


Inverted intercostal hernia of elastofibroma dorsi mimicking well-differentiated liposarcoma in the chest wall

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Abstract

Elastofibroma dorsi is a well-known benign chest wall tumor. Herein, we present a case in which an elastofibroma protruded into the thoracic cavity, leading to inverted intercostal hernia. Imaging revealed a soft tissue mass containing fat, typical of elastofibroma dorsi; however, precise diagnosis was difficult owing to the location of this mass that protruded into the thoracic cavity. Liposarcoma had to be ruled out because it was a growing fat-containing mass. Considering that the tumor moved while the patient was undergoing computed tomography-guided biopsy in the prone position, a diagnosis of inverted intercostal hernia of elastofibroma dorsi was made. We report this case with a review of current literature.

Keywords

Inverted intercostal hernia, elastofibroma dorsi, liposarcoma, chest wall, MRI, CT

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Introduction

Intercostal hernia is a rare phenomenon that is defined as the protrusion of lung or abdominal viscera via an intercostal defect commonly associated with trauma or thoracic surgery^{1,2} (Figure 1(a)). By contrast, the protrusion of soft tissue into the thoracic cavity, that is, inverted intercostal hernia, is extremely rare^{3,4} (Figure 1(b)). Respiratory fluctuations and changes in the body position, including a transition to the supine position, displaces or invaginates the tumor into the intrathoracic cavity because of compression from its own weight. Large surgical wounds from thoracotomy before thoracoscopic surgery has become common increase the risk of incisional hernia.

Elastofibroma dorsi is a well-known benign chest wall tumor and is diagnosed when its characteristic imaging findings are noted.^{5,6} Herein, we present a case in which elastofibroma protruded into the thoracic cavity and caused inverted intercostal hernia. Case

report

A 74-year-old man was referred to the department of radiology for an incidental finding of an intrathoracic mass during a follow-up visit for other diseases (hepatocellular carcinoma and lung cancer with multiple metastases). The patient had a previous history (18 years ago) of thoracic surgery for lung cancer resection with unspecified details.

At the time of the current admission, no clinical symptoms indicative of an intrathoracic mass were

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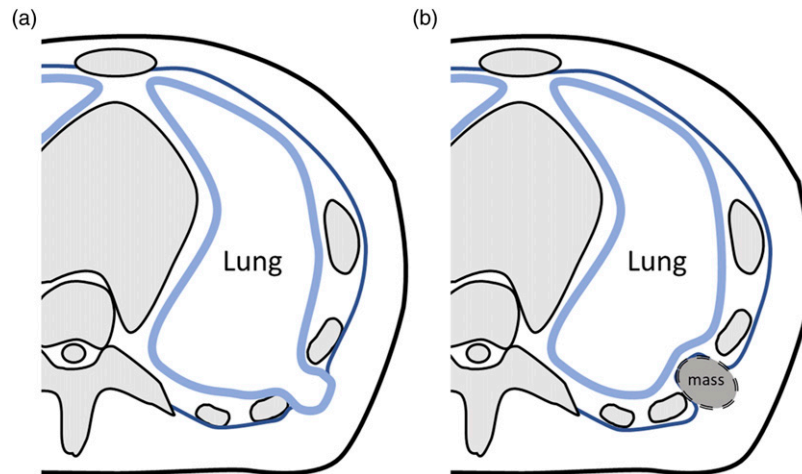


Figure 1. Schematic diagram of intercostal hernia and inverted intercostal hernia. (a). Intercostal hernia can be observed as the protrusion of lung via an intercostal defect. (b). Protrusion of soft tissue (mass) into the thoracic cavity, that is, inverted intercostal hernia.

observed. Physical examination revealed a 7-cm long incision scar from a previous surgery along the lower left rib on the left chest wall. Laboratory test results were within reference limits, except for elevated liver enzymes associated with chronic hepatitis.

