

Endless story of a spinal column hydatid cyst disease: A case report

Gokhan Cavus*, Vedat Acik, Emre Bilgin, Yurdal Gezeran, Ali Ihsan Okten

Department of Neurosurgery, Adana Numune Training and Research Hospital, Adana, Turkey

ARTICLE INFO

Article history:

Received 11 September 2017

Received in revised form

24 February 2018

Accepted 19 March 2018

Available online 7 May 2018

Keywords:

Hydatid cyst

Surgical treatment

Spinal hydatid cyst

Corpectomy

ABSTRACT

We describe a case of multifocal relapsing hydatid cyst following multilevel thoracic corpectomy and 360° instrumentation surgery. A 41-year-old male patient presented with cord compression and paraplegia due to a multiseptated cystic lesion at T10-T11 level. The cyst was excised with a combined anterior and posterior approach and 360° stabilization was performed. The patient received albendazole for 1 year after the surgery.

The patient presented with paraparesis 5 years after the surgery. Cystic lesions between C2-T1 and T10-T11 were detected on the spinal MRI and the patient was operated with removal of the lesions on both levels and adjuvant local 20% hypertonic saline application. The patient received albendazole for the postoperative 6 months.

After 3 months from the surgery, the patient's paraparesis recovered. There was no recurrence after 2 years from the last surgery.

© 2018 Turkish Association of Orthopaedics and Traumatology. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Background

Hydatid cyst is a disease caused by the larvae of the *Echinococcus granulosus* cestode and is commonly seen in the Mediterranean region, Middle East and India.¹ Diagnosis of this disease is usually late as its general course is painless.² The adult form of the cestode lives in dog intestines, which are the definitive host. Infection in humans occurs as a result of eating tapeworm eggs.³ 60–70% of hydatid cysts are formed in the liver while 10–15% involve the lungs.^{4,5} Bone involvement is rare and observed only in 0.5–2% of all hydatid cyst cases, with approximately half of them residing in the vertebrae.⁶ Thoracic spine is the most affected region.⁷

In our case, a patient with thoracic multilevel extradural, intraspinal, vertebral and paravertebral hydatid cyst was treated with two-stage combined anterior and posterior decompression.

The hydatid cyst multifocally had relapsed in the cervical and lumbar regions of the spine during the two-year follow-up period and decompression surgery was repeated.

Patients and methods

A 41-year-old male patient presented to our clinic with complaints of lower back pain for a year, paraplegia which developed 10 days ago and a urinary and fecal incontinence. On the MRI, a multiseptated lesion extending from the right paravertebral region to the midline at the T10-T11 level and to the intervertebral disc space, which resulted in height loss at the anterior and transverse colon level and an increase in the thoracic kyphosis, was observed. Hyperintense cystic components in sagittal cross-sections of T2-weighted images and hypointense cystic components in axial cross-sections of T1-weighted images, with a heterogeneous inner structure, were also observed (Fig. 1). No obvious contrast agent retention was observed in T1-weighted sagittal and axial cross-sections of the thoracic MRI (Fig. 2).

The thoracic and lumbar region tomography at the T10-T11 level revealed narrowing of the intervertebral disc space, height loss in the anterior body of the vertebrae, kyphotic angle formation at this level, and irregularities and destruction in the end plates. A

* Corresponding author. Department of Neurosurgery, Adana Numune Training and Research Hospital, Serinevler Mah., Ege Bagatur Bulvarı, Yuregir, Adana, 01260, Turkey. Tel.: +90 342 338 1731; fax: +90 342 338 8618.

E-mail address: dr.gokhancavus@gmail.com (G. Cavus).

Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology.

