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Case Report

Hydatid Disease of Spine: A Rare Cause of Chronic Back-Ache Not Responding to Conventional Treatment

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Abstract

Hydatid disease is a parasitic infection caused by tapeworms in human. In human, it is common in liver, spleen and brain. Musculoskeletal involvement is secondary and uncommon, with an incidence of less than 2.5%. Spinal involvement is even rarer with an incidence of 1%. We came across an Afghan national in his early 20s who presented to us with the complains of gradually worsening pain in the upper back for 1.5 years associated with weakness of both lower limbs for 3 months. Within 2 weeks of presentation the neurological deficit progressed to complete paraplegia with loss of sensation below the level of the chest and urine retention. The patient was thoroughly examined clinically. Essential radiological and hematological investigations were performed as well. And a consequent diagnosis of hydatid disease of spine involving the chest wall was made. He was then operated by a multidisciplinary team consisting of cardiothoracic surgeon, orthopedic surgeons and anaesthetists. The cysts from the chest wall and the spine were excised followed by decompression of spine. Lavage with hypertonic saline was done intra-operatively to prevent recurrence. The post-operative period was uneventful and patient showed signs of improvement gradually. He was discharged and prescribed Albendazole and Praziquantel. Patient remained symptom-free for around one year. After a period of one year, signs of recurrence developed. Patient presented to us with paraplegia and had to be re-operated and put on similar post-operative drug regime of albendazole and praziquantel.

Keywords: ***Musculoskeletal hydatidosis, Spinal hydatid disease, Vertebral hydatid disease, Parasitic infection of spine, Cystic echinococcosis of spine.***

Introduction

Hydatid disease, also called as cystic echinococcosis is a parasitic infection caused by a tapeworm. Echinococcosis is a zoonotic infection caused in humans by the larval stage of the Echinococcus genus which produce unilocular or polycystic lesions and are prevalent in areas where livestock is raised in association with dogs. Primary hydatidosis is common in the liver, spleen, and lungs. Musculoskeletal involvement is secondary and uncommon, with an incidence of less than 2.5% [1]. Spinal involvement is rare with an incidence of 1% [2]. Neural compression is common in vertebral hydatidosis. The prognosis is generally regarded as very poor [3].

We herein present a case of spinal hydatid disease that presented with upper back pain and subsequent paraplegia.

Case Report

An Afghan national in his early 20s presented with the complaints of gradually worsening pain in the upper back for 1.5 years associated with weakness of both lower limbs for 3 months. Patient came to us walking without any support with the above-mentioned complaints. He was examined clinically and advised to come for further investigations. After 2 weeks he was brought back on a trolley. His weakness had worsened in the last 2 weeks. He could not move his legs at all. There was sensory loss below the chest and urine retention as well. There was no history of fever, cough with expectoration or any constitutional symptoms. On taking a detailed history it was revealed that patient had had anti-tubercular drug therapy for 6 months on the advice of a local physician but to no avail. However, there were no proofs of any report confirming tubercular pathology as the diagnosis.

Clinical Examination

On performing a complete neurological examination

- Not able to perform Bilateral SLR (Straight Leg Raising)
- Power- Bilateral hip, knee, ankle all muscle groups- 0/5
- Reflexes – Ankle Jerk, Knee Jerk- Grade 2
- 40-50 % sensory hypoesthesia below D6
- Plantar response- mute bilateral

The Local examination findings were as follows

- Diffuse swelling noted in the upper back and left paraspinal region.
- Soft in consistency
- Not adherent to the skin but adherent to the underlying muscles

Investigations

- Blood investigations were done- CBC, PT, APTT, INR, KFT, ESR- all findings were within normal limits.
- Radiological investigations- X- Ray of the Chest, CT scan of the chest and MRI of the dorsal spine was advised

The Chest X-ray showed in-homogenous opacification of left upper and middle zone and expansile osteolytic lesions with surface irregularities involving posterior ends of 5th rib on left side (Figure 1).

