

Epidural Spinal Cord Compression by an Abscess from a Dumbbell Extension of a Non-Osseous Soft Tissue Paravertebral Hydatid Cyst in the Dorsal Region

Saleem Khadir Musalah*

Muhammad Azeez Sulaiman**

Anas Amer Mohammad***

Walid Wahab Al-Rawi****

Abstract

Background and objectives: Human being may become infested with *Echinococcus granulosus*, as an intermediate host, ending with the formation of hydatid cyst. Hydatidosis, affecting various parts of human body and should be considered in the differential diagnosis of any cystic mass lesion. Osseous Hydatid cyst is rare; however, it involves the spinal column in half of the cases. In the current study, the epidural hydatid cyst had presented with spinal cord compression due to the formation of an epidural abscess formation.

Methods: A young adult housewife lady who had been affected by the parasite, primary paravertebral hydatid cyst, between the seventh and ninth dorsal vertebral levels, presented with spinal cord compression due to epidural abscess lesion. Ultrasound, the contrast magnetic resonance, and computed tomography imaging are standard tests in the diagnostic work-up of the lesion and follow-up of the patient. Appropriate dorsal laminectomy, removal of the hydatid cyst, and the inflammatory epidural cystic compressing tissues were done.

Results: Operative intervention had resulted in a dramatic improvement in the patient's condition and reversal of her neurological deficit back to normal; the paravertebral ectocyst size had eventually shrunken and there was no recurrence. The excised tissues that were compressing the dorsal spinal cord proved, Histopathologically, to be inflammatory in its microscopical appearance.

Conclusions: Hydatid cyst epidural dumbbell extension may rupture and lead to the formation of an abscess, giving enhancement on contrast magnetic resonance and computed tomography scan studies. To our knowledge, this is the first case to be reported in the literature specifying spinal cord compression by an epidural abscess formation from a paraspinous dumbbell extension of hydatid cyst.

Key words: Abscess; Albendazole; *Echinococcus granulosus*; Epidural Hydatid cyst; Spinal Cord Compression.

Introduction

Most researchers mention that *Echinococcus granulosus* hydatidosis is endemic in the Eastern and Southern countries of Mediterranean sheep breeding areas, and west China. Body organs

infected with the hydatid cyst (HC) include, in descending order, the liver, lungs, spleen, kidney, heart, bone, and the central nervous system. Osseous HC affects 2% of those with hydatidosis and

* M.B., Ch.B., C.A.B.R. (Radiodiagnosis) Lecturer,

** M.B., Ch.B., F.I.C.M.S. (Neurology) Assistant Prof.,

*** M.B., Ch.B., F.I.C.M.S. (Anaesthesia and Intensive Care) Assistant Prof.,

**** M.B., Ch.B., F.R.C.S. (G) (Neurosurgery), Professor

** Department of Medicine, College of Medicine, University of Duhok, Region of Kurdistan, Iraq. *, ***, and

**** Department of Surgery, College of Medicine, University of Duhok, Region of Kurdistan, Iraq.

Correspondence: Prof. Dr. Walid Wahaib Al-Rawi, E-mail: wahidwahaib4960@gmail.com

that half of the cases affect the vertebral column, preferably the dorsal region. Hydatid cyst, as a surgical problem, should be thought of in our current clinical practice, in geographical regions endemic with this cestode worm infestation, Hydatidosis lesion may involve vertebral body and extends to epidural space producing spinal cord compression with its resultant general (e.g., backache) and neurological symptoms and signs (radicular pain, sensory level, motor deficit, and autonomic sequelae (e.g., pelvic sphincteric functional derangement)). They, furthermore, recommend the use of combined decompressive /reconstructive surgery and Albendazole anthelmintic therapy as therapeutic and curative measures.