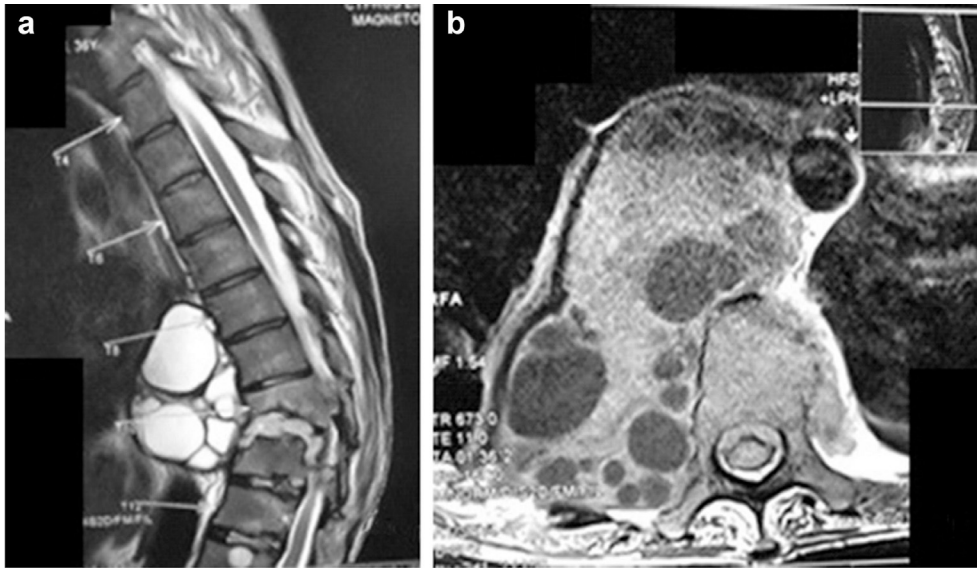


Fig. 1. Preoperative (a) T2-weighted sagittal and (b) T1-weighted axial cross-section MRIs.

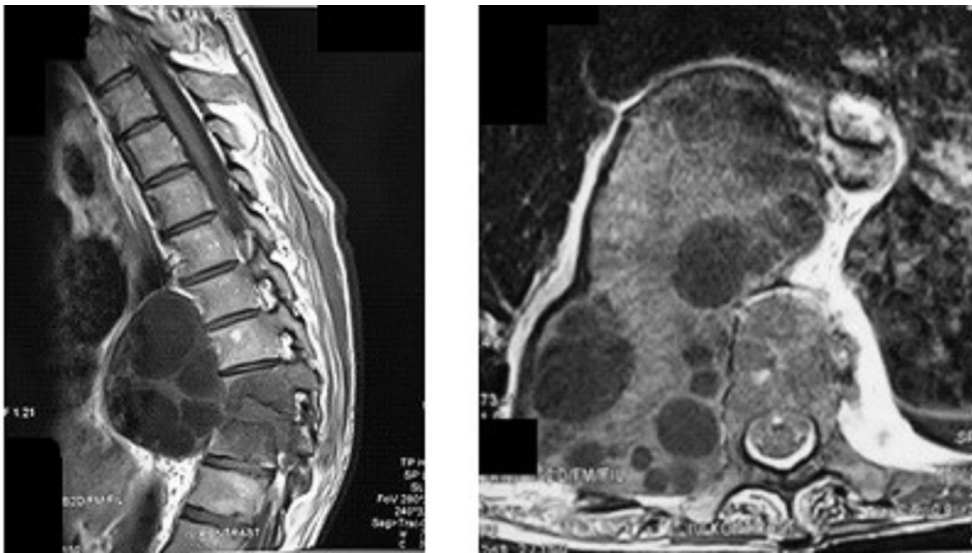


Fig. 2. Preoperative T1-weighted contrast sagittal and axial cross-section MRIs of the thorax.

heterogeneous-looking lesion containing hypodense cystic regions was also detected within the right anterior paravertebral region (Fig. 3).

In 2010, the cyst at the T10-T11 level was excised via combined anterior and posterior surgery in the same session (Fig. 4) and the surgical region was washed off using 20% hypertonic saline. Following corpectomy, a cage was placed at the T10-T11 vertebrae, and a posterior instrumentation and fixation with pedicle screws was applied at the T9-T12 vertebrae. Instrumentation and fixation with pedicle screws were also performed on the T6-8 and L1-2 vertebrae from the posterior to correct the deformity and to ensure stabilization (Fig. 5). The patient was administered albendazole for the postoperative one year.

After 18 months of long-term physiotherapy and rehabilitation, the patient's paraplegia almost fully recovered.