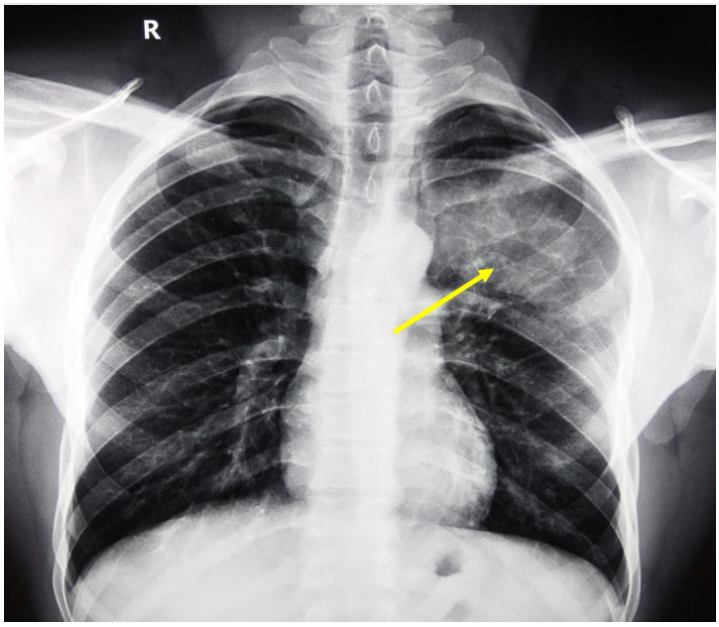

Figure 1: Chest X-ray showed in-homogenous opacification of left upper and middle zone

CT scan of the chest with 3d reconstruction of the bony architecture showed involvement of 5th and 6th ribs, cortical irregularity and erosion with involvement of the corresponding dorsal vertebrae (Figure 2). CT Scan axial cut through the chest showed lobulated, homogenous hypodense, pleural based lesions noted along the parietal pleura in the upper chest wall revealing fluid density with thin calcified walls and septae noted in few of them. Few round to oval hypo-dense lesion were noted within the left sided posterior para-spinal muscles at D5-D6 level (Figure 3). Left lateral cortices of D5, D6 vertebral bodies appears to be partially eroded and irregular (Figure 2 & 3)

Other relevant CT Scan findings

- Intra-spinal extension is likely at D5 level which shows widening of the left neural foramen with the destruction of the left side pedicle and erosion of the left lamina (Figure 3)
- Lung parenchyma- normal (Figure 3)

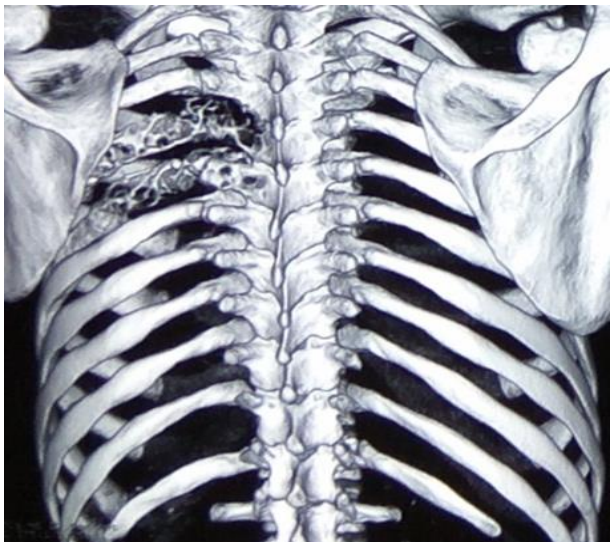

Figure 2- CT scan chest with 3d reconstruction of the bony architecture. Involvement of 5th and 6th ribs, cortical irregularity and erosion with involvement of the corresponding dorsal vertebrae.

