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Case report
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Primary mediastinal hydatid cysts

Primer mediastinal kist hidatikler

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Abstract
Hydatid cysts are a parasitic disease caused by *Echinococcus granulosus*, and 0.5 to 2.6% of all hydatid cysts are in the mediastinum. In this study, we presented primary mediastinal hydatid cysts. All patients underwent chest X-ray and thoracic computed tomography (CT), and only one patient underwent thoracic magnetic resonance imaging (MRI) due to the association with spinal cord. All lesions removed through thoracotomy were confirmed pathologically, and no postoperative complications were observed, except for one patient. Anthelmintic treatment was started in one patient with vertebral involvement. No recurrence was observed in any of the patients. Although mediastinal hydatid cysts are rarely encountered, it is difficult to differentiate mediastinal hydatid cysts from other mediastinal diseases. However, the diagnosis is often made intraoperatively. Despite being a benign disease, mediastinal hydatid cysts should be removed surgically, as they may lead to serious complications due to deformation.
Keywords: Mediastinum, Hydatid cyst, Echinococcosis, Ekstrapulmonary

Öz
Hidatik kist, Echinococcus granulosus'un neden olduğu paraziter bir hastalıktır. Tüm kist hidatik vakalarının %0.5-2.6'sı mediastinal yerleşimlidir. Çalışmamızda primer mediastinal kist hidatikleri sunduk. Bütün hastalarda akciğer grafisi, toraks bilgisayarlı tomografi (BT) çekildi. Bir olguda spinal kordla ilişkisi nedeniyle toraks manyetik rezonans görüntüleme (MR) çekildi. Torakotomi ile çıkarılan bütün lezyonlar patolojik olarak doğrulandı. Postoperatif bir hastada dışında komplikasyon gelişmedi. Vertebra tutulumu olan bir olguda antihelmitik tedavi başlandı. Hiçbir olguda nüks gözlenmedi. Mediastinal kist hidatikler nadir görülmekle beraber diğer mediastinal hastalıklardan ayrımı zordur. Çoğu zaman cerrahi sırasında tanı konulur. Benign bir hastalık olmasına rağmen dektrüksiyona yol açarak ciddi komplikasyonlara neden olabileceğinden cerrahi olarak çıkarılmalıdır.
Anahtar kelimeler: Mediasten, Hidatik kist, Ekinokokkoz, Ektrapulmoner

Introduction

Hydatid cysts are a parasitic disease caused by the larvae of *Echinococcus granulosus*, and are endemic in some regions. Although very rare, intrathoracic extrapulmonary hydatid disease may occur in the pleural space, pleural fissure, chest wall, mediastinum, pericardium, myocardium and diaphragm. Of all hydatid cyst cases, 0.5 to 2.6% were in the mediastinum [1]. Here, we present the cases with primary mediastinal hydatid cysts we operated between 2009 and 2019 to contribute to the literature.

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Case presentation

We reviewed 254 patients operated on due to intrathoracic hydatid cysts between 2009 and 2019. Of 254 patients, four (1.6%) were mediastinal hydatid cyst. All patients underwent chest X-ray and computerized tomography (CT), while magnetic resonance imaging (MRI) was performed in only one patient to determine the relationship between neural foramen and the lesion. All cases were operated on, and the lesions were completely removed. Pathological examination revealed hydatid cysts. Of all patients, three were female and one was a male. Their ages ranged between 47-74 years, with a mean value of 59 years. Except for one, all patients were symptomatic. Two patients displayed coughing, while one had dyspnea and back pain. Radiologically, three of the hydatid cysts were located in anterior mediastinum, and one was posteriorly located. Thoracic CT revealed diaphragm paralysis in one of the cases with anteriorly located hydatid cysts (Figure 1, 2). Except for one of the hydatid cysts located in the anterior mediastinum, no distinction could be made from pericardial cysts. MRI was performed to differentiate from neurogenic tumors in the posteriorly located lesion. The defined mass extended into the neural foramen, and due to the destruction of the vertebral corpus and posterior components, the patient was radiologically pre-diagnosed with a neurogenic tumor and underwent surgery (Figure 3). Thoracotomy was performed to remove all lesions totally in all cases. The drains were drawn in a mean of 4 days postoperatively (ranging 2-5 days). Mean hospital stay was 5 days (ranging between 3-6 days). Postoperative complications developed in one patient (Table 1) in the form of diaphragmatic eventration, in whom diaphragm plication was performed six months later due to dyspnea. No recurrence was witnessed in the patients during the mean 88-month follow-up period (range: 42-126 months).

Table 1: General characteristic of the patients

Cases	Age	Gender	Location	Surgical procedure	Postoperative drain removal time	Postoperative complications
1	59	F	Anterior mediastinum	Thoracotomy excision	5th day	Diaphragm eventration
2	56	F	Anterior mediastinum	Thoracotomy excision	2nd day	No
3	74	F	Posterior mediastinum	Thoracotomy excision	2nd day	No
4	47	M	Anterior mediastinum	Thoracotomy excision + Diaphragm plication	5th day	No