A full spinal MRI performed after the patient redeveloped paraparesis at the end of the five-year follow-up period revealed a cystic characterized mass lesion filling the anterior epidural space between the C2-T1 vertebrae and compressing the spinal cord (Fig. 6). On the thoracic MRI, a lesion clearly compressing the cord from the T10-T11 posterior epidural region and extending bilaterally to the paravertebral region, exhibiting clinical features of paraparesis, was also detected (Figs. 7 and 8).

The patient underwent surgery and the cystic lesion was completely removed without any cystic rupture via hemilaminectomy to the C2-C4 vertebrae from the left side and the surgical region was washed off using 20% hypertonic saline during



Fig. 3. Preoperative CT images of the thoracic and lumbar regions.

the same session. Then the cystic lesion at the T10-T11 level was excised by entering through the previous incision point in the thoracic region and the surgical region was washed off with 20% hypertonic saline. The patient received albendazole for the post-operative six months.

Results

Three months after the second surgery, the patient's paraparesis recovered. No recurrent lesion was observed in the CT and MRIs taken at the second year follow-up (Figs. 9–11).

Discussion

Hydatid disease mostly forms in the liver (60–70%) and in the lungs (10–15%).⁵ Bone involvement is rare and is observed only in 0.5–2% of all cases, with approximately half of the cysts involving the vertebrae.⁶ Hydatid cyst has been reported to have developed in 25–84% of cases with a neurological deficit.⁸ Abbassioun and Amirjamshidi⁹ reported that thoracic vertebrae were involved in 60%, lumbosacral vertebrae in 35% and cervical vertebrae in 5% of the cases. In 2005, Herrera et al¹⁰ accounted the involvement of chest and lumbar vertebrae in 20 cases. In our study, we present a rare case showing synchronous multifocal thoracic and cervical involvement.



Fig. 4. Excised hydatid cyst.

The parasite spreads to the paraspinal tissues or epidural space by perforating the cortex and the anterior part of the paravertebral body.⁹ The cyst may extend from the vertebral body to the anterior space, laterally to the extradural space or paraspinal tissues, or even to the spinal canal, compressing the spinal cord. However, involvement of multiple vertebrae is rare. Intervertebral discs constitute a strong barrier against the diffusion of hydatid cyst. The cysts in the spinal canal are mostly located at the posterior or posterolateral aspect of the spinal cord.⁹

Braithwaite and Lees⁶ classified the spinal disease in five types: 1) primary intramedullary hydatid cyst, 2) intradural extramedullary hydatid cyst, 3) extradural intraspinal hydatid cyst, 4) spinal hydatid cyst disease of the spine and 5) paravertebral hydatid cyst disease. In our case, the hydatid cyst was located in the vertebral and paravertebral regions of the thorax and extradural intraspinal regions of the cervical spine.

Clinical manifestation of the hydatid cyst depends on the level of the affected vertebrae and the stage of the disease. The disease shows no pathognomonic signs or symptoms other than those related to spinal cord compression.¹¹ When the bone is fractured, a neurological deficit occurs together with a continuous pain.^{12,13} Patients usually present with continuous back pain and radicular pain. Weakness in the extremities occurs afterwards causing paraplegia in 25–84% of the cases. The symptoms in our patient, diagnosed one year after the onset, were back pain, paraplegia and urinary and fecal incontinence.