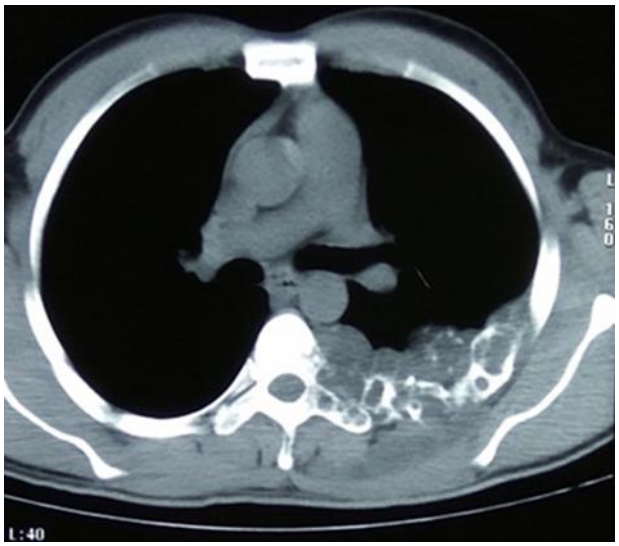

Figure 3: CT Scan axial cut through the chest shows lobulated, homogenous hypodense, pleural based lesions noted along the parietal pleura in the upper chest wall revealing fluid density with thin calcified walls and septa noted in few of them.

MRI of the dorsal spine sagittal cut T1 weighted images showed Iso to hyperintense lesion involving D4-D6 (Figure 4). MRI of the dorsal spine sagittal cut T2 weighted images showed hyperintense lesion involving D4-D6 (Figure 5). Theses cystic lesions showing smaller T1- iso to hyperintense and T2- hyperintense cystic foci within are likely suggestive of daughter cysts. MRI of the Chest

Coronal cut T2 weighted image showed hyperintense lesion over the left side upper chest (Figure 6). MRI axial cut dorsal spine shows lesions also involving posterior extradural space D4-D6 levels on left side causing compression of anterior, lateral and posterior walls of thecal sac at these levels. Evidence of compressive myelopathy was noted at these levels (Figure 7).

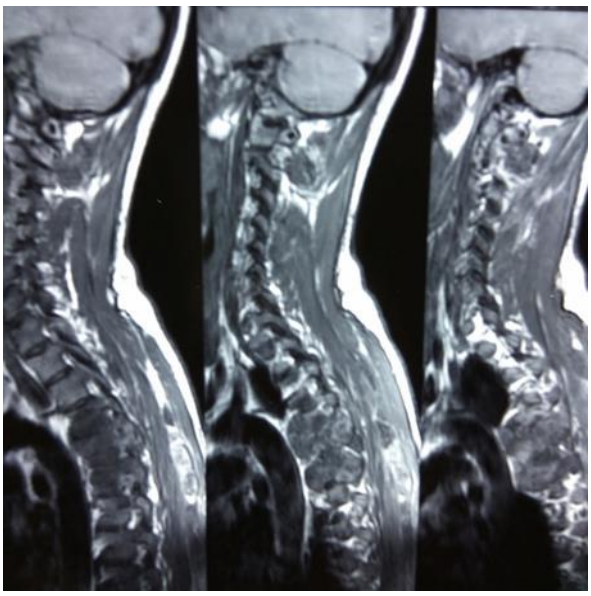

Figure 4: MRI dorsal spine sagittal cut T1 weighted images. Iso to hyperintense lesion involving D4-D6

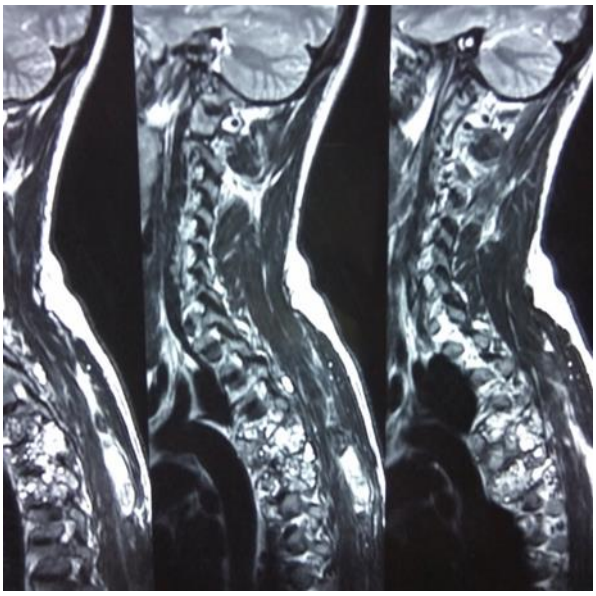

Figure 5: MRI dorsal spine sagittal cut T2 weighted images. Hyperintense lesion involving D4-D6

Figure 6: MRI Coronal cut T2 weighted image showing hyperintense lesion over the left side upper chest.

