

EXTENSIVE VERTEBRAL HYDATIDOSIS A STUDY

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An exceptional case of vertebral hydatidosis with involvement of the cervical, dorsal, lumbar and sacral segments of the spine is presented. Vertebral hydatidosis is a rare form of hydatid disease. It is found in less than 2% of all cases in echinococcosis. The initial parasite localization is usually univertebral. Extensive primary lesions were related to massive infestation by tapeworm ova at multivertebral levels. At this stage, the disease presents as a malignant tumor resistant to all surgical procedures. The prognosis is poor because of severe neurological complications and inexorable spinal destruction. The hope remains that in the future an effective medical parasitocidal treatment will be available.

Keywords : hydatidosis ; vertebral.

Mots-clés : kyste hydatique ; localisation vertébrale.

Vertebral hydatidosis is rare, although in endemic areas it must always be considered in the differential diagnosis. The purpose of this paper is to describe the clinical and radiological findings, treatment and evolution in one case of extensive spinal lesions.

A literature review on this subject is presented.

CASE REPORT

A 40-year-old woman of rural origin presented in the pulmonary department in April 1985 with a 4-year history of progressive low thoracic back pain radiating along the seventh, eighth, and ninth ribs with a tender right paraspinal mass to the right of T8, T9, and T10. Lumbar puncture was performed, and clear fluid came back into the

aspirating syringe with extension of hydatid membranes. After that the patient developed a sinus tract in her back with intermittent discharge of hydatid cysts and debris. After 6 months the sinus tract healed, and the swelling disappeared.

The patient was readmitted to the National Orthopedic Institute in July 1986. She was found to have a stiff lumbar spine with tenderness. The scar was well healed without swelling. Nevertheless the abdominal examination showed a large mass, the volume of a baby's head in the left lower quadrant. There were no abnormal neurological signs.

Radiographs of the spine showed destructive changes in the bodies of C4, C5, C6, C7, all thoracic vertebrae and the first two lumbar ones with destruction of the inner part of the right seventh, eighth and ninth ribs and corresponding transverse processes. The neural arch was also affected by the destructive process with either enlarged hazy or even absent pedicles, especially in the dorsal segment in the antero-posterior view (fig. 1).

In association with these changes there were bilateral paravertebral shadows with smooth polycyclic limits extending from T6 to L2.

The most severe destruction involved the last seven thoracic and the first lumbar vertebrae with areolar lucencies and a niconcave aspect on the lateral view (fig. 2). Moreover the sixth, seventh

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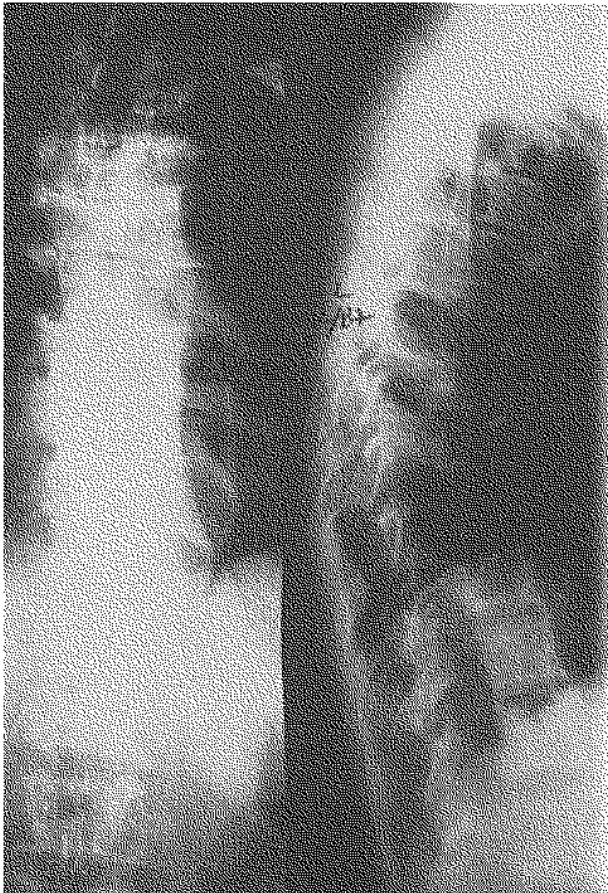


Fig. 1. — Osteolytic lesions involving all thoracic vertebrae. Left paravertebral shadow with polycyclic limits in the anteroposterior view.