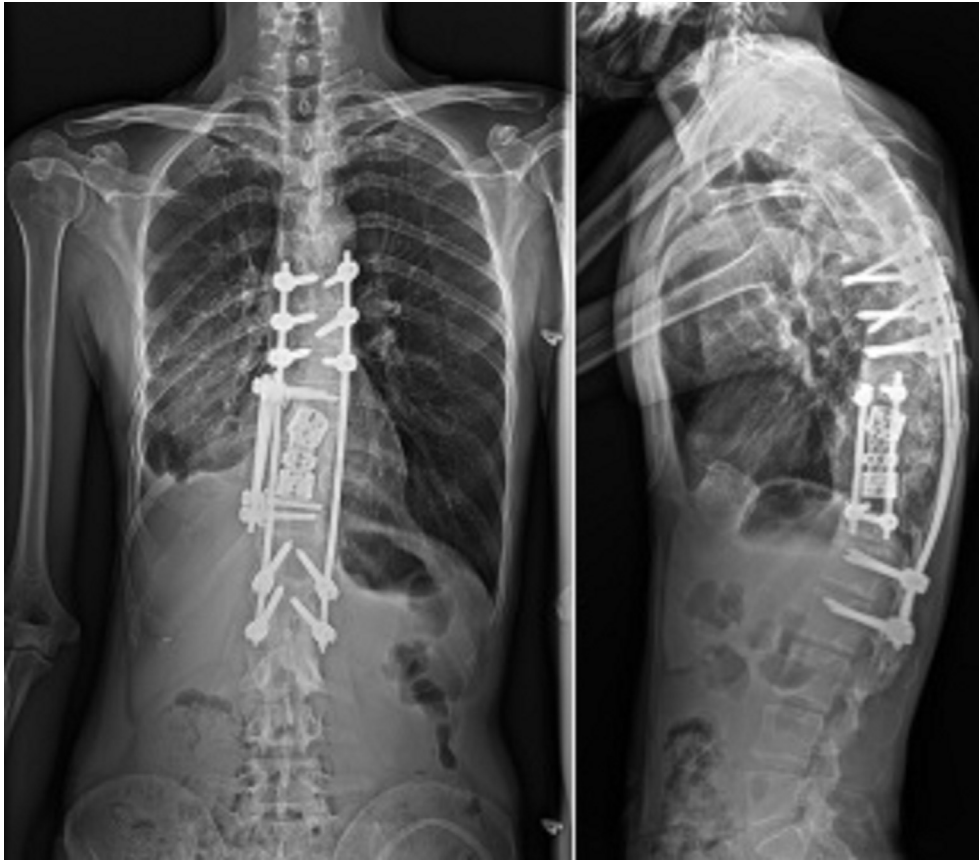


Fig. 5. Anteroposterior and lateral radiographs after the first surgery.

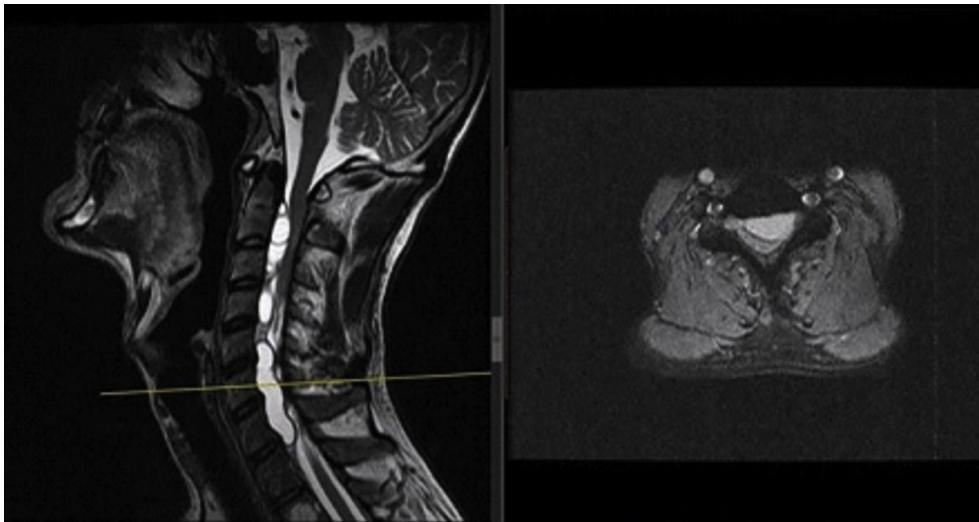


Fig. 6. T2-weighted sagittal and axial cross-section MRIs of the cervical spine taken five years after the first but prior to the second surgery.

The adjacent bones are involved in the late stages of the disease. CT and MRI are deemed compulsory for the diagnosis. CT is particularly beneficial for screening the damaged vertebral structures^{14,15} and MRI is considered the best imaging tool. A hydatid cyst mostly contains a single thin wall and the contents have the same density as the cerebrospinal fluid

(CSF).^{16,17} In T2-weighted imaging, a low-density content appears in the cyst highly signaling in a homogeneous manner on the cyst wall. The density of the cyst fluid is the same as the CSF in T1- and T2-weighted imaging. The lesion shows no gadolinium uptake.¹⁵ Our CT and MRI results conformed to those in the literature.