Figure 7: MRI axial cut dorsal spine shows lesions also involving posterior extradural space D4-D6 levels on left side causing compression of anterior, lateral and posterior walls of thecal sac at these levels.

Diagnosis

Based on the clinical and radiological findings the following diagnosis was made

- Hydatid Disease of the spine with extradural compression of spinal cord at D5, Sensory level D7 with Paraplegia and bladder involvement and
- Involvement of chest wall and ribs

Management

- Left postero-lateral thoracotomy with partial costectomy of left 5th-6th ribs
- Spinal decompression with excision of cyst from D4-D6 vertebral level and irrigation with hypertonic saline to prevent recurrence.

Other Intraoperative findings

- Left posterolateral thoracotomy incision (Figure 8)

- Hydatid cysts adhering to the chest wall and ribs (Figure 9)
- Partially resected 5th and 6th ribs infiltrated with hydatid cysts (Figure 10)
- Daughter cysts curetted out from the body of D6 vertebra (Figure 11)
- Excised hydatid cysts with daughter cysts (Figure 12)

Figure 8: Left posterolateral thoracotomy incision

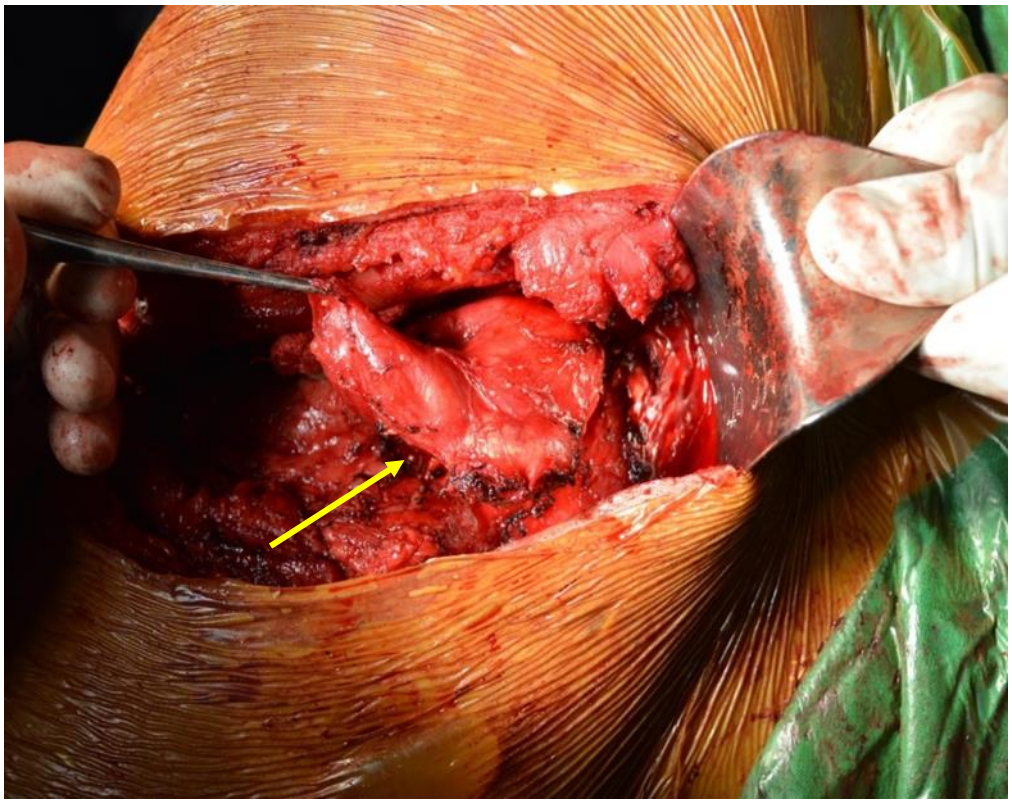

Figure 9: Hydatid cysts adhering to the chest wall and ribs (yellow arrow)