Patients and method

A 33-year-old housewife, displaced refugee camp inhabitant, in the north of Iraq, from a non-domestic-animal-raising family, had presented to our private neurosurgical practice with 2-month-history of mid-backache, radiating to the buttocks and back of both lower limbs during most of the day, neither relevant to weight lifting nor to movement. She also had experienced progressively bilateral leg weakness, paraesthesia from the umbilicus to the feet, and urinary dribbling; however, the bowel motion was unaffected. She felt low grade fever during the course of the disease that responded to self-ingestion analgesia. Body weight loss was negligible. She gave history that her elder sister was operated upon 15 years ago for HC of the liver. On examination, she was fully conscious, mobile with help, and cooperative but looked suffering and in marked discomfort. Peripherally, she was acyanotic and well perfused. There was no neck stiffness. The vital signs were

However, although recurrence is possible, most authors mention "dramatic" improvement and / or steadiness in the clinical course of the disease¹. In the current report, in a young female lady patient, the epidural hydatid cyst component of a dorsal paravertebral dumbbell extension of the hydatid cyst has become infected, either primarily, or due to its rupture and the release of irritant materials, leading to an intense inflammatory process and an abscess formation, clinically, producing spinal cord compression. To our knowledge, this is the first case to be reported in the literature specifying spinal cord compression by an epidural abscess formation from a paraspinal dumbbell extension of hydatid cyst.

normal. There was marked tenderness on palpation over the mid-dorsal region; there was some restriction of trunkal flexion and extension. The cranial nerves were intact. However, examination of the peripheral nervous system had revealed sensory level of hypoaesthesia and hypoalgesia to about the umbilicus (T9/10 level), brisk knee and ankle jerks, and up-going plantar responses bilaterally; both of the vibration joint position senses were diminished. Muscle power of the legs, hypertonic paraparesis, was 3-4/5 (Medical Research Council grading system). Routine blood tests were as follow: Hb 11.7 gm/dL, mild neutrophilia and eosinophilia, ESR 67 mm/hour, and highly sensitive C-reactive protein 5.3 mg/100 ml. Fasting blood sugar, renal functions, and serum lipids were normal; Zinc 11.7 mmol/L (normal 13.8-18.6 mmol/L), and serum 25 OH vitamin D-ELISA 11.4 ng/mL (30-100 ng/mL). Immunological tests for the HC were unavailable.

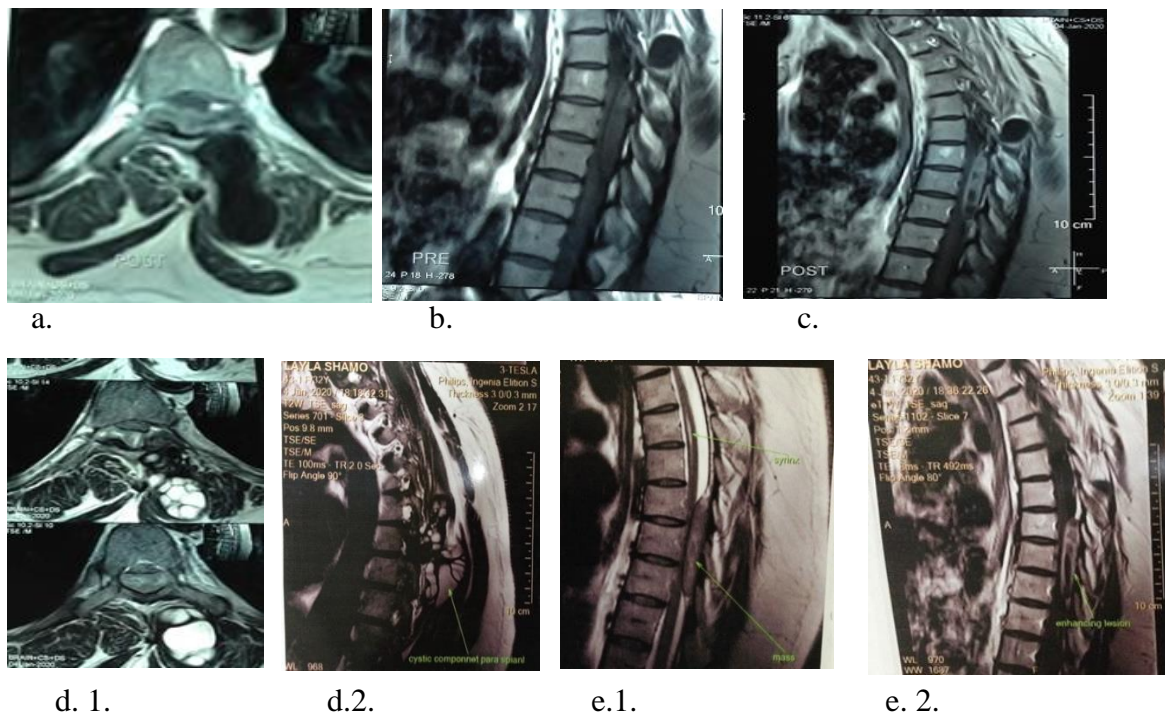


Figure (1): Pre-operative MR images, pre-and post-contrast T1W and T2W phases (a-e).

