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Re: Vertebral hydatid cyst infection (*Echinococcus granulosus*): a case report

In their case report, Normelli et al. [8] from Sweden presented a complicated case of a 36-year-old male Iraqi Kurd with extensive vertebral hydatid cyst infection caused by *Echinococcus granulosus*.

Our country, Turkey, is an endemic area for hydatid disease [1, 2, 3, 9, 10, 11, 12]. Spinal hydatidosis is rare and most of the patients are between 30 and 60 years old [2, 12]. The disease preferentially involves the thoracic spine [10]. Following a course through the liver, the embryos reach the extradural spaces via Batson's venous plexus when intra-abdominal pressure increases. When the embryo reaches the vertebral body, it develops into its larval stage, known as the intraosseous stage of hydatid infection. Histologically, there are three layers of the cyst wall from the outermost to the innermost – pericyst, acellular laminated membrane, and germinal

layer – as stated by the authors [8]. The organism grows within the intratrabecular space and *endogenous* vesiculation results in the formation of daughter cysts within the parental cyst, destroying the bone like a tumor. The organism then spreads beneath the periosteum and *exogenous* vesiculation, known as the extraosseous stage of hydatid infection, and forms multiple cysts separate from the main cyst in the extradural and paraspinal areas, causing the spinal cord compression and paraspinal extension.

There are, however, several points that were not made clear by Normelli et al. [8], and we would like to draw the attention of the readers of *European Spine Journal* to a few that we consider to be important.

Location

Vertebral hydatid cyst infection has been classified according to the anatomical location of the hydatid infection into five subgroups: (1) intramedullary hydatid cyst; (2) intradural-extradural hydatid cyst; (3) extradural intraspinal hydatid cyst; (4) vertebral hydatid cyst; and (5) paravertebral hydatid cyst [5]. However, as the authors mentioned, there is no pathognomonic clinical picture of the disease [1]. It should be kept in mind as a rare cause of progressive myelopathy if the patient lives in the

major sheep-raising areas of the world or if the patient has migrated from a region where it is endemic, as occurred in Normelli et al.'s case.

Imaging

Characteristically, the articular cartilage and intervertebral disc are resistant to invasion in spite of collapse due to the eroded vertebra [4]. Preoperative diagnosis can be made using neuroimaging, but the bone involvement may not be evident on spinal radiographs. As stated by Altınörs et al. [2], computerized tomography and magnetic resonance imaging (MRI) have increased diagnostic specificity during the last decade. The imaging tool of choice is MRI, because it allows early diagnosis of spinal cord compression and more accurate localization of intraspinal and paraspinal echinococcal infestation, if coronal and axial MR images in addition to sagittal ones are obtained. Moreover, the alteration in signal intensity can be used to demonstrate cyst viability. On MRI, the cyst appears as a multiloculated hypointense mass on T1-weighted imaging and a bright hyperintense mass on T2-weighted imaging. T2- and proton-density-weighted images are useful in determining the cyst viability, with decreased signal on these sequences being a characteristic indicator of a dead cyst [11].

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Unfortunately, Normelli et al. [8] presented only a sagittal thoracolumbar MR image of their case, and they state in their case report that at surgery the infection was more extensive than was indicated by the radiological information obtained from MR images regarding the extension of the disease within the spinal canal. We do not agree with Dr. Normelli et al.'s opinion of the results of the MRI investigation as "false-negative". There is no doubt that more information can be obtained from T1- and T2-weighted coronal and axial MR images, especially in a patient who has been receiving pharmacotherapy for a long period. Therefore, full imaging of such a case would be very interesting.

Surgical treatment

We agree with the authors that "extensive" laminectomy may cause a local gibbus deformity, resulting in paraplegia. In such cases, it is possible to use anterior and/or lateral decompression, but only with major tissue trauma. Alternatively, Açıkgöz et al. [1] have described an endoscopic surgical technique, a kind of minimal invasive procedure, for the removal of spinal lesions in an interesting case with multiple vertebral hydatid cysts.

Value of albendazole

Vertebral hydatid cyst infection has typically an invasive nature, and the

removal of all the cysts cannot be achieved without perforating the cyst wall in the majority of cases [3, 5, 9, 12]. Recently, successful treatment of hydatid disease with albendazole (ALB) for at least 1 year has been reported [3, 6, 7]. We do not agree with the authors' claim that ALB is a parasitostatic drug rather than parasiticidal, and that the disease is controlled rather than cured. Although there are very limited series from which to draw any conclusions regarding the value of the drug, the progressive shrinkage of the large viable spinal hydatid cysts and the cure of the small ones prove the efficacy of ALB in these cases. Based on recent experience in the management of hydatid disease, we think that ALB should be used as a "systemic or topical" scolical agent in the presence of cyst rupture or systemic disease, and recurrent cases in the intra- and postoperative period, as well as in cases of inoperable spinal hydatid disease. ALB treatment results in immobilization and death of *Echinococcus granulosus* as a result of low adenosine triphosphate (ATP) production, by preventing glucose uptake and glycogen storage.

In conclusion, Normelli et al. [8] apparently have some useful information on this serious condition, but they do not convey it in their article. For example, they do not offer any conclusions based on their patient in the Discussion section of their case report.

We invite Dr. Normelli, the lead author, on the behalf of his co-authors, to review these other reports cited above and welcome his feedback.

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