

An Unusual Location Of Hydatid Disease Hydatid Cyst Of Rib

Sameer MohIdeen

Department of Cardiothoracic & Vascular surgery. College of Medicine,
Alnahrain University.

Abstract:

The Hydatid cyst is endemic in our country, but bone lesions are less common. The disease often takes the appearance of abscess or malignant lesion. Osseous Hydatid disease is caused by parasitic tapeworm Genus Echinococcus. Patients usually present with pain , swelling, or pathological fracture.

The course of the disease is generally slow and laboratory tests are frequently negative . Diagnosis is generally made through the combined assessment of clinical, radiologic, and laboratory data.

Living in a rural area is an important factor for the disease/ The gold standard for therapy is radical removal of the ribs or chest wall.

We present the case of a 42- year old man with costal echinococcosis of the right (8th) rib ,complete excision of the rib ,followed by three months of Albendazole 10mg/kg/day.

Key word: Hydatid cyst, Chest wall, Rib, Echinococcosis, Albendazole, Rural area.

الخلاصة :

الأكياس المائية متوطنة في بلدنا، ولكن أكياس المائية للعظام هي أقل شيوعا. هذا المرض غالبا ما يظهر كحالة خراج أو آفة خبيثة. ويتسبب مرض الأكياس المائية للعظام من قبل الطفيلية الشريطية المشوكة . المرضى يقدم عادة مع ألم، تورم، أو الكسر مرضي. ويعتبر هذا المرض بطيء بشكل عام، والاختبارات المعملية سلبية في كثير من الأحيان. يتم عادة التشخيص من خلال التقييم المشترك ، الفحص السريري ، والإشعاعي، والبيانات المخبرية.

الذين يعيشون في منطقة ريفية يشكل عاملا مهما لهذا المرض / المعيار الذهبي لعلاج هو إزالة جذرية من الأضلاع أو جدار الصدر. دراسة عن مريض رجل يبلغ من العمر 42 سنة مع أكياس مائية في الضلع الثامن الأيمن من جدار الصدر وقد تم الاستئصال الكامل للضلع، تلاها اعطائه ثلاثة أشهر من دواء البندازول 10مليغرام /كغم /يوم .

Introduction:

Hydatid disease is primarily an illness of residents in rural areas who frequently come into contact with carnivores, sheep, and cows. Echinococcus granulosus is extremely widespread, with high rates of infection in eastern and southern Europe, the Middle East, northern Africa, and South America^[1].

IRAQ is also a very important area to be studied for this disease. The osseous hydatidosis, especially when located in the rib, is a very rare disease.

Human echinococcosis, commonly called hydatid disease is a zoonotic infection caused by larval forms of small tapeworms of the genus Echinococcus. In humans, the two main forms are due to Echinococcus granulosus and, less

frequently, Echinococcus multilocularis (alveolaris).^[2]

Hydatid disease most commonly involves liver and lung but is rarely encountered in the rest of body, including the skeletal system. Liver (60%) and lungs (20% to 30%) are the most affected by the disease^[3]. Osseous hydatidosis is uncommon (0.9% to 4%) especially the ribs^[3].

Bone lesions are always primary; secondary lesions are due to recurrence and occur as an isolated finding^[4].

The diagnosis is made through the combined assessment of clinical, radiological and laboratory findings^[5, 6].

The role of imaging in this condition is to distinguish it from aneurysmal bone cyst, giant cell tumour and metastasis so that appropriate surgical procedure can be

performed to prevent anaphylactic shock, which may be a fatal complication of surgery.

We present a rare cause of rib destruction by Hydatid disease, extending along the eight rib. To the best of our knowledge, there is no previous report of hydatid cyst involving a whole rib without apparent clinical symptoms.

Case Report:

42years old man from the north of Baghdad (rural areas), presented with chest pain right side, a sense of numbness in the upper abdomen for four months duration, he had no history of previous symptoms associated with the lesion. On physical examination, there was no fever, an arterial pressure was within normal limits. Chest was clear normal vesicular breathing.

The patient had history of operation right thoracotomy for hydatid disease (10-yaers) before. Local part x-ray and Contrast enhanced CT scan were performed. Penetrated view for right lower ribs showed presence of a lobulated expansile lytic lesion involving the posterior aspect of right 8th, rib (Fig.1)

Adjacent ribs were normal. On CT scan there was presence of a well-defined multi loculated predominantly cystic density lesion with internal septations noted involving the posterior part of the right 8th, rib (Fig.2).

Because of high suspicion of hydatid disease for the cystic lesion with benign nature, an operation was planned and performed on the patient without further investigation. An incision was performed along the lateral arch of the eight rib,(Fig.3) Was completely removed by disarticulation from the costo-verteberal joint.