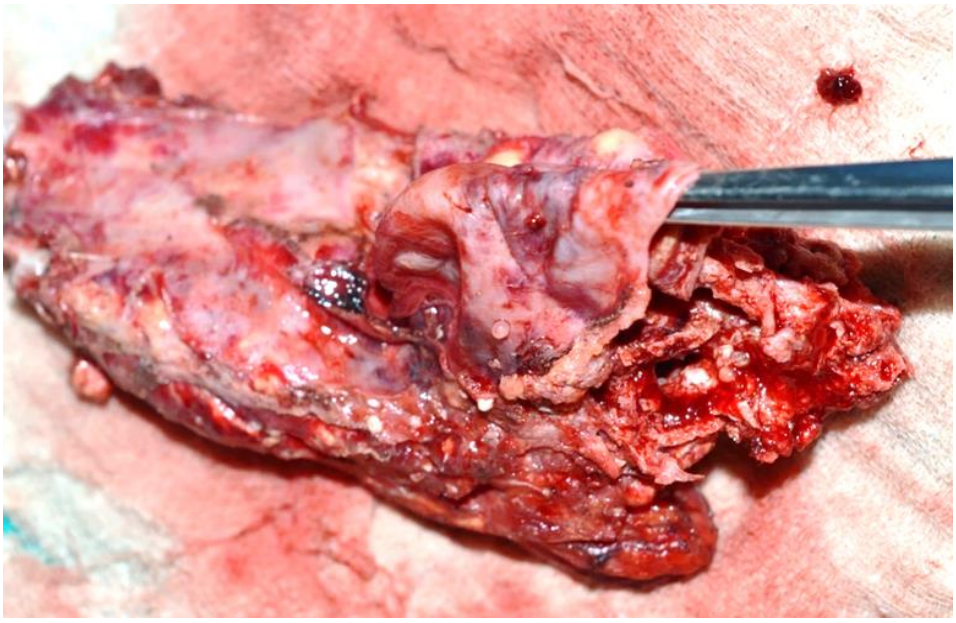

Figure 10: Partially resected 5th and 6th ribs infiltrated with hydatid cysts

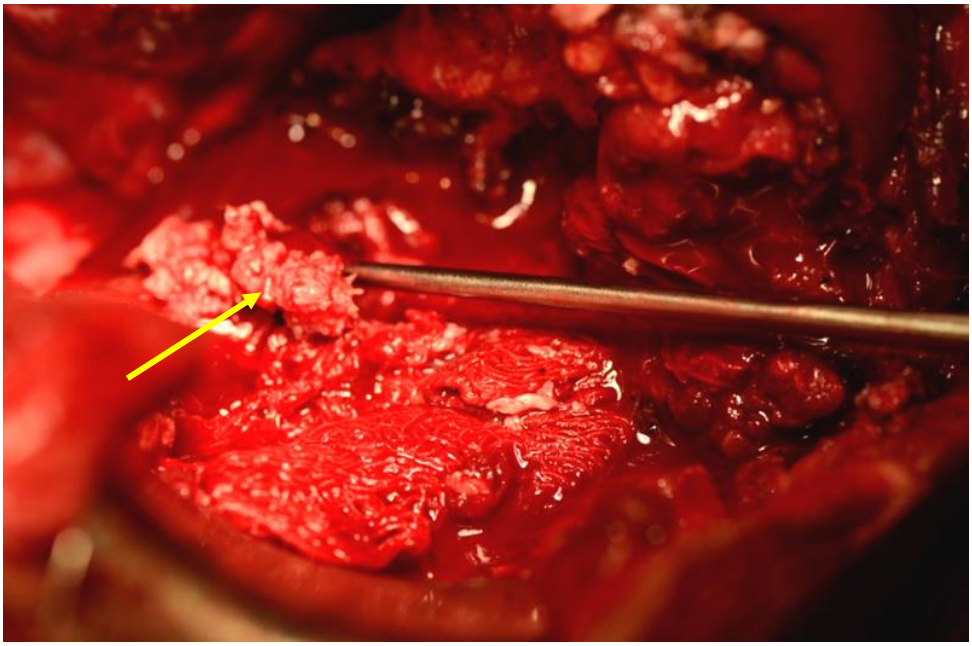

Figure 11: Daughter cysts curetted out from the body of D6 vertebra (yellow arrow)

