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Case Report

Surgical treatment of an intrathoracic rib causing cough in a young woman

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ABSTRACT

Intrathoracic rib is mostly asymptomatic and one of the rare congenital anomalies of thoracic wall. Keeping in mind its appearance and types on chest x-rays can prevent unnecessary examinations and invasive procedures. Locations and relationship to adjacent tissues of intrathoracic ribs determine its clinical course and surgical decision.

Keywords: accessory rib, anatomical variations, intrathoracic rib, rib anomalies, supernumerary rib

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Introduction

The term intrathoracic rib refers to the abnormal position of a rib within the thoracic cavity. It was first described by Lutz in 1947. Later, it was also used with the definition of accessory rib or supernumerary rib [1]. Although the etiology is not clear, genetic factors and chemical effects in the fetal period have been held responsible [2]. This rare congenital abnormality mostly has no effect on respiratory functions and usually detected incidentally by imaging studies. We report a case of supernumerary intrathoracic rib (SIR) located posterior to the abnormally developed T6 vertebrae in an 18-year old female who presented with dyspnea and back pain.

Case Report

An eighteen year-old female patient admitted to our clinic with the complaint of back pain, cough and dyspnea for several years. It was understood that her complaints were unrelated to effort and intensified especially in the evening. She has celiac disease. Vital signs were normal and stable. Oxygen saturation was %98 in room air. There was no history of operation, chest trauma or thoracic disease. There were no pathological findings in the physical examination findings and auscultation of respiratory sounds. Laboratory blood test results are within the normal range. On the conventional chest radiography, an opacity extending from the thoracic vertebra to the diaphragm was observed behind the cardiac shadow. For further examination, CT scan of thorax was performed, 3D and multiplanar reconstruction images were created (Figure 1a). These images revealed an abnormal osseous structure originating from the left lateral corpus of the T6 vertebrae which was protruded infero-laterally into the thoracic cavity (Figures 1b-1d). There was no intraparanchimal lesion. No other skeletal alteration was observed. We performed a postero-lateral muscle sparing thoracotomy incision The accessory rib had no articulation with vertebrae, covered by the parietal pleura and extended towards the diaphragmatic crus (Figure 2). There were no adhesions with the lung parenchyma. The accessory rib was resected completely by cutting from the vertebral corpus using a rib cutter. It was observed that SIR did not have its own intercostal muscle, artery, vein or nerve. The symptoms regressed after the operation, there was only pain at the incision site. There was no air leakage in the chest tube after the operation. The patient did not describe dyspnea, cough and back pain during the hospitalization period and was discharged on the postoperative day 2. There were no complications during the one year follow up. Written informed consent was obtained from the patient for publication of her data.

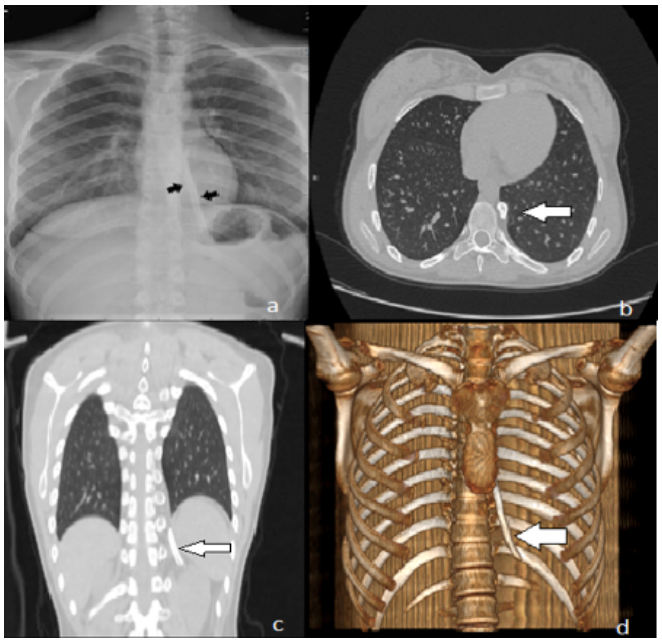

Figure 1. Posteroanterior chest graph shows an accessory rib (arrows) located on the left lateral side of corpus of T6 vertebrae **(a)**, lung window CT image shows an abnormal thoracic rib (arrow) **(b)**, coronal window CT image shows an abnormal thoracic rib (arrow) **(c),** three-dimensional CT recontruction image of the bone shows an abnormal supernumerary inthrathoracic rib (arrow) **(d).**