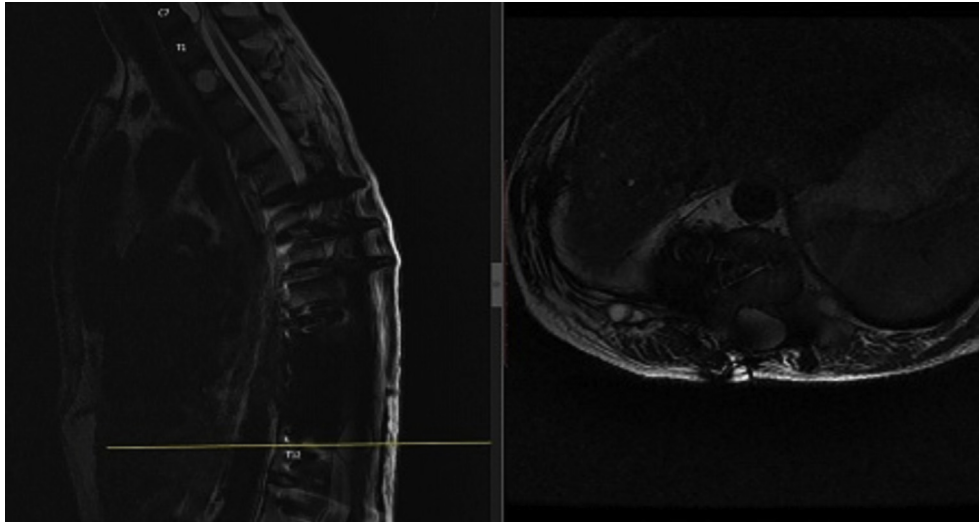


Fig. 7. T2-weighted sagittal and axial cross-section MRIs of the thorax taken five years after the first but prior to the second surgery.

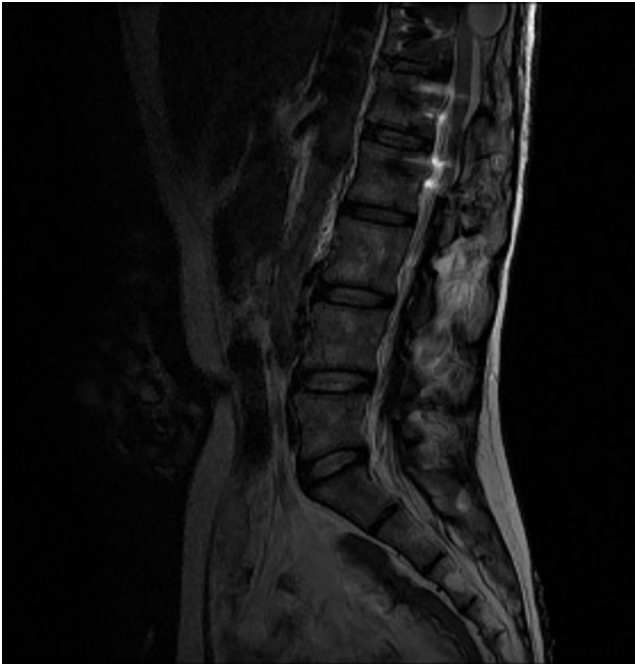


Fig. 8. T2-weighted sagittal cross-section MRI of the lumbar region taken five years after the first but prior to the second surgery.