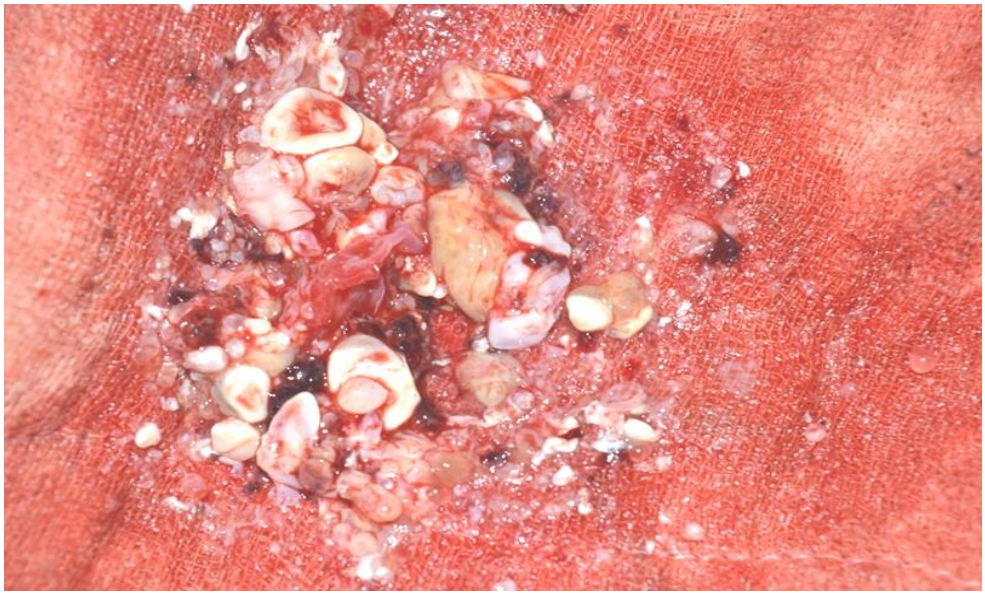

Figure 12: Excised hydatid cysts with daughter cysts

Post-operative management

Post-operative period was uneventful. The post-operative chest X Ray showed complete excision of the 5th and 6th ribs and absence of opacification as compared to the pre-operative X Ray (Figure 13). Clinically, patient showed gradual return of power and sensation in the lower limbs. Patient was prescribed Albendazole- 400mg BD for 28 Days followed by 14 drug-free days. This was repeated for 3 cycles. Praziquantel was prescribed additionally once per week in a dose of 40 mg/kg during treatment with albendazole

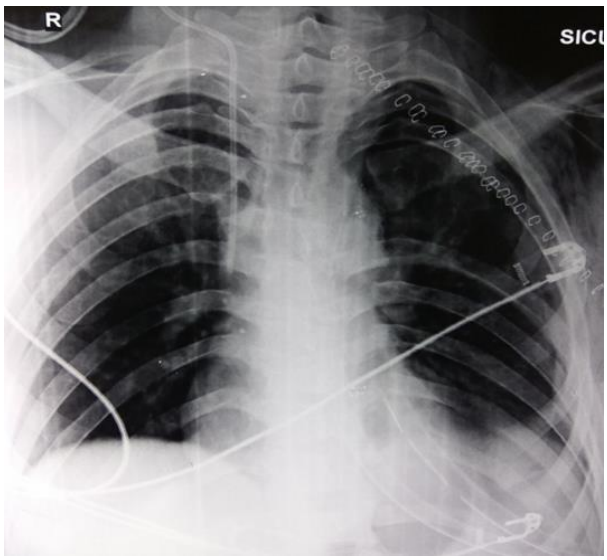

Figure 13: Post-operative Chest X ray.