Contrast-enhanced computed tomography (CT) revealed a heterogenous low-density mass containing fatty tissue in the lower left thoracic cavity; the mass protruded from the chest wall, that is, with an extra pleural sign. CT also showed a tumor with a diameter of approximately 40 mm (Figure 2(a)). The fat-containing hypovascular mass showed a slight degree of contrast enhancement in the equilibrium phase. No similar lesion was visible on the right side. CT performed 2 years ago showed that the same mass was small and located outside the rib cage (Figure 2(b)).

Magnetic resonance imaging (MRI) performed for other diseases showed that the mass was a smooth and well-defined tumor with a heterogenous high and low signal intensity on a T2-weighted image (Figure 3(a)). Diffusion-weighted images (Figure 3(b)) did not show any abnormal diffusion restriction, and the apparent diffusion coefficient (ADC) value was elevated (Figure 3(c)). The decrease in the signal intensity between the dual gradient-echo in-phase and opposed-phase MRI revealed the presence of internal fat content, including microscopic fat content (Figure 3(d) and (e)).

Percutaneous biopsy was performed to exclude liposarcoma. Percutaneous biopsy was performed under CT guidance with the patient in the prone position (Figure 4(a)); however, the mass moved out of the chest cavity and settled on the caudal side of the left scapula, compared with contrast-enhanced CT (supine position) (Figure 4(b)).

The pathological examination of the biopsy specimen showed increased collagen fibers, spindle-shaped mesenchymal cells, and adipose cells following hematoxylin and

eosin (HE) staining (Figure 5(a), HE $\times 10$). Furthermore, multiple elastic fibers were visible after Elastica van Gieson's (EVG) staining (Figure 5(b), EVG stain $\times 10$). Therefore, the mass was pathologically diagnosed as an elastofibroma.

Discussion

In the present case, imaging results showed the typical characteristics of elastofibroma dorsii. However, the diagnosis was difficult because it presented in the thoracic cavity and the past surgical history of lung surgery was initially unknown.

Elastofibroma is a well-known, slow-growing benign lesion that is typically found beneath the scapula with highly characteristic imaging results.^{5,6} It can typically present on imaging as a soft tissue mass containing a fat layer (7,8). MRI commonly shows a homogenous to heterogenous hypointensity on T1- and T2-weighted images, reflecting abundant collagenous fibrous tissue. Moreover, fatty tissue inside the mass shows up as high signal intensity on T1- and T2-weighted images.⁶⁻⁸ Diffusion-weighted images do not show abnormal diffusion restriction, and the ADC value is often reported to be elevated.⁶ After the administration of intravenous contrast, a slightly delayed contrast enhancement is observed.

Intercostal hernia is a rare phenomenon that is commonly associated with trauma or an inadequate closure of the chest wall following thoracic surgery.^{1,2} Seder et al.⁴ identified certain potential risk factors for chest wall hernia. This included a history of thoracotomy, obesity, chronic obstructive pulmonary disease, oral steroid use, and diabetes mellitus.⁴ Of these, the present case had a history of thoracotomy, chronic obstructive pulmonary disease, and diabetes mellitus.