(a.) T1W precontrast, axial reconstruction; it shows the dumbbell extension.

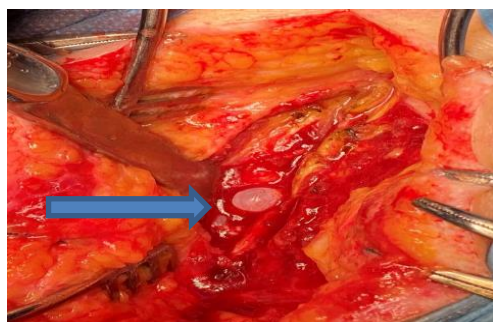
(b.) T1W precontrast, sagittal reconstruction.

(c.) T1W postcontrast, sagittal reconstruction. It clearly shows the dumbbell extension and the enhancing epidural inflammatory cystic mass.

(d. 1&2.) T2W pre-contrast, axial and sagittal reconstruction. It shows the left sided paraspinal multicystic mass and its dumbbell extension into the spinal canal through the intervertebral neural foramen.

(e. 1&2.) T2W pre- and post-contrast sagittal reconstruction. It shows a syrinx proximal to the enhancing epidural cystic inflammatory mass (abscess). Imaging studies were as follow: Abdominal U/S and chest X-ray and computed tomography (CT) scan were normal. Pre-operative contrast magnetic resonance (MR) scan had shown, at T7-T9 levels, left sided paravertebral paraspinal non-enhancing

multicystic mass, 78X73X36 mm dimensions, hypointense on T1W and hyperintense on T2W phases, dumbbell extension through intervertebral foramen to the inside of the spinal canal at T8 level communicating with the contrast-enhancing-multicystic and thickened epidural tissue, 58X11X25 mm dimensions, markedly compressing the spinal cord (Figure1, a-e).



(a.1.)



(b.2.)

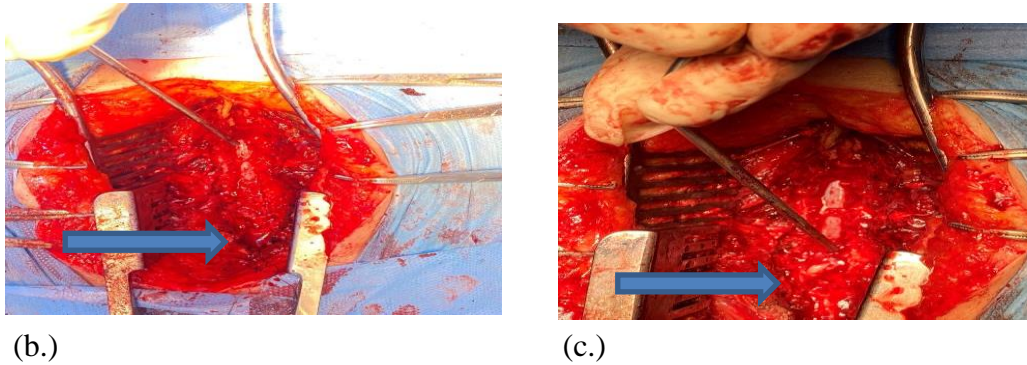


Figure (2): Operative findings.

(a.1.) The accidental rupture of the mother cyst in the left paraspinal muscular tissues and the extrusion of one of the daughter cysts; see arrow.

(a.2.) The laminated membrane of the mother cyst with one of the daughter cysts; see arrow.

(b.) The thickened inflammatory tissues are seen over the lower part of the picture over the dura; see arrow.

(c.) Purulent fluid (pus) coming out during dissection and removal of the inflammatory tissue mass seen in picture; see arrow.