The presence of the visceral hydatid cyst should alert the physician against the spinal hydatid cyst disease. Biopsy or aspiration of the cyst has the risk of anaphylaxis and diffusion, and it should never be considered.¹⁸ If possible, the lesion should be fully excised as a block, and if required a fixation should be performed. The cystic space and the surrounding surgical region should be washed off using hypertonic saline.¹⁹ Laminectomy is widely used in the treatment of spinal hydatid cysts. The purpose of the surgery must be to excise any cyst without causing any rupture.⁷ However, this is almost impossible to do due to radical excision, indefinite anatomical boundaries and existing neural

structures. The preferred treatment method should be early surgical decompression, radical resection and stabilization of the surgical region, washing off the surgical region using various solutions and postoperative adjuvant chemotherapy. Although the efficacy of certain solutions are not proved yet, they are widely used in the reported studies and include: (3%, 10%, 20%) hypertonic saline, 0.5% betadine, 0.5% silver nitrate and 2% formalin.⁹ The unfavorable results may be associated with the localization of the cysts (intradural and extramedullary) and poor penetration of anthelmintics (albendazole or mebendazole) and its metabolites into the intradural space via a passive diffusion transferring mechanism.²⁰ The use of albendazole has been recommended to be limited to six months to one year in order to prevent any redevelopment of the cyst. Our patient received albendazole for a year after both surgeries.

Any cyst would probably burst during the surgery and its relapse rate will exceed 40%.²¹ Turtas et al reported a relapse rate of 50% following posterior decompression surgery.²² In spinal cyst hydatidosis, recovery through excision is possible in case of a single cyst with a solid cyst wall.⁹ In our case, the hydatid cyst multifocally relapsed although a wide resection and anterior and posterior fusion was performed on the affected vertebral body.

In conclusion, the spinal hydatid cyst is a severe disease with significant morbidity. The disease may not be fully resolved via surgical or medical treatment. Medical treatment is recommended by almost all authors although it is not absolutely effective in preventing the relapse. Albendazole treatment alone may be ineffective on the recurrent spinal hydatid cyst. The recommended treatment is vertebrectomy with anterior or posterior approach and spinal cord decompression via instrumentation in suitable cases, and subsequent adjuvant medical therapy. The results are rarely satisfactory and the prognosis is usually poor.

Conflicts of interest

No funds were received in support of this work. The authors have no conflicts of interest. No clinical presentation at a conference.