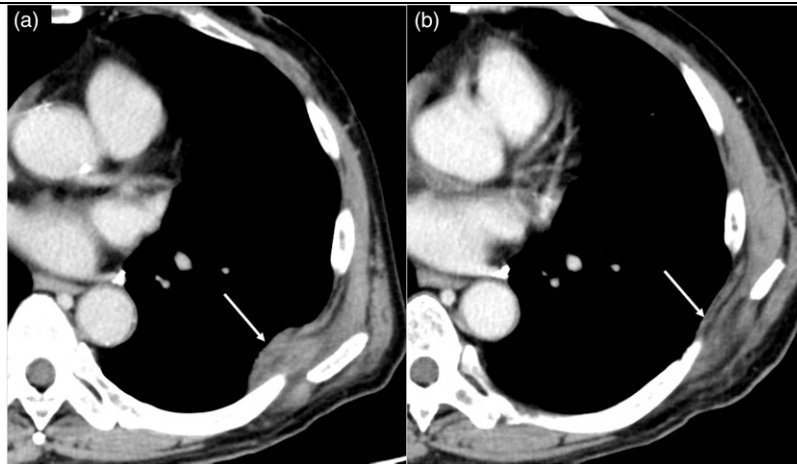


Figure 2. Contrast-enhanced computed tomography (CT). (a). Contrast-enhanced CT scan (equilibrium phase). (b). Contrast-enhanced CT scan (equilibrium phase) 2 years prior to the one performed in (a). Contrast-enhanced CT revealed a heterogenous low-density mass containing fatty deposition in the left posterior chest wall, that is, with an extra pleural sign. CT also revealed a tumor measuring approximately 40 mm at the maximum diameter (Figure 2(a)). The hypovascular mass containing fat showed a slight degree of contrast enhancement in the equilibrium phase. No similar lesion was visible on the right side. CT scan performed 2 years ago (Figure 2(b)) showed the same small mass located outside the rib cage.

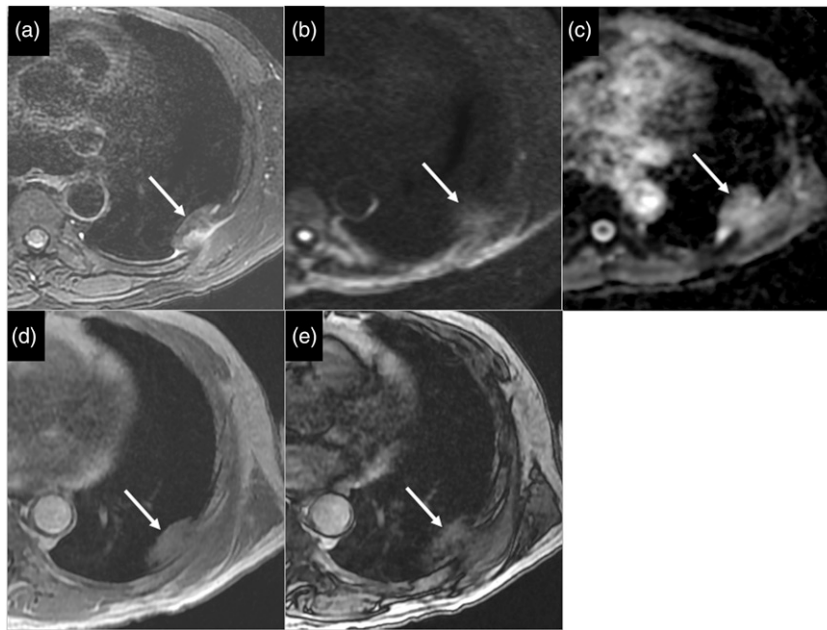


Figure 3. Magnetic resonance imaging (MRI). (a). Fat-suppression T2-weighted image. (b). Diffusion-weighted image (b-value, 800 s/mm²). (c). Apparent diffusion coefficient (ADC) map. (d). Chemical shift in-phase image. (e). Chemical shift opposed-phase image. The mass was a smooth, well-defined tumor with a heterogenous high and low signal intensity on fat-suppression T2-weighted imaging (Figure 3(a), arrow). Diffusion-weighted images (Figure 3(b)) did not show any abnormal diffusion restriction, and the ADC value was elevated (Figure 3(c)). The decrease in the signal intensity between the dual gradient-echo in-phase and opposed-phase MRI suggested internal fat content, including microscopic fat content (Figure 3(d) and (E), arrow).

