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CASE REPORT Open Access

Primary para-vertebral hydatid cyst in the sub-occipital area of the neck: an unusual case of echinococcosis

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Abstract

Background: Hydatid disease is a parasitic infectious disease caused by *Echinococcus granulosus*. The parasite can form cysts in any part of the body with the liver and lung being the most commonly involved organs. It can rarely occur in other organs like the muscle, bone, pericardium, myocardium, spleen, spine, and neck. Para-vertebral hydatid cyst is very rare and can present with a variable clinical presentation. Surgical excision is the treatment modality of choice and accepted to be curative with a very low recurrence.

Case presentation: We here present a case of primary hydatid cyst in the para-vertebral space in sub-occipital area of the neck who presented with complaints of pain and swelling in the right posterior-lateral side of the neck. X-ray cervical spine revealed suspicious lesion in the sub-occipital area. Magnetic resonance imaging (MRI) was suggestive of multi-loculated cystic lesion. There were no such cysts found in peritoneal viscera or any other organ. Patient was planned for surgery and the cyst was excised. Histopathological examination confirmed the hydatid disease. Post-operatively, the patient was put on albendazole and patient's symptoms disappeared after surgery.

Conclusion: Cysts in any part of the body should be evaluated for hydatid disease especially in the endemic areas. Radiological imaging and serology are important for diagnosis, and surgical excision is the best modality of treatment.

Keywords: Hydatid cyst, Parasite, Para-vertebral space, Surgery

Background

Hydatid disease is caused by larval form of *Echinococcus granulosus*. It is commonly seen in cattle rearing areas like Australia, New Zealand, South America, Mediterranean countries, and countries with poor socio-economic conditions [1–3]. It is commonly seen in the liver and lung [4–6] and rarely involves some unusual locations such as the bones, heart, brain, spleen, pericardium, myocardium, and muscles [7–10]. Primary hydatid cyst of muscle is very rare because of the presence of lactic acid creating unfavorable environment for its growth [11, 12]. It constitutes only 2–3% of all cases [13]. We present a case of hydatid cyst in para-vertebral space in sub-occipital area of cervical region in a 50-year-old male presented to our neurosurgery outpatient clinic at our institute.

Case presentation

A 50-year-old male patient was presented to our neurosurgery outpatient clinic with chief complaints of pain and swelling in the right posterior-lateral side of the neck from the last 6 months. The pain was dull and boring without any radiation. There was no history of fever, vomiting, altered sensorium, loss of consciousness, or vertigo. There was no history of tuberculosis, trauma, surgery, or other significant medical history. General physical examination was unremarkable. There was no cranial nerve involvement. Neurological examination was grossly normal. Power of both upper limbs was 5/5 both proximally and distally. Sensations were intact. Hematological investigations were also within normal range. X-ray cervical spine showed suspicious circular

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Fig. 1 X-ray showing suspicious lesion just blow the occipital area, posterior to spinous process of cervical spine C1 and C2

lesion in the sub-occipital area, at the level of cervical spine C1 and C2 (Fig. 1). Chest X-ray was also grossly normal. Abdomen ultrasonography (USG) was normal. Magnetic resonance imaging (MRI) showed multi-loculated cystic lesion about 77 × 74 × 44 mm in the right posterio-lateral para-vertebral space at the level of cervical vertebrae C1 and C2 with an extension into posterior triangle (Fig. 2a–d). There was no extension in the spinal canal, and the spine was absolutely normal. There were no such hydatid cysts found in peritoneal viscera or any other organ. On the bases of the MRI findings, a diagnosis of hydatid cyst was made, and hydatid serology was done (ELISA for *Echinococcus granulosus*), which was reported to be positive.

The patient was planned for surgery after proper workup. The cyst was approached by posterior sub-occipital incision, and the cyst was en-masse excised without any spillage (Fig. 2). Hydatid cyst about 8 × 8 × 5 cm was found in para-vertebral space in the sub-occipital area just posterolateral to the C1 and C2 cervical vertebrae. The cyst was located in the intramuscular space. Operative area was packed for 7 min with a 3% sline soaked gauge after removal of the cyst. The excised cyst was opened; daughter cysts along with hydatid fluid were seen within the cystic cavity. Histopathological examination confirmed the hydatid disease. Post-operative period was uneventful. The patient was discharged on 5th post-operative day and was put on albendazole 600 mg per day (400 mg in the morning and 200 mg in the evening) for 3 months. The patient became symptom free after the surgery.