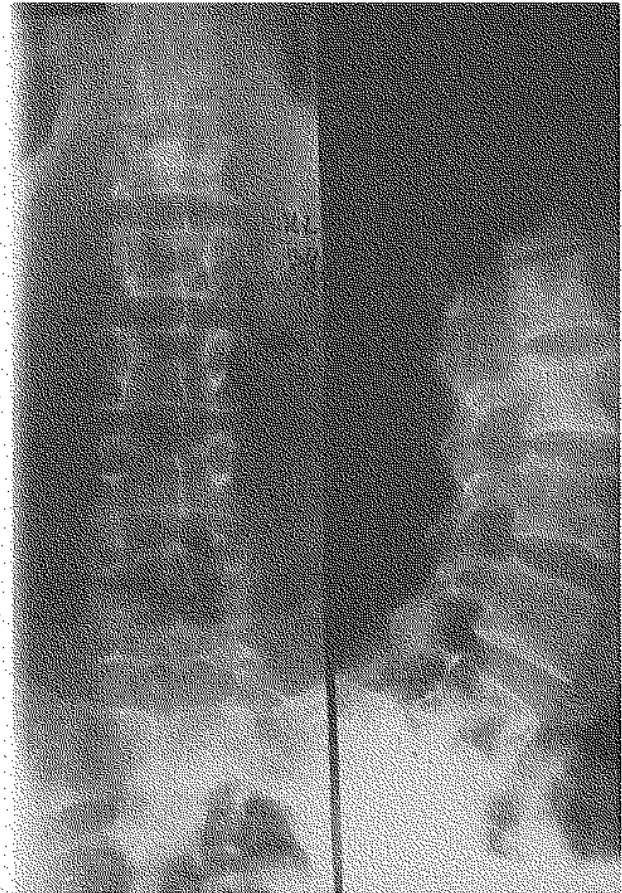


Fig. 2. — Osteolytic biconcave shape of the two first lumbar vertebrae with destruction of the corresponding pedicles and neural arches.

and eighth intervertebral disc spaces were narrowed with moderate collapse of T8. The patient was at full-term pregnancy. She left the hospital and returned one year after delivery in October, 1987 with a right lumbar paravertebral mass (10-cm diameter) and a lower abdominal mass of increasing size. The gait was spastic with hyperreflexia but without any motor strength or sensory neurological changes. The other main clinical finding was dorsal scoliosis convex to the right and a stiff flat lumbar spine. Radiological examination showed moderate progression of the bony destruction (fig. 3).

Computed tomography (CT) confirmed the involvement of the cervical, dorsal and lumbar vertebrae with an evident osteolytic aspect of the

left sacral wing not yet detectable on plain radiograms (fig. 4).

CT also revealed an extensive bilateral prevertebral cystic mass extending from the sixth cervical vertebra to the pelvis causing anterior displacement of the descending aorta with invasion of the spinal canal.

The left lumbar and intrapelvic cystic mass was multilocular, bilobed and compatible with the clinical findings. It extended into the spinal canal and behind the sacrum with involvement of the posterior part of the left ilium (fig. 5).

At operation in November 1987, through a left lumpectomy we drained a huge cystic collection containing 2 liters of degenerative hydatid debris. The cavity was filled with 10% formalin solution.