Under general anaesthesia, intravenous dexamethasone and ceftriaxone, in the prone position, the patient was operated upon through T6-T10 laminectomy. Although unfortunately, the main HC had ruptured during the dissection of the paraspinal muscles, however, there was no anaphylaxis. Grossly, we could achieve total removal of the main HC laminated membrane, daughter cysts, and the epidural thickened inflammatory tissue and sucking out all of the purulent collection inside (FIG 2, a-d). The dura was not violated and remained intact along the operative field. No instrumental vertebral stabilization was done. The excised tissues that were compressing the dorsal spinal cord proved, Histopathologically, to be inflammatory in its microscopical appearance. The purulent fluid that was drained during the decompressing procedure, "pus", was sterile. Post-operative recovery was uneventful; she

was given two-monthly courses of Albendazole, 400 mg three times daily, in addition to nutritional supplements (see above). During follow-up visits, the patient regained her full neurological functions. Post-operative contrast MR scan had confirmed the shrinkage of the paraspinal main ectocystic mass, removal of the inflamed epidural tissues, and freeing of spinal cord from any compression (FIG 3, a-b); however, post-operative CT scan had shown the absence of bony involvement with HC lesion, removal of T6-T10 vertebral laminae, and the normal alignment of the thoracic vertebrae (FIG 4, a-c). The research complies with the guidelines and regulations of Clinical Ethical Committees of both, The Ministry of Health, Region of Kurdistan and College of Medicine, University of Duhok, Region of Kurdistan, Iraq. Consent for approval has been taken from the patient, and her identity has been concealed.

Discussion

Although, in vertebral HC disease, the dorsal (thoracic) spine region is the most commonly part to be affected by the HC lesions,² however, any part of the vertebral column can be involved by the hydatidosis

process, as mentioned in the literature. Furthermore, the site(s) of the HC can be intra-osseous vertebral, paravertebral and paraspinal with dumbbell extension into the spinal canal,³ epidural,⁴ intradural,⁵

and posterior mediastinal.⁶⁻⁷ The spectrum of lesions produced by the HC may include local tissue destruction by invasion, pressure and vascular damage, pathological fracture and bony deformities and instability, neural tissue damage such as root irritation and/or spinal cord compression, cauda equine syndrome, spondylodiscitis, arachnoiditis, and even rupture into the pleural cavity had been

reported.⁸ The dumbbell extension may appear like a neurofibroma tumour and causes a diagnostic confusion. In the current case, however, it has caused an epidural abscess compressing the spinal cord tissue at T7-T9 vertebrae from a dumbbell extension through the T8 intervertebral foramen from neighboring left sided paravertebral paraspinal site.

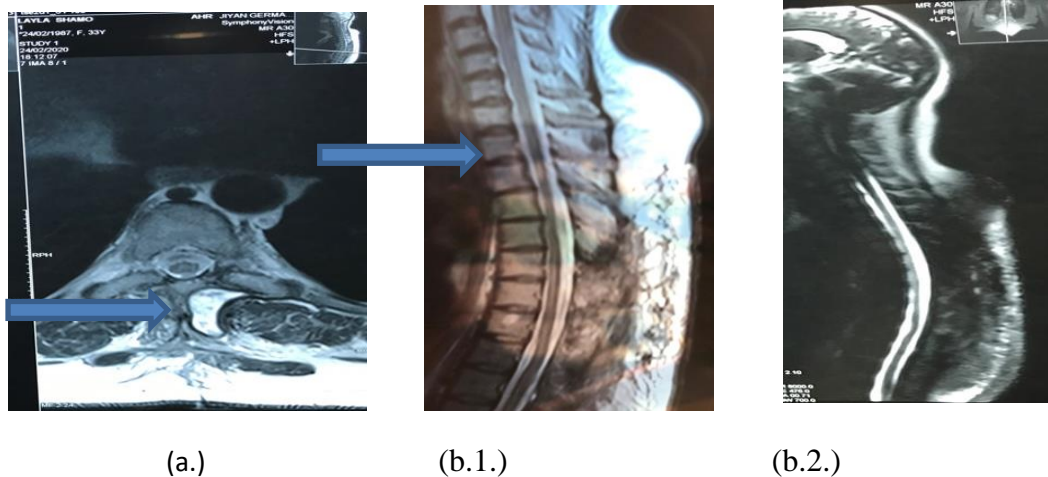


Figure (3): Post-operative MR images, Pre- and post-contrast, T2W phases (a-b).

- (a.) Axial view showing marked shrinkage of the size of the paraspinal mother cyst (arrow); it also shows the circular cross section of the spinal cord free from any compressive lesion.
- (b.)(b.1&2.) Two sagittal views showing freedom of spinal cord from the compressing inflammatory mass (epidural abscess); it also shows the proximal syrinx that has been seen before (arrow).

