Case Report

Mediastinal Hydatid Disease: An Unusual Presentation

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ABSTRACT

Hydatid disease is a significant health problem in endemic areas. While occurrence of the cysts in the liver and lung is common, mediastinal localisation is extremely rare. We report the case of a 35-year-old male who presented with a painless swelling on the right side of the neck and features of superior vena caval obstruction. Chest radiography and computed tomography (CT) suggested a cyst in the right upper lobe, extending into the right supraclavicular region as well as another cyst in the left lung. Thoracotomy revealed that the right-sided cyst was actually mediastinal in location and had herniated through the thoracic inlet compressing the superior vena cava (SVC). Both cysts were removed in two separate operations and symptoms of SVC compression subsided after removal of the right-sided cyst. Histopathology was consistent with a hydatid disease. **[Indian J Chest Dis Allied Sci 2010;52:245-247]**

Key words: Mediastinal hydatid cyst, Echinococcosis, Mediastinal cyst.

INTRODUCTION

Hydatid disease remains a serious health problem in endemic countries, like India. A rural setting with sheep, goat and cattle as intermediate hosts is an important risk factor for the disease. Hydatid cysts are usually located in the liver, lung and sometimes in other sites, such as the spleen, kidney, bone and brain. Mediastinal hydatid disease is distinctly uncommon,[1, 2] but important because it can compress vital structures.

We report here a rare presentation of mediastinal echinococcosis with herniation to the neck and symptomatic superior vena caval compression. Our search has revealed only three reported cases of cervico-mediastinal echinococcosis[3-5] in the available literature.

CASE REPORT

A 35-year-old non-diabetic, non-hypertensive farmer residing in a village in eastern India presented with a painless, gradually increasing swelling in the right supraclavicular area for six months. It was associated with slowly progressive breathlessness without wheezing or orthopnoea. For the last three months, he had developed facial puffiness with swollen right upper limb and dry cough. There was no history of haemoptysis, fever, voice change, dysphagia, weight loss or any other noticeable swelling elsewhere. The patient had smoked 12 to 15 *bidis* (unfiltered, rolled tobacco-leaves) daily for the last 12 years and had been in contact with dogs for the last 10 years.

On general examination, the patient had a firm, smooth, non-tender, non-pulsatile, non-compressible lump (2cm×3cm) in the right medial supraclavicular area (Figure 1) with engorged non-pulsatile neck veins and prominent veins over the right upper anterior

Figure 1. Photograph of the patient's neck showing the supraclavicular swelling.

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chest wall with flow from above downwards. There was a diffuse swelling of the right upper limb without any vascular or neurological abnormality. The vital signs were not remarkable; there was no lymphadenopathy or hepatosplenomegaly. Respiratory system examination revealed an impaired percussion note over the apex of the right lung, dullness over left 6^{th}, 7^{th}, 8^{th} intercostal spaces along the dorsal scapular line with diminished breath sounds. Other systems were normal.

Investigations showed raised neutrophil and eosinophil counts (71% and 7% respectively in a total leukocyte count of 10,000/cmm). Blood biochemistry was normal and sputum for acid-fast bacilli was negative. Chest radiograph (postero-anterior view) showed two homogeneous rounded opacities sharply circumscribed in the right upper and the left mid zones (Figure 2). Computed tomography of the chest (Figure 3) revealed a cystic opacity (5.1cm×4.0cm) in the right upper lobe extending into the supraclavicular region, compressing the SVC at its formation leading to prominence of vessels around it.

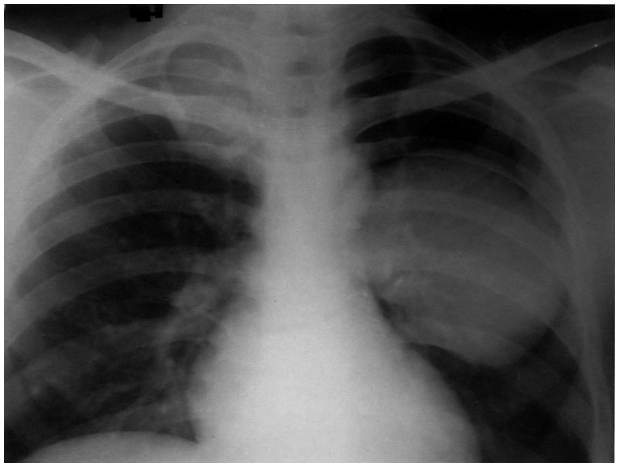

Figure 2. Chest radiograph (postero-anterior view) showing two homogeneous rounded opacities sharply circumscribed in the right upper and left mid zones.