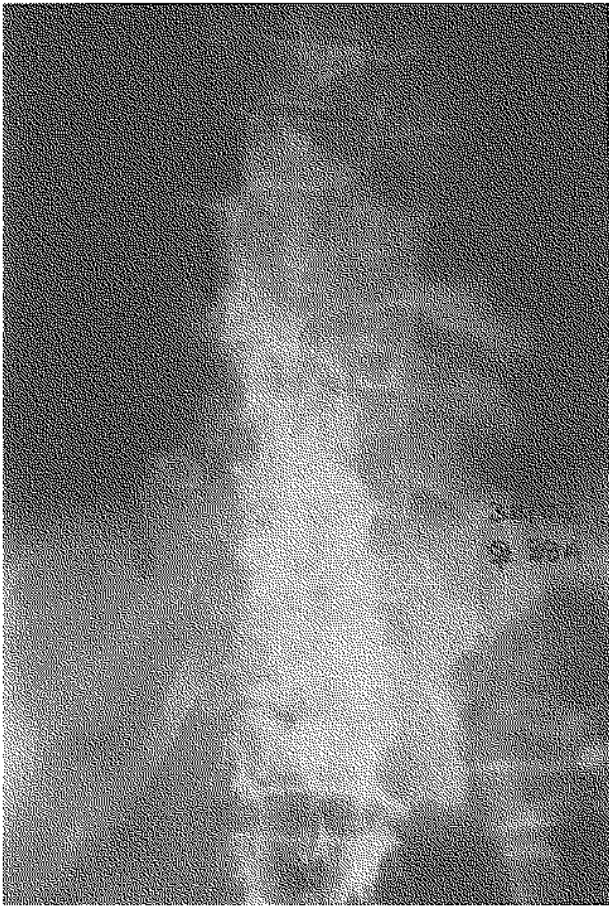


Fig. 3. — Progression of the destructive bipolar hydatid process in thoracic vertebrae and in the right costovertebral junctions.

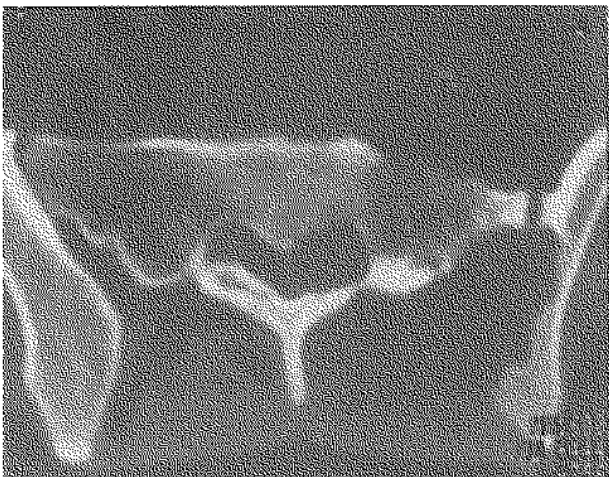


Fig. 5. — Destructive lesion of the left part of the sacrum resulting in involvement of the posterior segment of the ilium with a giant lateral perivertebral abscess.

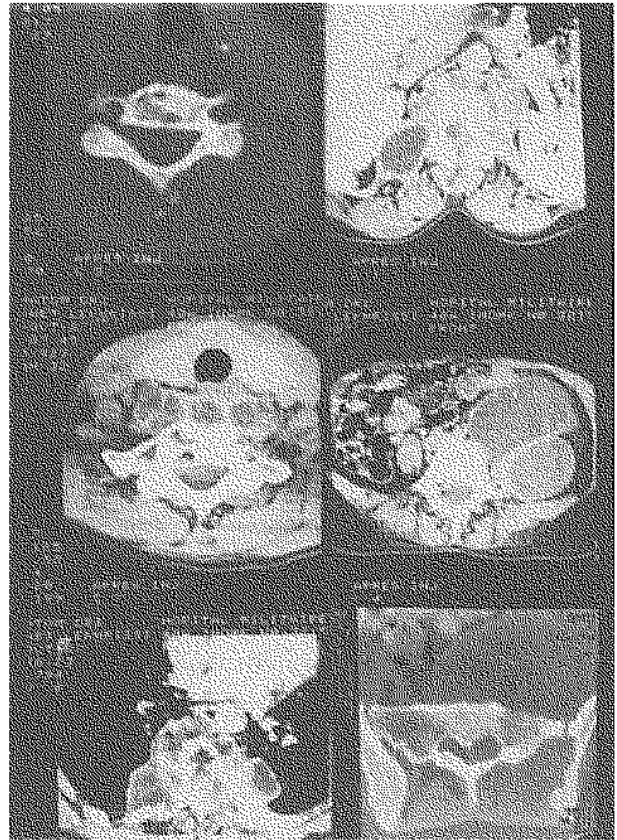


Fig. 4. — Extensive lesions involving cervical, dorsal, and lumbar segments and even the lateral left sacral process.