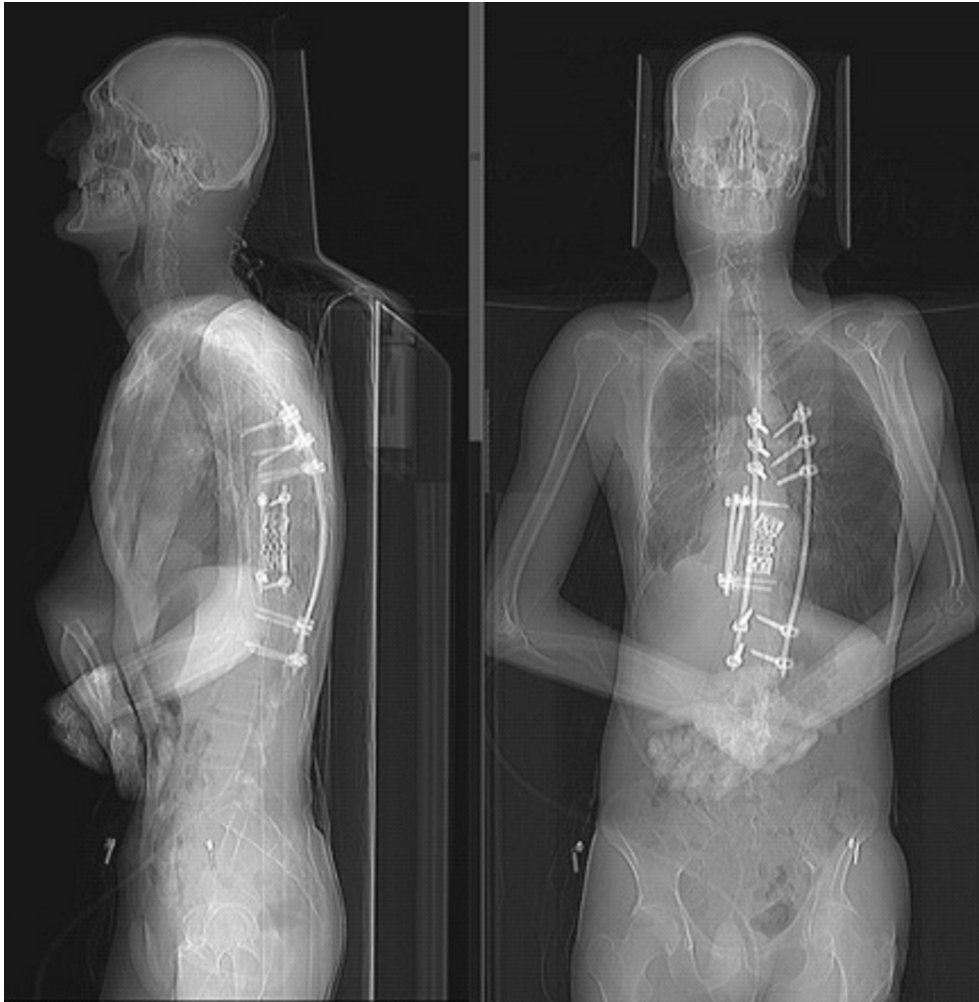


Fig. 9. Lateral and AP CT scan of the cervical spine at the second year follow-up after the second surgery.

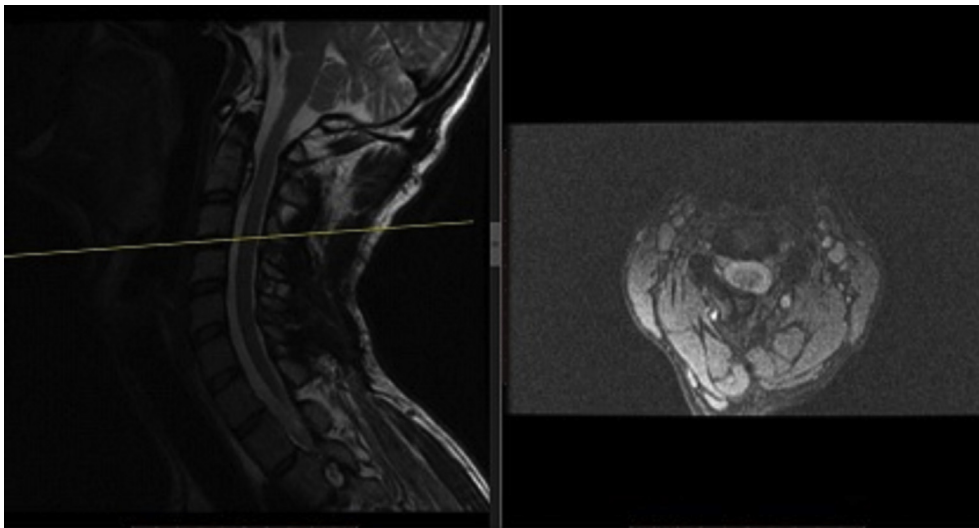


Fig. 10. T2-weighted sagittal and axial cross-section MRIs of the cervical spine taken at the second year follow-up after the second surgery.

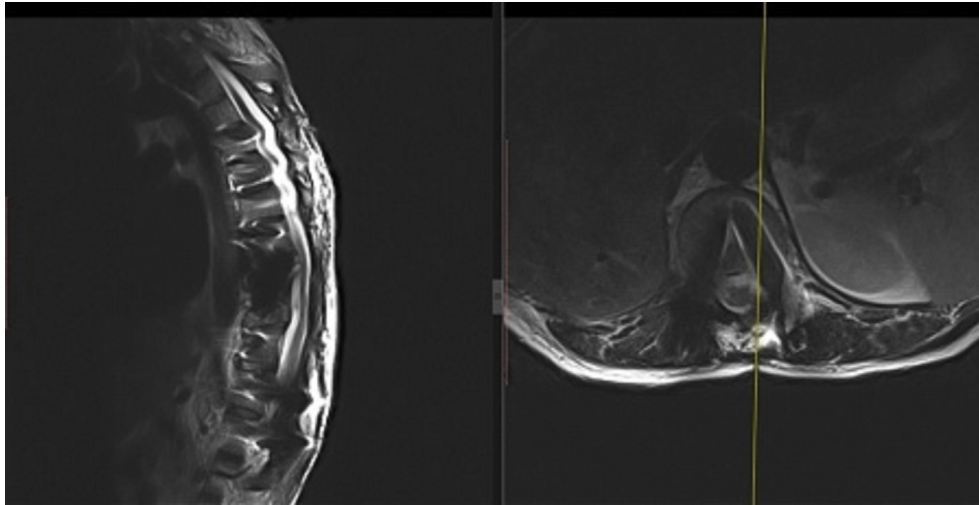


Fig. 11. T2-weighted sagittal and axial cross-section MRIs of the thorax taken at the second year follow-up after the second surgery.