Prognosis

Patient remained symptom-free for around one year. After a period of one year, signs of recurrence developed. Patient came to us with paraplegia. He had to be re-operated and put on similar post-operative drug regime of albendazole and praziquantel.

Discussion

Skeletal hydatid disease is most common in spine and pelvis, followed by the femur, tibia, humerus, skull, ribs [4]. In osseous locations it manifests as different-sized areas of pure osteolysis and extends to the surrounding soft tissue [5]. Spinal hydatid disease occurs as a direct extension from pulmonary, abdominal or pelvic infestation. It affects thoracic spine most commonly followed by lumbar spine, cervical and sacral spine [6]. Chest wall involvement occurs as the disease spreads by sub-periosteal or by sub-ligamentous path leading to paraspinal extension and involvement of contiguous ribs [7]. Spinal hydatid disease presents with radiculopathy, myelopathy, local pain owing to bony destructive lesions, pathological fracture and consequent cord compression [8,9].

The most common radiological manifestation of skeletal hydatid disease is a lucent expansile lesion with cortical thinning [10]. Bone hydatid disease lacks a typical clinical appearance and image characteristics on X-ray or computed tomography (CT) scan are similar to those of tuberculosis, metastases, giant cell tumour or bone cysts. The primary role of CT in diagnosis is in the recognition of the extraosseous spread of the hydatid disease within the soft tissues [10]. Magnetic resonance imaging (MRI) is the preferred imaging tool in the diagnosis of hydatid cysts located in various parts of body. It shows distinctive diagnostic features. The signal intensity pattern of the hydatid daughter cysts reflects their contents and may vary depending on whether cysts are dead or alive [10]. Fine needle aspiration cytology used is not recommended due to risk of spillage and consequent hypersensitivity reaction and systemic spread of hydatid cyst.

Various literature suggests that Mebendazole and Albendazole, are the only anti-helminthics effective against cystic echinococcosis but Albendazole is the preferred anti-helminthic agent used before and after surgery [10]. Praziquantel is used as combination therapy along with albendazole. Although, surgery is considered as the preferred treatment of choice in spinal hydatid disease [10], surgery alone is not curative. Adjuvant drug therapy as well as intraoperative prophylaxis is recommended [10].

Conclusions

Chronic back-ache with worsening of symptoms and not responding to conventional treatment should be considered as a red flag. Not all cases of backaches should be attributed to common causes such as mechanical back pain, disc disease or tubercular infection especially in the absence of trauma, as differential diagnoses. A high degree of suspicion is indispensable for cases involving parasitic infection of spine to reach at a definitive diagnosis. The geographical location of the patient needs to be elicited and hence the importance of a detailed history. In this case, the patient belonged to a region where people live in close association with dogs and live-stock. This should tickle a cord in the investigator's mind that investigations should also be thought of in the line of infections or disease conditions those are prevalent in that particular geographical location. A thorough investigation including hematological and radiological is of prime importance. As plain radiograph and CT Scan findings appear similar to that of other conditions like tubercular infections, cysts, metastasis or even giant cell tumour, MRI is the investigation of choice. Surgical intervention and intra-operative prophylaxis along with adjuvant drug therapy is the recommended treatment. Despite all these measures, the recurrence rate is quite high and overall prognosis is poor.

Ethics approval and consent to participate

Ethics committee approval- Not Applicable.

Informed consent

Informed consent was obtained from the patient.

Conflicts of Interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

Funding Statement

None/ Not Applicable

Authors' contributions

Dr Alok Sahu collected data including relevant pictures and led the manuscript preparation, incorporating inputs from all authors. Dr Rajesh Senapati and Dr Gopal Krishna Hembram helped in search of literature and contributed to the manuscript especially in writing the discussion and conclusion. Dr Manoj Kumar Sahoo helped in understanding the pharmacological aspect of the treatment and overall supervision of the manuscript.

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