The local tissues were swabbed with the same product. The wound was closed in layers.

Recovery was rapid : muscle tone became normal and hyperreflexia disappeared. The patient left the hospital on December 15, 1987 with a corset. She was seen at follow-up in May 1988. On examination the dorsal scoliosis had increased, the abdomen was free of a palpable mass and the neurological status was normal.

Plain radiographs confirmed the worsening of the bony destructive process with a left lateral collapse of T8 resulting in the increase in scoliosis. The right ninth, tenth and eleventh ribs were completely destroyed (fig. 6). A repeat CT showed intraspinal invasion with a hydatid abscess surrounding the cord ; it showed extensive destruction of the vertebral bodies, the neural arches and transverse processes (fig. 7). It appeared clearly



Fig. 6. — Worsening of vertebral destruction leading to collapse of T8 and resulting in right thoracic scoliosis. Destruction of last two left ribs.

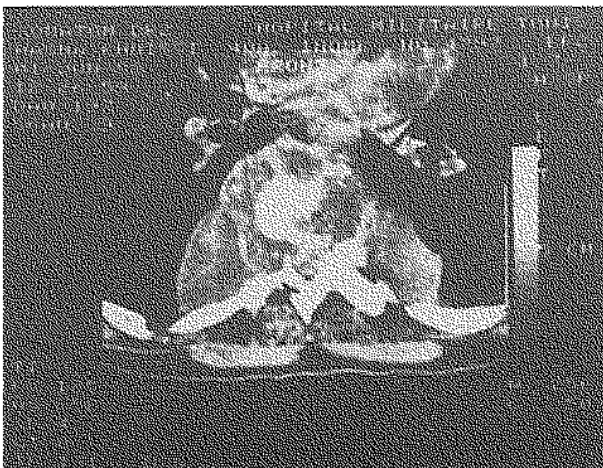


Fig. 7. — Extensive vertebral destruction involving the body as well as the neural arch with an intraspinal hydatid abscess.

that for such a malignant form of hydatid disease nonoperative treatment could be suggested for this patient. Medical treatment could be of some aid to improve the poor prognosis in this case.

Unfortunately after discharge from the hospital, the patient failed to return for follow-up examination.

DISCUSSION

In Tunisia, hydatid disease is caused by ingestion of the ova of the dog tapeworm, “*Echinococcus granulosus*”.

Man is an accidental host who replaces the usual one, the sheep. The intestation occurs after ingestion of food or water contaminated by the tapeworm ova excreted in dog feces.

The most usual localizations are liver and lungs, which are the first barriers to the larva during its bloodstream travel. Nevertheless, bone involvement by hydatid cysts is a rare condition. It is found in 0.37 to 2.4% of all cases of hydatidosis (1, 2, 13, 14, 15, 23, 28). The common site of bone lesions in hydatid disease is usually considered to be the spine. Most authors found this to be involved in 50% (4, 15, 25).

In bone tissue the larva behaves differently than in soft tissue. It grows in the direction of least resistance infiltrating and damaging the bone like a tumor by exogenous vesiculation. By relentless progression it may reach the dense cortical bone which will be eroded causing spontaneous pathologic fractures. In this way, the articular cartilage and periosteum are invaded and the neighboring soft tissues are involved. In this case large hydatid cysts were formed, enlarged by endogenous vesiculation resulting in the formation of daughter cysts. Extension may occur into the extradural space or into the paraspinal tissues.

The vertebral body, owing to its rich vascularization, is the common primary site of ova infestation. The intervertebral disc is spared and the shape of the body is maintained for a long time in spite of extensive destruction of bone.

The radiological features and differential diagnosis of vertebral hydatidosis have been described by many authors (4, 9, 12, 119, 24, 28). For the

diagnosis it is important to point out that there are no pathognomonic radiological signs.

Nevertheless, progressive vertebral destruction involving the body and posterior arch with multilocular osteolysis and a hazy image of the bone without periosteal reaction or processes of condensation are highly suggestive. Moreover when paravertebral lesions such as involvement of the ribs or round shadows in the soft tissues as found in our patient are present, the diagnosis of vertebral hydatidosis becomes highly probable, particularly if the patient comes from an endemic area.

