and immunoelectrophoresis) is negative. The patient received surgical treatment followed by medical treatment with benzimidazole derivatives (albendazole). The clinical evolution is favorable.

#### **Case report 2:**

The patient FB 51-year-old, living in the urban area of Algiers, with a pathological history of penicillin allergy and goitre undergoing treatment, was admitted on 21 January 2016 to the neurosurgery department of of Mustapha PACHA Hospital in Algiers, as part of the emergency for the management of dorsal spinal cord compression. At the admission, the clinical examination finds patient conscious and copperante who presents an injury syndrome made of back pain with intercostal neuralgia, a sub lesional syndrome made of spastic paraparesis. She presents hypoesthesia of both lower limbs, urinary incontinence sphincter disorders. Babinski's reflex is bilateral and osteotendinous reflexes are abolished. The tele-thorax objectified a left basithoracic opacity. To better explore the consequences on the medullary axis, magnetic resonance imaging (MRI) was performed, confirming the diagnosis of vertebral echinococcosis by showing images of hydatid vesicles characterized by a hyposignal on T1 sequences and a hyper signal on T2 sequences, intraductal extradural epidural posterior to the extended thoracic stage of T4 to T8 (Figure 2). There is no peripheral neuropathic involvement detected with EMG



Figure2: Coronal T1 gado cut on the right, showing an extended thoracic paravertebral mass exerting a mass effect on the vertebral column and its contents The patient benefited from bone decompression marrow by performing а laminectomy from T4 to T8 followed by a puncture of the two cystic formations with respect to T5-T6 and T7-T8 and ablation of the cystic parts. The punctured liquid was clear. The parasitological study favors a hydatid cyst. The patient is put on medical treatment based on benzimidazole derivatives (albendazole). The evolution is favorable with a decline of one year. On August 30, 2018, the patient was readmitted for the second time as part of the emergency for the management of spinal cord compression, she presented the same clinical picture of the first hospitalization; Magnetic resonance imaging (MRI) was performed urgently, confirming the diagnosis of the recurrence of 3 vertebromo medullary hydatid cysts, extended from T3 to T8 with an intraductal extension that represses and compresses the spinal cord, with T6 myelopathy. recurrence of hydatid cysts The induced somesthesic disorders of the lower limbs. The patient benefited from a dorsal medullary decompression followed by the evacuation of the 3 hydatid cysts and fibrosis. The clinical evolution is favorable.

#### Case report 3:

Mr. M. D, 37 year old, a worker, lives in the rural area of CHLEF, operated in May 2013 for a right lung hydatid cyst. This patient received three hospitalizations at the neurosurgery department of Mustapha PACHA Hospital in Algiers, for the management of spinal cord vertebral cysts. 1. 1st hospitalization: in June 2013 for cervical vertebro-medullary cervical cyst extended from C7 to D1 (Figure 4).

2. 2nd hospitalization: in July 2016, for the management of the recurrence of an extensive medullary vertebro cystic cyst from C7 to D3 (Figure 4).

3. 3rd hospitalization: in February 2017, for the management of the recurrence of an extensive medullary vertebro hydatid cyst from D1 to D2.

During the three hospitalizations, the patient presents the same clinical picture: a pyramidal syndrome with a spastic paraparesy, ROT are abolished at the level of the lower limbs, hyperesthesia at the level of D1, sphincter disorders with urinary incontinence type. The thoraco CT scan performed at the second hospitalization in 2016 showed the presence of aggressive pleuro-parenchymal and aggressive bone cysts in the right lung(Figure 3). Measures : Cervico-thoracic pleural fluid mass: 80x70mm Parenchymatous fluid mass: 2cm Aggressive 10mm bone mass interesting the vertebral body of d1 with medullary extension and the anterior arches of the first 2 right sides.



Figure 3: Cervico-thoracic CT in favor of aggressive pleuro-parenchymal and aggressive bone cysts of the right lung.



Figure 4 :T2 cervico-thoracic MRI: multi cystic osteolytic lesion, blowing and breaking the cortex of the superior cervicothoracic vertebrae from C7 to D3.Measuring 94 \* 60 \* 42mm

Secondary compression of the dorsal cord with signs of myelopathy extended to 20mm in height. REAR SPINAL SCREW PLATE (2013) The treatment was surgical, aggressive, resumption of the initial incision and enlargement of the laminectomy. Evacuation of the vesicles and the fibrous shell which surrounds them. The evolution was favorable, significant motor recovery. The pulmonary localization has been entrusted to the good care of the thoracic surgeons. Medical treatment with Metronidazole due to lack of availability of Albendazole. 2nd recurrence of hydatid cyst in February 2017

The patient returned to the same clinical picture of spinal cord compression:

- Spastic paraplegia.
- ROT vivid and diffuse.
- Bilateral babinski.



