

## An unusual location of hydatid disease: an asymptomatic case with rib destruction

*Kist hidatiğin nadir bir yerleşimi: Kosta destrüksiyonlu asemptomatik bir olgu*

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A 57-year-old man was admitted to our hospital with a pleural based lesion which was incidentally detected on a chest radiograph. A chest tomography revealed a mass lesion extending left sixth rib. The mass was removed by a left postero-lateral thoracotomy. Daughter vesicles were seen when the lesion was incised postoperatively. In this article, we presented a rib hydatidosis which is very uncommon asymptomatic presentation of hydatid cyst disease with its surgical management.

**Key words:** Asymptomatic; rib destruction; hydatid disease; surgery.

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)*.<sup>[1]</sup> Hydatid disease most commonly involves liver and lung, but is rarely encountered in the rest of body, including the skeletal system. Musculo-skeletal involvement is reported in only 1-4% of cases, and primary hydatid disease originating from the ribs are extremely rare.<sup>[2]</sup>

We present a rare cause of rib destruction by hydatid disease, extending along the sixth 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

A 57-year-old male farmer was admitted to hospital for inguinal hernia operation. Routine preoperative evaluation chest X-rays, incidentally detected a mass lesion, and he was referred to our department.

Akciğer grafisinde tesadüfen plevral tabanlı kitle tespit edilen 57 yaşındaki hasta kliniğimize başvurdu. Göğüs tomografisinde sol altıncı kot trasesi boyunca uzanan ve altıncı kotu destrükte eden kitle lezyon tespit edildi. Sol arka-yan torakotomi yapılarak kitle rezeke edildi. Ameliyat sonrası kitlenin kapsülü açıldığında kız veziküller görüldü. Bu yazıda kist hidatik hastalığında çok nadir ve asemptomatik seyreden kot kist hidatiğinin cerrahi tedavisi sunuldu.

**Anahtar sözcükler:** Asemptomatik; kosta destrüksiyonu; kist hidatik; cerrahi.

He had no history of previous symptoms associated with the lesion. On physical examination, there was no fever, and arterial pressure was within normal limits. Chest examination showed diminished expansion of left thoracic cage on inspiration, dull to percussion and diminished breath sounds over the left middle hemithorax on auscultation. His chest X-ray revealed a pleural based, well-circumscribed and lobulated mass lesion which nearly filled the left hemithorax, with absence of the sixth rib ipsilaterally (Figure 1a). The computed tomography scan of chest showed a sharply demarcated cystic lesion benign in nature extending along the sixth rib trace by destructing it (Figure 1 c-f). Routine laboratory tests were normal. However, indirect hemagglutination test for hydatid disease was positive (1/512).

Because of high suspicion of hydatid disease for the cystic lesion with benign nature, surgical removal was planned without further investigation, and postero-lateral thoracotomy was performed. Beyond the m. latissimus dorsi and m. serratus anterior, extrathoracic protrusion of the lesion extending along the

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6<sup>th</sup> rib (Figure 2a) was seen. The 6<sup>th</sup> rib was completely removed by disarticulation from the costo-vertebral joint. The lesion was 7x26 cm in diameter. The thoracic wall was reconstructed by using prolene mesh; to prevent lung herniation, since the thoracic wall defect was too large (7x26 cm in diameter) following removal of the mass lesion causing rib destruction. When the capsule of the specimen was incised, the daughter vesicles were revealed (Figure 2b). Histopathological examination of the specimen also confirmed the diagnosis of hydatid cyst. No complication occurred postoperatively (Figure 1b). Albendazole at a dose of 10 mg/kg per day was administered after the operation for three months. At the 10 month follow-up, the patient was healthy.

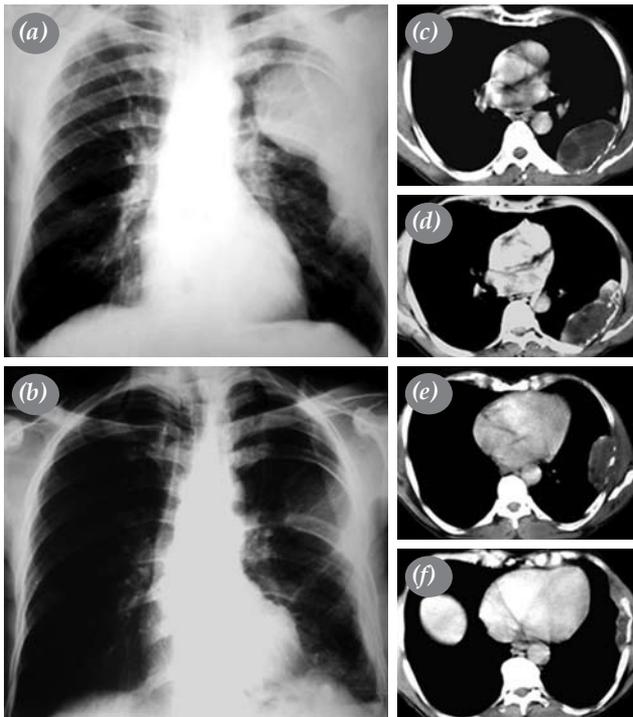
## DISCUSSION

*Echinococcus granulosus* is extremely widespread with high rates of infection in southeastern Europe, Middle East, North Africa and South America including our country.<sup>[3,4]</sup> *Echinococcus granulosus* is encountered much more frequently than *Echinococcus multilocularis* and causes multi-loculated lesions in soft tissues and viscera more frequently compared with *E. multilocularis*.<sup>[5]</sup>