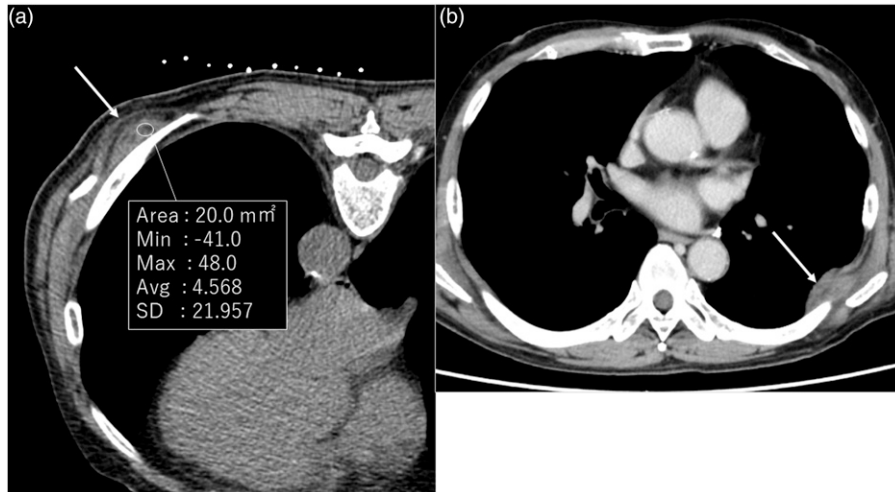


Figure 4. Computed tomography (CT) for percutaneous biopsy. CT (Figure 4(a)) for percutaneous biopsy in the prone position revealed the shifting of the mass to outside the rib cage, eventually settling on the caudal side of the left scapula (Figure 4(a)), compared with contrast-enhanced CT (supine position) (Figure 4(b)). The region of interest of the mass suggested fat component inside the tumor as the CT value included a negative component.

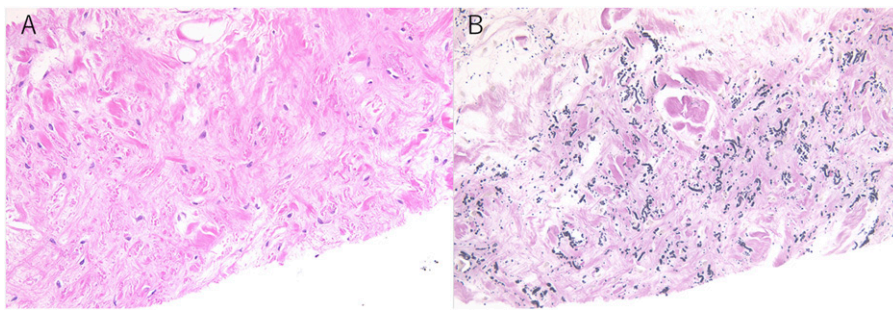


Figure 5. Pathological examination of the biopsy specimen. (a). Hematoxylin and eosin stain (HE, × 10). (b). Elastica van Gieson's stain (EVG, × 10). The pathological examination of the biopsy specimen revealed increased collagen fibers, spindle-shaped mesenchymal cells, and adipose cells on HE staining (Figure 5(a), HE × 10). Multiple elastic fibers were dyed black with EVG staining (Figure 5(b), EVG stain × 10).

Inverted intercostal hernias are, however, extremely rare. We believed that the inverted intercostal hernia resulted from the forcing of elastofibroma into the thoracic cavity from a fragile area of the left posterior chest wall because of the pressure of the body's weight while it was in the supine position. By contrast, the pressure on the body's weight was released during CT-guided biopsy as the body was in the prone position, resulting in the movement of the mass to the more typical area at the subscapular chest wall.

In general, elastofibroma dorsi is not eligible for treatment unless certain symptoms, such as pain and discomfort, are present. Biopsy is considered when it is necessary to distinguish it from malignant tumors, such as liposarcoma, if it presents at an unusual site.

In conclusion, we reported a rare case of inverted intercostal hernia of elastofibroma dorsi mimicking well-differentiated

liposarcoma in the chest wall. The diagnosis of inverted intercostal hernia of elastofibroma dorsi is not difficult if CT and MRI show typical imaging findings and location as well as if the mass's mobility is confirmed.

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Declaration of conflicting interests

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Ethical approval

All procedures performed in this study involving human participants were conducted in accordance with the ethical standards of the institutional and national research committees and the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards where appropriate.

Informed consent

This case report has obtained Institutional Review Board approval and the formal informed consent from this patient was waived. We had obtained informed patient consent about all procedures such as CT, MRI, and CT-guided biopsy.

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