# Figure 5: Persistence in place of a bulky expansive cervical thoracic vertebral process staged from C7 to T3.

It extends widely to the peri-medullary space from C7 to T2 responsible for severe medullary narrowing at T1-T2 with signs of secondary myelopathy(Figure 5). The patient to benefit from a third evacuation of the cystics, decompression of the marrow as well as a large excision of the vesicles invading the para vertebral muscles. Abundant wash with hypertonic saline. The patient is then referred to infectious diseases for further medical treatment

#### **Discussion :**

Widespread in the world, hydatidosis is endemic in Algeria. Its incidence is estimated at 0.9% (in 2017). It is an anthropozoonosis due to the development of the larval form of echinococcus granulosus. The man represents an accidental host. Bone involvement is rare: 0.5 to 2.5% of all hydatid sites (21). It is due to the phenomenon of "paradoxical embolism": during a sudden increase in intra-abdominal pressure, the blood from the portal system would drain into the spinal plexus, avoiding the hepatic and pulmonary filters (4). Vertebromedullary hydatidosis affects the spine, which accounts for 40 to 50% of all bone sites and is of decreasing interest in the dorsal (80%), lumbar (18%), sacral and cervical segments (1%) (1, 7, 9). The vertebral body is more frequently affected than the posterior arch (13,16). In our observation, the Vertebromedullary hydatidosis is located at the dorso-lumbar hinge.

#### Mri interest:

The hydatid vesicles, in their typical form, appear oblong MRI, in "flattened sausage", with very thin walls, without septa, their signal is pure liquid hypersignal (hyposignal in T1. on T2). Sometimes, the appearance is less typical and it is the difference in intensity on the T2-weighted sequences that allows, according to some authors, to determine the viability of the cysts (3, 11, 20). MRI is therefore an excellent examination for the pretreatment assessment of the Vertebromedullary hydatidosis since it allows, firstly, to confirm the hydatid nature of the ossifying abscess, second, to specify the extent of the lesions and finally to determine the degree of viability of vesicles and thus the prognosis. It also allows prolonged surveillance of possible recurrences (6).

## Cause of reccurence of vertebromedullary hydatidosis (infiltration):

In fact, contrary to conventional visceral localizations, the development of the parasite in the bone medium responds to a particular mode resulting in a microvascular infiltration without precise limits, by diverticular budding and exogenous vesiculation without encystment and with diffuse infiltration of the spongy tissue, conferring on the disease all its seriousness and explaining the therapeutic difficulties (19). The vertebral localization represents the most frequent form (41%) and the most serious of the injuries of the hydatidosis bone, unlike the visceral localizations, the bone attack is infiltrative diffuse, badly limited because of the absence of weed. The extension is low noise, exogenous vesicles, hence the diagnosis late. Faced with this aggression, the bone does not react. In the majority of cases, it is a somatic bone disease with an extension in the spinal canal Medullary involvement is secondary to the development of cysts within the canal. This preponderance somato-epidural is explained by the richness of the vascularization of the vertebral djaafer m et. al. / Vertebro-Medullaire Hydatidosis: About Three Cases Report And Review Of Literature

body and the epidural space compared to the vascular poverty of the marrow and its envelopes. Perop : The chemical sterilization of scolex intraoperatively (hydrogen peroxide and formalin) remains of uncertain efficacy (5).

#### Treatment:

The treatment of echinococcosis is always surgical, it consists in lifting the radiculomedullary compression.

The most commonly adopted attitude is decompressive laminectomy, but it is often difficult, sometimes mutilating, and does not protect against recurrence. The recurrence is very frequent because the resection is often incomplete because the bone attack is poorly limited, infiltrating.

#### Albendazole :

The place of medical treatment based on benzimidazole derivatives (albendazole) is not negligible for some authors. Indeed, it seems to improve the painful symptomatology in some cases (2,15). However, its effectiveness on the evolution and sterilization of lesions is still debated (5, 6). It has a place especially in the inoperable forms, or in case of refusal of the surgery, but also as adjuvant therapy to the surgical treatment (1, 4).