Computed tomography has also proved to be an effective and sensitive method to define the limits of the destructive lesions in bone and to show the spread of the hydatid cysts in adjacent soft tissues with their specific water density (6, 7, 17).

Magnetic resonance has recently been added to the arsenal of the radiological procedures. It has proved its powerful ability as a noninvasive modality in demonstrating the multiloculated echinococcal spinal as well as paraspinal lesions and in evaluating the effect of the intraspinal extension of the disease (17, 20).

Besides the radiological data, sensitive immunological tests, especially immunoelectrophoresis, are of great diagnostic importance in hydatidosis (5, 16).

In fact, our reported case should not present any diagnostic difficulties because of the extensive destructive lesions: more than 18 vertebrae with extension to 5 adjacent ribs, the left part of the sacrum with the contiguous ilium.

To the best of our knowledge no similar example of such extensive spinal localization has been reported in the literature. Usually the lesion involves initially one vertebra and then extends to the adjacent segments. It takes a period of many years until the clinical manifestations, because of slow and insidious growth of the parasite.

At the onset of illness, the presenting symptoms are mainly neurological, due to spinal cord or radicular compression found in 25 to 100% according to various reports in the literature (5, 9, 10, 11, 16, 18, 23, 24, 25, 26). Other main symptoms are vertebral fractures and paraspinal masses caused by hydatid abscesses. This latter situation was found in our case.

At this stage there are usually no more than three or four affected vertebrae.

When the lesion was seen for the first time many vertebrae were involved; it is acceptable to explain the extension of the lesions by massive initial infestation and multivertebral independent localization of the parasite.

This suggestion is reinforced in our case by the presence of a left sacral lesion which is separated from the affected third lumbar vertebra by two normal vertebrae.

Nevertheless in this situation the passage of ova without any localization in the liver and lungs seems to be a strange phenomenon which could justify the exceptional findings in our case.

This very advanced stage of the disease has all the criteria of a locoregional malignancy. In such a situation all operative procedures are unable to cure the lesions. Symptomatic surgical drainage of large abscesses or transitory decompression by laminectomy in case of neurological complications are only palliative procedures (10, 11, 12, 22, 23, 26, 27). Relapses and progression of illness are not avoided even when radical removal of the lesions has been performed. This is generally related to a poor delimitation of the hydatid process in bone which justifies its description as a "white cancer" (9).

Medical treatment with either mebendazole or albendazole with recurrent drainage seems to give encouraging results (1, 3, 21, 24). This combination improves the gloomy prognosis in extensive hydatid lesions by slowing down the inexorable destructive progression of the hydatid disease.

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SAMENVATTING

S. KARRAY, M. ZLITNI, M. KARRAY, M. DOUIK, N. SLIMAN en T. LITAIEM. Extensieve hydatosis van de wervelkolom.

De auteurs rapporteren een uitzonderlijk geval van extensieve vertebrale hydatosis. Beschrijving van de klinische en radiologische tekenen van de aandoening. Bespreking van de etiopathogenie van de diffuse letsels. De heelkundige behandeling bereikt slechts een tijdelijk en gedeeltelijk resultaat; de prognose is even slecht als voor een maligne tumor.

Mogelijks zal in de toekomst de associatie van chirurgie met een medische behandeling de bothydatosis kunnen stabiliseren.

RÉSUMÉ

S. KARRAY, M. ZLITNI, M. KARRAY, M. DOUIK, N. SLIMAN et T. LITAIEM. Kyste hydatique vertébral extensif.

À propos d'un cas exceptionnel d'hydatidose vertébrale extensive, les auteurs ont étudié les caractéristiques cliniques et radiologiques de l'affection.

L'étiopathogénie des lésions diffuses est discutée.

Les auteurs démontrent que le traitement chirurgical n'est que palliatif et que le pronostic reste aussi mauvais que pour une tumeur maligne.

L'association de la chirurgie à un traitement médical pourrait avoir un effet curatif sur la maladie osseuse hydatique.