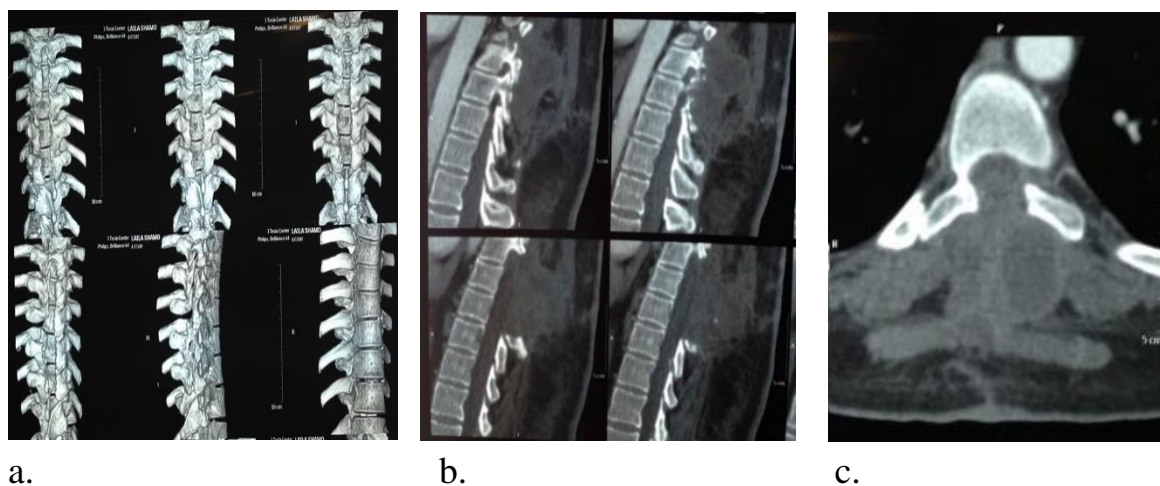


Figure (4): Post-operative CT pictures of part of the dorsal spine (a-c). All confirm local spinal stability.

- (a) 3 D view showing the excision of spinous processes and laminae of T6-T10.
- (b) Sagittal reconstruction. It shows the "hypodense" operative site. (c) Axial reconstruction. It shows the left sided "hypodensity" representing the previous site of ectocyst.

Although there are no specific clinical features, however, taking a detailed history and clinical examination are mandatory. In keeping with the other researchers' findings, the diagnostic imaging, namely, the US, contrast MR and CT, remains the cornerstone in the localization and/or the presumptive pre-operative diagnosis, and in designing the appropriate surgical procedure⁹. Immunological and serological tests, e.g. ELISA IgG, IgM, IgE, and IgA, vary in their specificity and sensitivity; In their study, they evaluated the diagnostic value of semi-purified antigens using ELISA tests; the results had shown that the 53 KDa antigen gave the best specificity (97.5%) and sensitivity (80%)¹⁰; however, they were unavailable at our health unit. Obviously, both the operative finding and the histological examination can achieve the definite diagnosis, as it is the case in the current report. Once the diagnosis is obtained, it is highly recommended to give the patient the appropriate course of Albendazole therapy, with liver function monitoring. However, there are authorities who advise giving Albendazole pre-operatively in order to achieve an acceptable scolical blood level in case rupturing of the HC occurs inadvertently. It is advisable to practice meticulous care during tissue dissection and resection, to avoid rupturing the HC in order to prevent anaphylaxis and recurrence. In case the rupture happens unintentionally, the operative field should be isolated with gauze soaked with 20% Sodium Chloride (NaCl) solution and irrigated with copious amount of the same solution in order to prevent recurrence. The epidural dumbbell