When the capsule of the specimen was incised, the daughter cysts were reveled (Fig-4). Histopathological examination of the specimen also confirmed the diagnosis of hydatid cyst.

No complication occurred post-operatively. Albendazole tablets 10mg/kg/day for three months after the operation. Follow-up the patient for two years is still in good condition and has no cyst relapse.

Discussion:

Echinococcosis granulosus is extremely widespread with high rates of infection in southeastern Europe Middle East, North Africa and South America including our country^[7,8]. Echinococcosis granulosus is encountered much more frequently than Echinococcosis multilocularis, and causes multi-loculated lesions in soft tissues and viscera more frequently compared with E. multilocularis.^[9] Although hepatic and pulmonary localization is the most frequent, it may be determined in any part of the body from head to toe.^[10] However, bony localization particularly in the rib(s), is exceptional. When costo-vertebral echinococcosis occurs, patients usually are admitted with complaints, sometimes with neurological complaints according to localization of the cyst^[11]. In some cases, concomitant lesions elsewhere ,especially in the lungs, may be detected^[12]. In our case, there was neither any other lesion nor any symptom including neurological and non-neurological ones.

Although nearly the whole rib was destroyed in the case, no extension of the cyst to adjacent tissues was detected. Sometimes size of hydatid cyst may increase without apparent clinical symptom; however, an osseous involvement without symptoms is exceptional.

In 1978, Panahi ^[13] reported that 39 costal echinococcosis cases were published. Rong and Nie ^[14]. presented 20 cases of hydatid disease of bone revealed by roentgenogram examination during the period of 1957 to 1980. The ribs were involved with 2 of the patients. In subsequent years, Stamatis and Greschuchna^[15]. Also published studies of 14

patients with hydatid disease of the lung and chest wall that had operations at the Ruhrlandklinik in Essen, Germany since 1976.

In two of these patients, echinococcosis of the rib was seen. These patients resided in Mediterranean countries, the clinical symptoms were not very marked, and the specific laboratory tests were seldom positive. The exact incidence of rib echinococcosis is unknown. In 2004, less than 50 cases of costal echinococcosis had been reported.^[16] A retrospective study by Thameur et al found eight cases (0.49%) with costal involvement out of 1619 cases with thoracic hydatid disease^[17].

Osseous involvement in hydatid disease is seen in the spine, pelvis, femur, tibia, humerus, skull and ribs. The posterior ends of the ribs are most commonly involved in costal echinococcosis. Cysts grow along the long axis of the rib causing expansion of the cortex where it meets more resistance from the solid cortical portion of the rib^[11, 12].

Costal echinococcosis may be classified as an intraosseous form and an extraosseous form. The intraosseous form may be further classified into a solitary costal form and a costo-vertebral form. The solitary costal form represents an area of multiloculated rib destruction without periosteal reaction.

The differential diagnosis of such a radiographic picture includes giant cell tumour, osteolytic metastases, plasmacytomas, aneurysmal bone cyst and cystic neurofibromas^[12].

Biopsy is contraindicated in echinococcosis due to fear of dissemination of scolices and other potentially fatal complications^[11]. However, review of recent literature suggests that aspiration cytology is the procedure of choice in suspected cases of skeletal echinococcosis^[5].

The gold standard in the therapy of this disease is the radical resection of the rib(s) involved. It has been proposed that better results are obtained by combining surgery with anthelmintic drugs like mebendazole or Albendazole for a period of 3 months should be considered^[3].

However, there are sporadic case reports of percutaneous aspiration of these cysts using ultrasound or CT guidance with pre-medication with Albendazole.

In cases of osseous hydatidosis, even after radical removal of the parasites, the World Health Organization (WHO) suggests adjuvant chemotherapy with mebendazole or Albendazole for at least 2 years after surgery.

In cases where only a palliative treatment is possible, the anthelmintic drug administration can be continuous. To conclude, Computed tomography (CT) is still the best method for diagnosis and post therapy follow-up of osseous hydatidosis.

On CT, skeletal cystic hydatidosis appears as one or several closely related, well-defined, osteolytic lesions.

There may be bone expansion, cortical thinning, cortical destruction, sclerosis, honeycomb appearance, and extension into adjacent soft tissues as depicted in the case above.



Figure-1: C-T Scan shows hydatid disease of the rib.