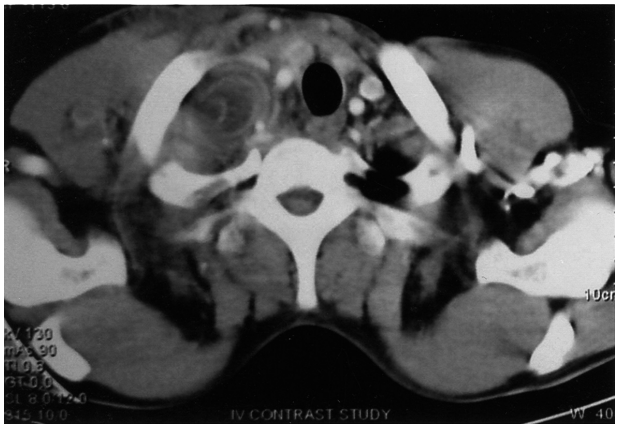

Figure 3. CT of the chest showing a cystic opacity in the right anterior-superior mediastinum.

There was another homogeneous, circumscribed opacity (8.41cm×6.2cm) in the left lung. No abnormality was seen on CT or ultrasound of liver.

After initiating albendazole presumptively, the patient was subjected to a right thoracotomy. Contrary to the CT findings, the right-sided cyst was actually seen to occupy the superior mediastinum in relation to but free from the right lung apex. It had herniated through thoracic inlet into right supraclavicular fossa and was adherent to and compressing the SVC. The intact cyst was deactivated with hypertonic saline solution; cystotomy was performed and the cyst was removed piece-meal owing to its adherence to SVC. The left-sided cyst was removed by pericystectomy three weeks later. The patient made an uneventful recovery with disappearance of features of SVC compression after the first operation. Histopathology was consistent with hydatid disease. Post-operatively, albendazole was continued for three months.

DISCUSSION

Hydatid cysts rarely present in the mediastinum posing difficulties in diagnosis. Eroglu *et al*[1] has reported 11 (2.6%) cases of primary mediastinal hydatid disease among 427 patients with thoracic hydatid disease in Turkey, where hydatid disease is common. Kabiri *et al*[6] in a retrospective review of 2332 cases of intrathoracic hydatid cysts found seven (0.3%) cases with mediastinal localisation. Of the mediastinal hydatid cysts reported in the literature, an overwhelming majority were in the posterior mediastinum.[7] The location of this cyst in the superior mediastinum is a rare clinical presentation.

Presenting symptoms and complications of mediastinal hydatid cysts depend on their location and size. Complications, such as mechanical compression of neighbouring structures, and rupture, leakage or infection of the cysts may result. Symptoms reported in the literature range from cough, dyspnoea, chest pain, dysphagia to Horner's syndrome, Pancoast's syndrome,[8] paraparesis,[9] though many cases are asymptomatic.[1,6] Rupture of the mediastinal hydatid cyst into the pleural cavity[10] or aorta,[11] and hydatid pulmonary embolism as a result of rupture into the inferior vena cava or heart have been reported,[12] but herniation into the neck is extremely uncommon.

Beji *et al*[3] reported a case of cervico-mediastinal hydatid disease presenting with a right-lower-neck mass. Alvarez *et al*[5] reported a case of primary mediastinal hydatid cyst presenting with a painful right supraclavicular mass, dyspnoea on exertion and dysphagia. Purohit *et al*[13] reported a case of primary mediastinal echinococcosis in a young female with Horner's syndrome and SVC obstruction, in whom the diagnosis was made on surgery.

Our patient presented with a right-lower-neck mass and features of SVC syndrome. Chest radiography suggested an intrathoracic cyst; ultrasonography and CT confirmed intrathoracic localisation and a cervical extension of the right-sided cyst; however, presence of the cyst in the left lung was an important supportive evidence. The localisation of the right-sided cyst in the anterior-superior mediastinum was actually demonstrated on thoracotomy and the final diagnosis was confirmed by histopathology. Although total pericystectomy is the operation of choice, owing to the intimate adherence to SVC, a small part of the cyst wall attached to the SVC was left in place. There was complete regression of features of SVC obstruction soon after the operation and the patient had no symptoms at six months follow-up.

In conclusion, primary hydatid cyst of the mediastinum with cervical herniation is a distinctly uncommon clinical entity which can present with compressive symptoms, such as superior vena caval obstruction at a young age, especially in endemic areas. Surgery is the treatment of choice and excision must be carried out without delay to avoid or relieve compression of the surrounding structures.

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