Discussion

Hydatid disease also called echinococcosis (or hydatidosis) is a zoonosis, caused by the larval stage of cystode, *Echinococcus* [14]. There are four species of the genus *Echinococcus* responsible for the infection in humans: *Echinococcus granulosus*, *Echinococcus multiloculatis*, *Echinococcus vogali*, and *Echinococcus oligarthus* [15]. Life cycle of *Echinococcus granulosus* involves definitive and intermediate host to complete its life cycle. Carnivores (dogs, wolves etc.) are the definitive hosts while the herbivores (sheep) are the intermediate hosts. Humans are the accidental dead-end hosts in the life cycle. Humans ingest the eggs from which embryo develops. Embryo penetrates the intestinal wall, enter the portal circulation, and get trapped in the liver [16], while few may manage to reach other organs via systemic circulation. Hydatid cyst may be present in any part of the body. The liver and lung are involved in 90% of cases [17] while the other organs like the brain (1–2%), kidney (3%), and bone (1–4%). The muscles, heart, spleen, breast, and pancreas are very rarely affected. Muscle hydatidosis have mostly associated with hepatic cysts [18]. Few case reports have been reported in the pectoralis major muscle [19], adductor muscles of the thigh [20], rectus and spinal muscles [21], and gluteal and neck muscles [18]. Our patient presented with a primary hydatid cyst in para-vertebral space in the sub-occipital area at the level of cervical vertebrae C1 and C2 (without any evidence of hydatid cysts in other body parts) which to our knowledge has not been reported in medical literature till date (Fig. 3).

Hydatid cyst generally remains asymptomatic with a very slow growth rate. The latency period ranges from 5 to 20 years [22, 23]. Clinical manifestation depends on the anatomical site involved and size of the cyst. Clinical symptoms are usually due to the pressure on adjacent structures or obstruction. Secondary infections and anaphylactic reaction to ruptured cystic fluid are the most common complications [24]. Our case presented with neck swelling in the sub-occipital area with neck pain.

Hydatidosis being asymptomatic is usually diagnosed by imaging done for other reasons. X-ray, USG, computed tomography (CT), and magnetic resonance imaging (MRI) are the main investigations used for the