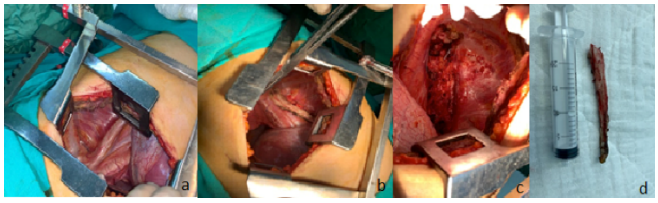

Figure 2. İntraoperative view of the accessory rib **(a)**, releasing the accessory rib from surrounding tissues by stripping the overlying pleura **(b),** view of the thoracic wall after separation of the accessory rib from the vertebral body with a rib cutter **(c)**, image of the accessory rib that has excised **(d).**

Discussion

Intrathoracic rib is a congenital anomaly that occurs with abnormal fusion of sclerotomes in embryological life [1,2]. When more than fifty cases in the literature are analyzed, it has been determined that it is more common on the right hemithorax without gender dominance. Most of the cases were diagnosed in pediatric age. SIR is mostly single and unilateral. Its location and relationship to adjacent tissues determines its clinical course, but it is usually asymptomatic. SIR is usually detected incidentally by imaging studies or in autopsy series. The most common symptoms are chest pain, hemoptysis, and dyspnea [1,2]. Etiological factors of accessory rib is

not clear. It is suggested that it is due to the incomplete sclerotome fusion that caused by the change in intrathoracic pressure during embryologic development. Alterations of Myf5/Myf6 expression, the Pax-1 gene, and the Hox gene family have been implicated [3].The intrathoracic rib is important in differential diagnosis. In the differential diagnosis intrathoracic rib, accessory lobe, asbestos plaques and calcified sequestration should be kept in mind. This unusual opacity detected on chest X-ray can cause concern and unnecessary further examination. Therefore, the benign course in asymptomatic cases prevents over-diagnose and over-treatment. Being aware of its benign course prevents unnecessary examinations and operations. In 2006, Kamaro et al classified accessory ribs according to their origin [1]. Accordingly, Type 1 refers to the supernumarary intrathoracic rib that is articulated from the vertebral corpus or its proximal rib. Type 2 bifid intrathoracic rib. In type 3, the rib is locally compressed into the cavity. In 2021, Xue et al proposed a new classification with five distinct categories that include all anomalies not included in the previous classification (Table 1) [2].

Table 1. The classification of intrathoracic rib [2].

Type 1	Separated from the vertebral corpus by a joint
1A	Originating from the lateral side of the vertebral corpus (right or left)
2B	Originating from the anterior of the vertebral corpus (unilateral or bilateral)
Type 2	Originating from the proximal rib
Type 3	Bifid or forked intrathoracic rib
Type 4	Ribs locally compressed into the cavity
Type 5	Floating intrathoracic rib (without any rib or osseous connection to the vertebrae)

According to this current and useful classification, our case is in category type 1-A. Sometimes this intrathoracic rib can be attached to the diaphragm with a fibrotic band or fatty tissue. Intrathoracic rib is mostly common with scoliosis [2]. Intrathoracic rib can be easily overlooked, especially if the image is not taken under optimal conditions in the chest X-ray. It can be hidden in cardiac shadow or hidden behind the clavicle, especially at the upper levels. Barreiro et al. analyzed the tomography images of 650 patients and found the incidence of accessory thoracic ribs to be 0.15% [4].CT scan is a steady method to show intrathoracic ribs. Also, with CT, parenchymal diseases and diaphragmatic extensions can be observed and relations with neighboring organs can be determined. In our case, three-dimensional reconstructions and multiplanar CT were very supportive for a successful operation. The newly defined contrast-enhanced ultrasound (CEUS) also provides reliable information (in) for the identification of masses in the subpleural area [5]. Chronic cough is an indication for bronchoscopy. However, we did not perform bronchoscopy because tracheobronchial system pathologies was not considered in our patient. The cough complaint vanished in the patient who completed the recovery period after the surgery. This may suggest that the supernumerary rib is the cause of the irritation. It is necessary to evaluate thoroughly before deciding on surgery. All possible complications of the intrathoracic rib according to localization and the patient's symptoms and possible complications of surgery should be well evaluated in the decision of surgery. The doctor's decision should be shared with the patient and the final decision should be made with the patient's participation. It has been suggested in the past that surgery does not improve respiratory functions and may have additional secondary negative effects [5]. However, in the study of Coyan et al the symptoms disappeared after surgery, as in our case [6]. We think that asymptomatic patients may become symptomatic or the existing mild symptoms may be exacerbated, especially in physiological conditions such as pregnancy that may cause elevation of the diaphragm.

In conclusion, the intrathoracic rib should be kept in mind in the differential diagnosis of opacities in chest X-rays. Although it is a benign anomaly, localization and the symptoms of SIR determines decision of the rib resection.

Declaration of conflicting interests

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Authors' contributions

DK,MŞİ; conceived and designed the study, HI,MŞİ; reviewed and recorded the data, DK,MŞİ; wrote the paper, SG,ES; reviewed and edited the manuscript. All authors read and approved the manuscript.

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