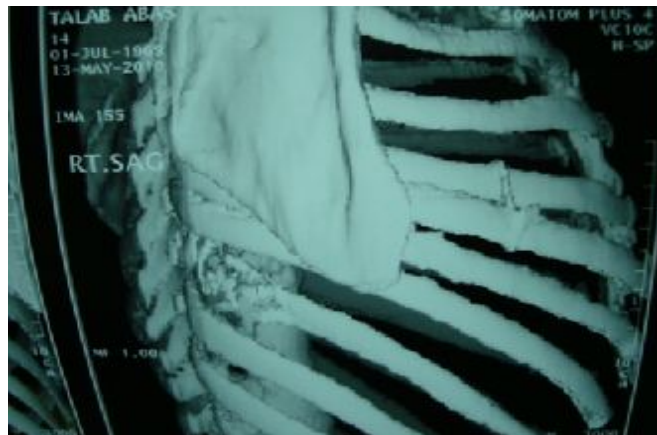


Figure- 2: bone destruction involved by hydatid cyst.



Figure -3: shows the resection of the affected rib from the cost-vertebral junction.



Figure -4: shows the specimen of the rib and the daughter cysts.

References:

- 1- Petersen, C. and Mills, J. Parasitic infections. In: Murray J. F. Nadel J. A. eds. Textbook of respiratory medicine. Philadelphia: WB Saunders Company, 1994. Pp: 1201-1243.
- 2- Dogan, R.; Yuksel, M. and Cetin, G. Surgical treatment of hydatid cysts of the lung: report on 1055 patients. Thorax 1989. Vol. 44. Pp: 192-199.
- 3- Tomos, P.; Kakaris, S.; Lachlan's, E. and Karakatsani, A. Secondary echinococcosis of the rib and soft tissues. Respiration. 2005. Vol. 72. (5). Pp: 542.
- 4- Gezer, S. T.; Altinok, Y.; Agaçkara, A. and Tastepe, I. Hydatid disease of the first rib causing thoracic outlet syndrome. Medical Principles and Practice. 2007. Vol. 16. (1). Pp: 68-70.
- 5- Karaoglanoglu, N.; Gorguner, M. and Eroglu, A. Hydatid disease of the rib. Ann Thorac Surg 2001. Vol. 71. Pp: 372-3050
- 6- Rong, S. H. and Nie, Z. O. Hydatid disease of bone. Clin Radiol 1985. Vol. 36. Pp: 301
- 7- Karaoglanolu, N.; Gorguner, M. and Eroglu, A. Hydatid disease of rib. Ann Thorac Surg 2001. Vol. 71. Pp : 372-37
- 8- Saglam, L.; Akgun, M.; Kaynar, H.; Gorguner, M.; Mirici, A. and Polat, P. Human, pulmonary, cystic echinococcosis in eastern Turkey. Ann Trop Med Parasitol. 2003. Vol. 97. Pp: 531-3.
- 9- Bonakdarpour, A.; Zadeh, Y. F.; Maghssoudi, H.; Shariat, S. and Levy, W. Costal echinococcosis. Report of six cases and review of the literature. Am J Roentgen Radium Ther Nucl Med. 1973. Vol. 118. Pp: 371-7.
- 10- Polat, P.; Kantarci, M.; Alper, F. ; Suma, S.; Koruyucu, M. B. and Okur A. Hydatid disease from head to toe. Radio graphics. 2003. Vol. 230 Pp: 475-94.
- 11- Raut, A. A.; Nagar, A. M.; Narlawar, R. S.; Bhatgadde, V.L.; Sayed, M. N. and Hira, P. Echinococcosis of the rib with epidural extension a rare cause of paraplegia. Br. J. Radial. 2004. 77. Pp: 338-41
- 12- Sebit, S.; Tunc, H.; Gorur, R.; Isitmangil, T.; Yildizhan , A. and Us, M. H. et al. The evaluation of 13 patients with intrathoracic extrapulmonary hydatidosis. J Int Med Res. 2005. Vol. 33. Pp: 215-21.
- 13- Panahi, F. Costal echinococcosis. Report of one case and review of the literature. Sem Hop. 1978. Vol. 54. Pp: 1389-1392.
- 14- Rong, S. H. and Nie, Z.Q. Hydatid disease of bone. Clin Radiol 1985. Vol. 36. Pp: 301-305.
- 15- Stamatas, G. and Greschuchna, D. Echinococcus cysticus costalis: report of 2 cases and review of the literature. Pneumologie. 1989. Vol. 43. Pp: 213-216.
- 16- Thameur, H.; Chenik, S.; Abdel-moulah, S.; Bey, M.; Hachicha, S. and Chemingui, M. *et al.* Thoracic hydatidosis. A review of 1619 cases. Rev Pneumol Clin 2000. Vol. 56. Pp: 7-15.
- 17- Kilic, D. F.; Tercan, E.; Sahin, A. ; Bilen, and Hatipoglu, A. Unusual radiologic manifestations of the echinococcus infection in the thorax. Journal of Thoracic Imaging, 2006. Vol. 21. (1). Pp: 32-36.