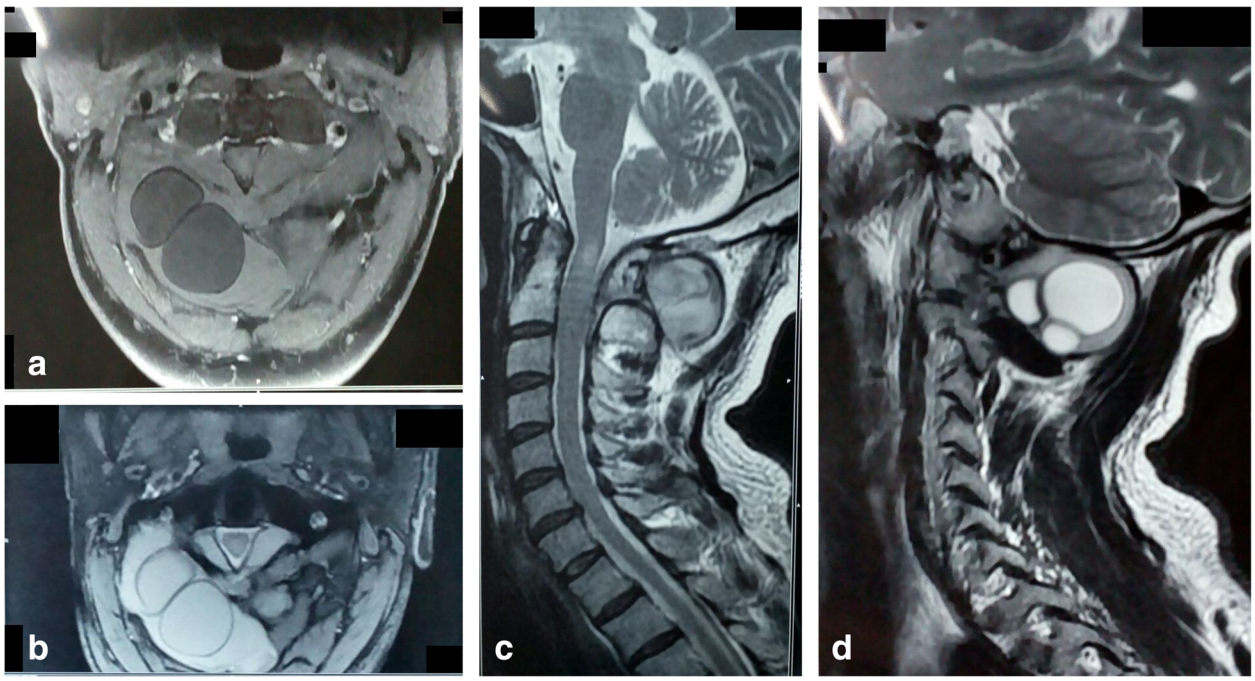

Fig. 2 **a** MRI neck (T1 weighted, axial view) showing cyst the in sub-occipital area. **b** MRI neck (T2 weighted, axial view) showing cyst in the sub-occipital area. **c** MRI neck (T1 weighted, sagittal view) showing cyst in the sub-occipital area. **d** MRI neck (T2 weighted, sagittal view) showing cyst in sub-occipital area

Fig. 3 Showing intraoperative picture of the cyst in intermuscular space in the sub-occipital area

detection of cysts. CT and MRI have very high accuracy in establishing diagnosis of hydatid cyst [25, 26]. Diagnose is confirmed by serological tests. Various serological tests are used to detect specific serum antibodies and circulation antigens: hemagglutination test, immunoelectrophoresis (IEP), ELISA for echinococcal IgG, etc. ELISA have sensitivity-95% and specificity-94% [27]. Serological tests can have false negative and false positive results; therefore, positive tests do not confirm the diagnosis and negative test do not exclude the disease [28]. In the present case, X-ray cervical spine showed a suspicious lesion in the sub-occipital area, posterior to C1 and C2 cervical vertebrae. The diagnosis was then made by MRI and confirmed by ELISA.

Treatment for the hydatid disease depends on the location of the cyst, size, and health status of the patient. Surgical excision is the optimal treatment of large and symptomatic hydatid cysts [29]. Other modalities of treatment include anti-helminthic drugs (for small and asymptomatic cysts) and scolecidal agents. Depending on the clinical situation, radical or conservative approach can be adopted. Total cystectomy has the least recurrence rate but is associated with high morbidity. Cyst in the intraperitoneal viscera can be approached by simple deroofing and enucleation, but care should be taken to prevent the spillage. Residual cavity can be managed by various methods [29]. Puncture-aspiration-injection-reaspiration (PAIR) is an ultrasound-guided technique consisting of

puncture of the hydatid cyst and evacuation of the contents, injection of scolecidal agents such as 95% ethanol, and reaspiration of the contents of the cyst. Although this technique has long been discouraged because of the potential complications such as iatrogenic spread of disease and anaphylactic shock, there is an expanding literature suggesting that PAIR is effective for the treatment of primary uncomplicated hepatic cysts [30]. This technique has also been used as a modality of treatment in muscular hydatid cyst [31]. In the present case, we have performed total cystectomy with captionage of the residual cavity.

Conclusions

Hydatid cyst can occur in any part of the body, so cysts in any part of the body should be evaluated for hydatid disease especially in the endemic areas. The radiological imaging and hydatid serology play an important role in diagnosis with high accuracy. Treatment varies from medical management to surgical excision. We consider surgical excision as the best modality of treatment.

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Availability of data and materials
The data and material used during the current study is available with the corresponding author on reasonable request.

Authors' contributions
DPMUD, WAA, and MLA conceived and designed the manuscript. DPMUD, MLA, and GBA helped in the definition of the intellectual content. DPMUD, WAA, KAR, GBA, and WNG contributed in the literature search. DPMUD, WAA, and MLA provided the clinical studies. DPMUD, MLA, and WNG involved in data acquisition. DPMUD, WAA, MLA, and GBA analyzed the data. DPMUD, MLA, and KAR gave statistical analysis. DPMUD, WAA, MLA, and GBA prepared the manuscript. DPMUD, MLA, KAR, and WNG edited the manuscript. DPMUD, WAA, MLA, and KAR reviewed the manuscript. DPMUD, WAA, MLA, KAR, GBA, and WNG are guarantors of the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate
Not applicable.

Consent for publication
Written informed consent was obtained from the patient to participate in the treatment and publication.

Competing interests
The authors declare that they have no competing interests.

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