Conclusions

In regions endemic with tape worm hydatidosis, when a patient presents to neurosurgical/orthopaedic services with spinal cord compression by a cystic lesion, clinicians should keep in mind the possibility of HC as the offending causative parasite. Laboratory and imaging techniques are quite helpful in the

extension, from the hydatid cyst mass lesion through the neighboring intervertebral neural foramen into the spinal canal, seen as abscess formation, was presumably due to either infection of a hydatid cyst, or rupture of one, or more, of the cysts with the liberation of the intensely chemically irritant and immunogenic materials causing the inflammatory pyogenic response which ended up with an abscess formation. Histologically, there was confirmation, by the specialist pathologist, of the inflamed tissue that was removed during surgery to be "inflammatory tissue". The culture of the purulent fluid (pus) recovered during the operative procedure was "sterile"; the reason for that was that either due to the "prophylactic antibiotic" given by the anaesthetic team routinely during the peri-operative period of neurosurgical procedures, or it was a chemically-induced abscess formation by the irritant fluid locally released during inadvertent rupture of the hydatid cyst during the course of illness, before arriving to surgical operative moment. In the current case, there was no need to perform any additional instrumental stabilizing procedure as the HC lesions involved the soft tissues only and there was no osseous involvement; following the T6-T10 laminectomy, the post-operative CT confirmed the stability of the spine. Although recurrence is quite possible¹¹, however, both surgery and chemotherapy may achieve a cure in the vast majority of cases; a strategy of follow-up, for quite few years, at least, should be adopted¹²⁻¹³.

diagnostic and localization processes. As the uncomplicated HC shows no contrast enhancement on imaging studies, however, when there is intense contrast enhancement of a dumbbell extension of the HC, this should alert the clinician the possibility of the inflammatory process. Appropriate and successful surgical

removal of HC and associated inflammatory tissue lesion, combined with Albendazole therapy, are highly curative. However, follow-up should be continued

Conflicts of interest

The authors have no conflict of interest to declare. Concerning the scientific and intellectual aspects of the manuscript, all

for quite few years looking out for any possible recurrence.

authors have critically reviewed and approved the final text and are responsible for its content.

References

1. Foad AF, Satir AA. Skeletal manifestations of hydatid cystic disease in Sudan. *J Taibah Univ Med Sci.* (2018); 13(6), 564-71.
2. Liand Q, Xiang H, Leilei X, et al. Treatment experiences of thoracic spinal hydatidosis: a single-center case-series study. *Int J Infect Dis.* 2019; 89:163-8.
3. Cavus G, Acik V, Bilgin E, Gezercan Y, Okten AI. Endless story of a spinal column hydatid cyst disease: A case report. *Acta Orthop Traumatol Turc.* 2018; 52(5):397-403.
4. Kamat AS, Thompson C, Ben Husien M. Staged Surgical Management in the Treatment of Primary Epidural Hydatidosis of the Spine: A Case Series and Review. *Cureus.* 2015; 7: e401.
5. Medjek L, Zenini S, Hammoum S, Hartani M. Intradural hydatidosis of the thoracic spine. Apropos of a case. *Ann Radiol (Paris).* 1991; 34:251-5.
6. Goenka AH, Das CJ, Goel P, Srinivas M, Pangtey GS. Review Giant primary posterior mediastinal hydatid cyst in a child: report of a case and review of literature. *Pediatr Surg Int.* 2009; 25:647-9.
7. Kumar S, Satija B, Mittal MK, Thukral BB. Unusual mediastinal dumbbell tumor mimicking an aggressive malignancy. *J Clin Imaging Sci.* 2012; 2:67.
8. Singh S, Sardhara J, Singh AK, et al. Spinal intradural hydatid cyst causing arachnoiditis: A rare etiology of cauda equina syndrome. *J Craniovertebr Junction Spine.* 2016; 7:282-4.
9. Mrabet FZ, Achrane J, Sabri Y, El Hassani FE, Hammi S, Bourkadi JE. Contribution of imaging in diagnosis of primitive cyst hydatid in unusual localization: pleura. A report of two cases. *Case Rep Radiol* 2018; 2018: 6242379
10. Iraqi W. Diagnostic value of semi-purified antigens of hydatid cyst fluid in human cystic echinococcosis. *Acta Parasitol.* 2016; 61:144-50.
11. Mon ST, Li Y, Shepherd S, Daniel S, Poonnoose S, McDonald M. Recurrence of chest wall hydatid cyst disease involving the thoracic spine in an Australian patient. *J Clin Neurosci.* 2016; 30:132-6.
12. Jain A, Prasad G, Rustagi T, Bhojraj SY. Hydatid disease of spine: multiple meticulous surgeries and a long-term follow-up. *Indian J Orthop* 2014; 48 (05) 529-32.
13. Baykaner MK, Doğulu F, Oztürk G, Edali N, Tali T. A viable residual spinal hydatid cyst cured with albendazole. Case report. *J Neurosurg.* 2000; 93(1 Supl