References

- Charles RW, Govender S, Naidoo KS. Echinococcal infection of the spine with neural involvement. *Spine*. 1998;13:47–49.
- Papanikolaou A. Osseous hydatid disease. *Trans R Soc Trop Med Hyg*. 2008;102:233–238.
- Romig T, Dinkel A, Mackenstedt U. The present situation of echinococcosis in Europe. *Parasitol Int*. 2006;55:S187–S191.
- Scarlata F, Giordano S, Saporito L, et al. Cystic hydatidosis: a rare case of spine localization. *Infez Med*. 2011;19:39–41.
- Schnepper GD, Johnson WD. Recurrent spinal hydatidosis in North America. Case report and review of the literature. *Neurosurg Focus*. 2004;17:E8.
- Braithwaite PA, Lees RF. Vertebral hydatid disease: radiological assessment. *Radiology*. 1981;140:763–766.
- İşlekel S, Zileli M, Erşahin Y. Intradural spinal hydatid cysts. *Eur Spine J*. 1998;7:162–164.
- Morshed AA. Hydatid disease of the spine. *Neurochirurgia (Stuttg)*. 1977;20:211–215.
- Abbassioun K, Amirjamshidi A. Diagnosis and management of hydatid cyst of the central nervous system: Part 2: hydatid cysts of the skull, orbit, and spine. *Neurosurg Q*. 2001;11:10–16.
- Herrera A, Martínez AA, Rodríguez J. Spinal hydatidosis. *Spine*. 2005;30:2439–2444.
- Pamir MN, Akalan N, Ozgen T, Erbenli A. Spinal hydatid cysts. *Surg Neurol*. 1984;21:53–57.
- Ozdemir HM, Oğün TC, Tasbas B. A lasting solution is hard to achieve in primary hydatid disease of the spine: long-term results and an overview. *Spine*. 2004;29:932–937.
- Bonifacino R, Dogliani E, Craig PS. Albendazole treatment and serological follow-up in hydatid disease of bone. *Int Orthop*. 1997;21:127–132.
- Hamdan TA, Al-Kaisy MA. Dumbbell hydatid cyst of the spine: case report and review of the literature. *Spine*. 2000;25:1296–1299.
- Kovoor JM, Thomas RD, Chandrashekar HS, Jayakumar PN, Pillai S, Shankar SK. Neurohydatidosis. *Australas Radiol*. 2007;51:406–411.
- Kalkan E, Cengiz SL, Çiçek O, Erdi F, Baysefer A. Primary spinal intradural extramedullary hydatid cyst in a child. *J Spinal Cord Med*. 2007;30:297–300.
- Coates R, von Sinner W, Rahm B. MR imaging of an intracerebral hydatid cyst. *AJNR Am J Neuroradiol*. 1990;11:1249–1250.
- Zlitni M, Ezzaouia K, Lebib H, Karray M, Kooli M, Mestiri M. Hydatid cyst of bone: diagnosis and treatment. *World J Surg*. 2001;25:75–82.
- Salduz A, Koyuncu LO, Dikici F, Talu U. Long-term result of treatment of paraspinal and extradural hydatid cyst: a case report. *Acta Orthop Traumatol Turc*. 2009;43:267–271.
- Takayanagui OM, Bonato PS, Dreossi SA, Lanchote VL. Enantioselective distribution of albendazole metabolites in cerebrospinal fluid of patients with neurocysticercosis. *Br J Clin Pharmacol*. 2002;54:125–130.
- Arif SH, Zaheer S. Intradural extramedullary primary hydatid cyst of the spine in a child: a very rare presentation. *Eur Spine J*. 2009;18:179–182.
- Turtas S, Viale ES, Pau A. Long-term results of surgery for hydatid disease of the spine. *Surg Neurol*. 1980;13:468–470.