F: Female, M: Male

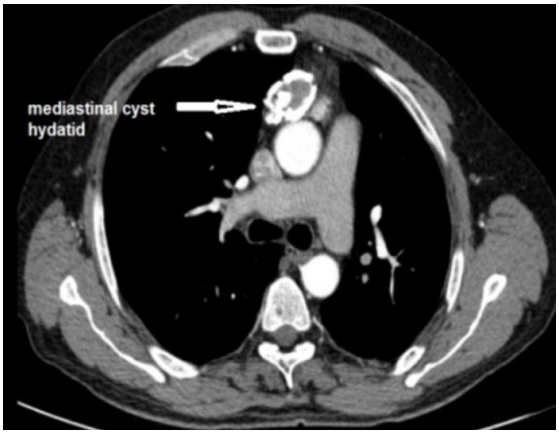

Figure 1: Thoracic Computed Tomography showing the lesion

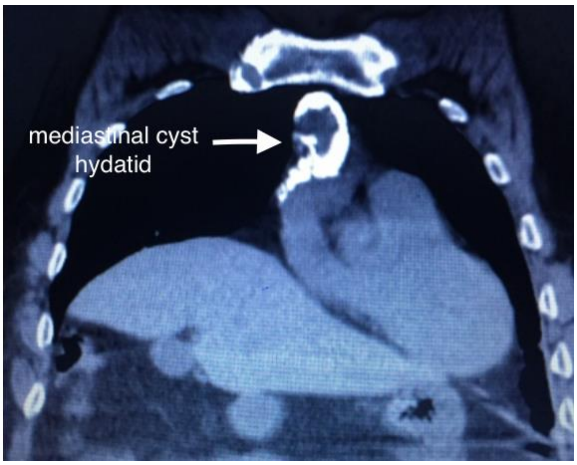

Figure 2: Eventration of diaphragm on the right

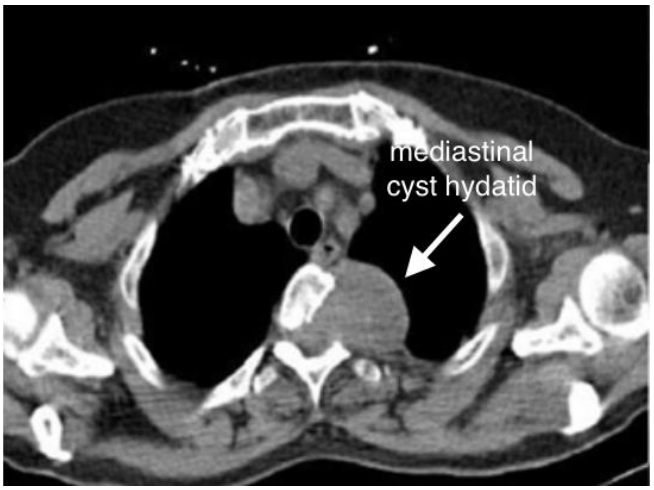

Figure 3: Vertebral invasion thorax Computed Tomography image

Discussion

Hydatid cysts are rarely observed in mediastinum. The parasite possibly migrates to the region through the arterial branches of the thoracic aorta or lymphatics after passing through the hepatic and pulmonary filters. The symptoms are caused by cyst size, location, mediastinal pressure, and degree of erosion. Among the symptoms commonly witnessed are chest pain, cough, dyspnea and recurrent nerve palsies [2]. As consistent with literature, cough, dyspnea and back pain were also observed in our study.

Today, many serological tests are recommended to diagnose hydatid cysts; however, most are not performed routinely in clinical practice due to their low diagnostic value, variable sensitivity, and specificity [3]. In our study, we also did not routinely use any serological tests.

In a study performed by Rakower and Milwidsky [4], primary mediastinal hydatid cysts were located most in posterior, anterior and middle mediastinum, respectively. In our study, however, hydatid cysts were more frequently located in anterior mediastinum.

In general, mediastinal hydatid cysts cannot be distinguished clinically and radiologically from other mediastinal cystic lesions. Radiological examinations, such as chest radiography, thoracic CT and MRI facilitate the diagnostic procedure. Although the diagnostic procedure involves taking all clinical, radiological, laboratory and historical data into consideration, the diagnosis of mediastinal hydatid cysts is usually made during the surgery [5]. In all our patients, chest X-

ray and thoracic CT were performed; in addition, MRI was performed to determine the association with spinal nerves in a patient with a posteriorly located cyst. Despite these procedures, all but one case was diagnosed during surgery in our study.

Mediastinal hydatid cysts may lead to complications such as rupture, fistulas, embolism, and pressure on vital organs [6-7]. Posteriorly located hydatid cysts may destruct the vertebrae, compress on the spinal nerve, and be confused with neurogenic tumors [8]. In our case with the posteriorly located cyst, the lesion extended into the neural foramen, causing a destruction of the vertebral corpus and posterior elements; therefore, the case was pre-diagnosed as a neurogenic tumor and operated. During the surgery, it was considered as a daughter vesicle and the case was diagnosed with a hydatid cyst. The lesion was totally excised with the simultaneous participation of neurosurgery. The diaphragmatic paralysis is an exceedingly rare entity in primary mediastinal hydatid cysts, and as its treatment modality, simultaneous diaphragmatic plication is also performed during surgery [9]. We detected diaphragmatic eventration in a case with an anteriorly located cyst but did not evaluate the diaphragmatic paralysis was due to the hydatid cyst because the location of the lesion was far from the phrenic nerve. We also performed the diaphragm plication simultaneously during the surgery. A variety of techniques have been described to treat hydatid cysts, such as percutaneous drainage and capitonnage. However, when the hydatid cysts are strongly associated with vital mediastinal structures, total excision may not be possible, and partial excision is achieved (9). During surgery, dissemination or seeding of the cysts, and anaphylactic reaction may occur [10]. In such cases, anti-helminthic drugs are administered in the postoperative period [9]. In our case, we administered the treatment of andazol for 12 months only to the case with vertebra dextration, and observed no recurrence during the follow-up.

Conclusion

Although mediastinal hydatid cysts are rarely encountered, it can be difficult to differentiate them from other mediastinal lesions, and the diagnosis can be made during the surgery. Despite being a benign disease, because they may cause serious complications due to destructions and pressure on the surrounding tissues, the mediastinal cysts must be operated and removed totally, if possible. In addition, long-term anthelmintic treatment should be started in patients with bone involvement.

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