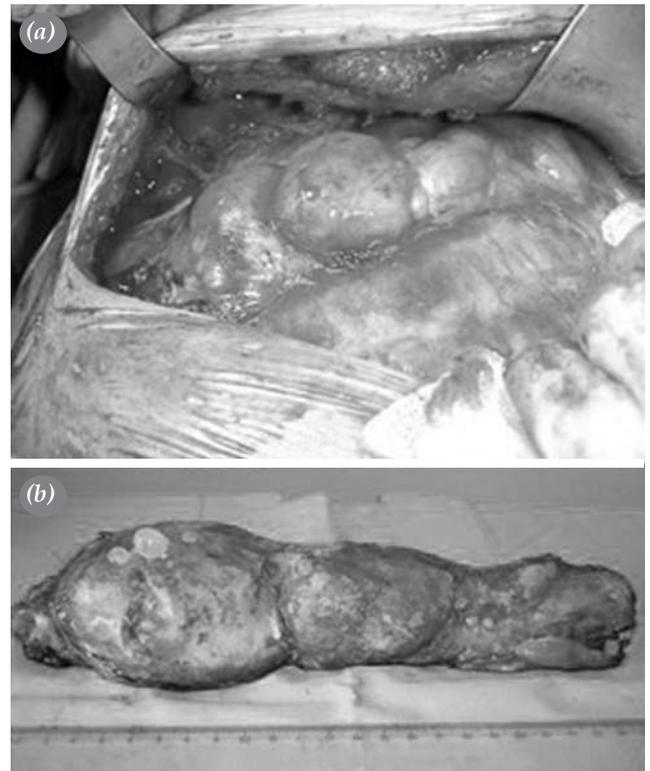
Although hepatic and pulmonary localization is the most frequent, it may be determined in any part of the body from head to toe.<sup>[6]</sup> However, bony localization particularly in the rib(s), is exceptional. When costo-vertebral *echinococcosis* occurs, patients usually are admitted with complaints, sometimes with neurological complaints according to localization of the cyst.<sup>[7]</sup> In some cases, concomitant lesions elsewhere, especially in the lungs, may be detected.<sup>[8]</sup> 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.

The exact incidence of rib *echinococcosis* is unknown. In 2004, less than 50 cases of costal *echinococcosis* had been reported.<sup>[8]</sup> A retrospective study by Thameur et al, found eight cases (0.49%) with costal involvement out of 1619 cases with thoracic hydatid disease.<sup>[9]</sup>

Osseous involvement in hydatid disease is seen in the spine, pelvis, femur tibia, humerus, skull and ribs. In the cases with osseous hydatidosis, absence of pericyst formation allows aggressive proliferation of the parasite



**Fig. 1.** (a) Pre- and (b) postoperative chest X-rays and (c-f) pre-operative thorax computed tomography images of the case. (a, b) The cyst, which is located around the 6<sup>th</sup> rib of left chest wall, was totally removed surgically. Thorax computed tomography images show extension of the cyst along the sixth rib trace, as well as rib destruction.



**Fig. 2.** (a) Intra- and (b) postoperative images of the cyst. The daughter vesicles were revealed after incision of cyst capsule.

along the lines of least resistance, especially along the bone canals. The posterior end of rib is 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.<sup>[5,8-10]</sup>

The typical appearance of hydatid disease in chest X-ray and CT of thorax are mostly sufficient for the diagnosis, and diagnostic difficulties in non-complicated cases are very limited in our clinical experience since the disease is endemic in our country, especially in our region, eastern Anatolia. However, we usually confirm our preoperative diagnosis with histopathological examination of surgical specimens as it was done in our case.

The treatment of choice of this disease is the radical resection of the rib(s) involved. However, medical treatment is limited to cases with inoperable disease. It may be use as an adjuvant therapy in operable cases. Use of antihelminthic drugs for both preoperative treatment and postoperative prophylaxis helps in the reduction of recurrence of this disease. We gave a medical treatment for prophylactic purpose (10 mg/kg/day albendazole) for three months in postoperative period without observing any recurrence or side effect associated with the treatment.

The present case depicts an unusual presentation of hydatid disease with rib involvement.

#### **Declaration of conflicting interests**

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