#### **Prognosis:**

The prognosis of the HV is thus reserved because of frequent recurrences and the neurological risk emphasizing the interest of prevention (6, 10, 21).

#### **Conclusion:**

Vertebromedullary hydatidosis poses diagnostic difficulties but especially therapeutic because of the large number of recurrence even in the surgery is often large and mutilating. It is a local malignancy affection "white cancer". The neurological and functional prognosis depends on of the lesions the severity preoperative. Monitoring by CT and MRI allows the early diagnosis of recurrence even before the appearance of clinical signs. Albendazole appears to be the molecule of choice, although the duration of treatment is controversial; The best weapon is prevention.

#### References :

1. akahaddar a, gourinda h, el alami z, el madhi t &miri a - kyste hydatique du sacrum chez une adolescente.a propos d'un cas. rev rhum, 1999, 66, 331-333.

2. bhatoe hs, bakshi ss & swamy gln – trichinoechinococcosis t5. j neurosurg, 2000, 92, 225-228.

3. briant jf, richez p, belliol e, barea d, raillat a et al- atteinte osteo-articulaire d'origine parasitaire l'echinococcose osseuse. j radiol, 1998, 79, 1351-1357.

4. bruschi f, ortona e, ioppolo s, siracusano a & bonadio m - immunochemical and molecular characterization of vertebral hydatid fluid. scand j infect dis, 1999, 31, 322-323.

5. el andaloussi m, yousri b & aboumaarouf m – hydatidose vertebrale. à propos de trois cas. rev chir orthop, 2001, 87, 392-396. 6 2001, 87, 392-396.

6. evliyaoglu c, keskil s. possible spontaneous "birth" f a hydatid cyst into the lateral ventricle. chlids nerv syst, 2005; 21:425-8.

7. garcia-vicuna r, carvajal i, ortiz-garcia a, lopezrobledillo jc, laffon a & sabando p - primary solitary echinococcosis in cervical spine. spine, 2000, 25, 520-523.

8. goasgun j, bequet d. parasitologie du systeme nerveux central. encycl med chir neurologie, editions techniques, 1991 ; n°17052

9. hamden ta & al-kaisy a - dumbbell hydatid cyst of the spine. spine, 2000, 25, 1296-1299.

10. hernigou p, nabih a & goutallier d- hydatidose vertebrale : complication, apport de l'imagerie mo

11. kabbaj-el kouhen n, dafiri r, el ouahabi a, el khamlich & imani f - kyste hydatique intradural lombaire isole.j.radiol, 1999, 80, 147-149.

12. kalaitzoglou i, drevelengas a, petridis a, palladas p. albendazole treatment of cerebral hydatid disease : evaluation of results with ct and mri. neuroradiology, 1998; 40: 36-9.

13. keller tm, schweitzer js, helfend lk & chappel t -treatment of progressive cervical spine instability secondary to hydatid disease. a case report. Spine, 1997, 22, 915-917.

14. lam ks, faraj a, mulholland rc & finch rg – medical decompression of vertebral hydatidosis. spine, 1997, 22, 2050-2055.

15. larbaoui d, alloula r, (1979). «etude epidemiologique de l'hydatidose en algerie: resultat de deux enquetes retrospectives portant sur 10 ans». La tunisie medicale, 6: 318-326.

16. pandey m & chaudhari p - primary hydatic cyst of sacral spinal canal: case report. Neurosurgery, 1997, 40, 407-409 djaafer m et. al. / Vertebro-Medullaire Hydatidosis: About Three Cases Report And Review Of Literature

17. robert-gangneux f, tourte-schaefer c.valeur comparee de deux techniques de western-blot pour le diagnostic de confirmation d'une hydatidose. Parasitologie, 1998, manuscrit  $n^{\circ}1991$ .

18. seimenis, a. (2003). "overview of the epidemiological situation on echinococcosis in the mediterranean region". Acta tropica, 85: 191-195.

19. tekkouk ih & benlik - primary spinal extradural hydatid disease : report of a case with magnetic resonnance characterisis and pathological correlation. neurosurgery, 1993, 33, 320-323.

20. tsitouridis i & dimitriadis as - ct and mri in vertebral hydatid disease. eur radiol, 7, 1207-1210.

21. zlitni m, kooli m, ezzaouia k, lebib h & mestiri m - manifestations osseuses des parasitoses. emc appareil locomoteur, 1996, 14-021-b-10.