Radiology Case Reports 16 (2021) 710–713

Available online at www.sciencedirect.com

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Case Report

Bronchial carcinoid with bronchocele masquerading as Scimitar syndrome on chest radiograph ☆

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ARTICLE INFO

Article history:
Received 25 December 2020
Revised 3 January 2021
Accepted 3 January 2021

Keywords:
Bronchocele
Bronchial carcinoid
Scimitar syndrome

ABSTRACT

Bronchial carcinoid tumors occur predominantly in the perihilar location and can be asymptomatic. They may present in early stages with only obstructive features such as mucus plugging of airways resulting in bronchocele formation. We report the case of a 44-year lady with no chest complaints, who underwent chest radiograph for a routine preanesthetic checkup. A vertically oriented, tubular, soft tissue density lesion was observed in the right lower lung, which mimicked a Scimitar vein. Scimitar syndrome is a congenital disorder in which an anomalous vein drains the middle and lower lobes of right lung and enters into the IVC most commonly. It may present asymptomatically in adults and on chest radiograph appears as a vertical tubular opacity paralleling the right cardiac border. However, CT angiography revealed the lung lesion to be a bronchocele, distal to a central intensely enhancing spherical mass, completely occluding the right lower lobe bronchus. This perihilar mass had been missed on the chest radiograph. Bronchoscopic biopsy revealed a carcinoid tumor. As the patient was asymptomatic, she refused surgery in the ongoing COVID-19 pandemic.

Introduction

Bronchoceles are formed due to mucus impaction of lung airways commonly as a consequence of airway obstruction. Common causes are bronchiectasis due to cystic fibrosis or allergic bronchopulmonary aspergillosis. Bronchoceles may also be seen distal to a central airway obstruction, such as due to a foreign body, benign strictures, bronchial atresia or neoplasms such as bronchial carcinoid [1]. A lobar or segmental bronchus is the common site of origin of 80% carcinoid tumors, which are central and perihilar in location. They may be asymptomatic and in the early stages may present with only

☆ Competing Interests: The authors have declared that no competing interests exist.

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https://doi.org/10.1016/j.radcr.2021.01.013

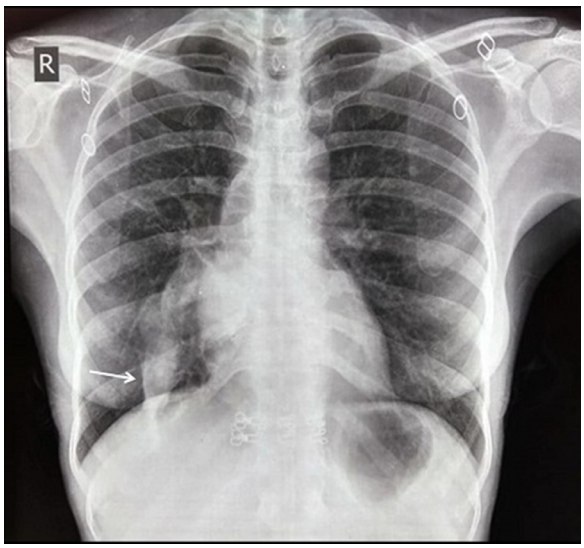

Fig. 1 – Chest radiograph showing a tubular, vertically oriented radio-opacity (arrow) in the paracardiac region in the right lower lung, separate from the heart border.

obstructive features such as mucus plugging and bronchocele formation [2]. Chest radiograph showing a bronchocele mimicking a "Scimitar vein" (which can be a congenital asymptomatic disorder presenting in adults) distal to a central carcinoid tumor being overlooked, has not been reported till date.

Case report

A 44-year lady presented with abdominal pain for 5 days. She gave a history of pulmonary tuberculosis 4 years back for which she had completed anti-tubercular therapy; and presently had no chest complaints. She was a non-smoker and had no history of diabetes or hypertension. Vital signs were normal. Chronic cholecystitis with cholelithiasis was diagnosed on ultrasound and an elective cholecystectomy was planned. A chest radiograph done for pre-anesthetic work-up revealed an elongated, vertically oriented radio-opacity, separate from the heart border in the right paracardiac region of lung, extending from the hilum to just below the diaphragm (Fig. 1). Differential diagnosis of bronchocele, pulmonary arteriovenous malformation and Scimitar vein were entertained.

CT angiography of the chest demonstrated a well-defined, rounded,4.5 × 5.3 × 4.7 cm, homogenous, infrahilar soft tissue density mass in the apical segment of right lower lobe, showing calcification within (Fig. 2). It had a nodular endobronchial component completely occluding the right lower lobe bronchus. Retrospectively, a retrocardiac focal mass-like density was identified on the right side, in the chest radiograph. Hypertrophied bronchial arteries arising from the descending thoracic aorta were supplying the mass which showed intense, heterogenous enhancement (Fig. 3). On CT distal to the mass was a lobulated, well marginated, vertically oriented, fluid density structure suggestive of a bronchocele

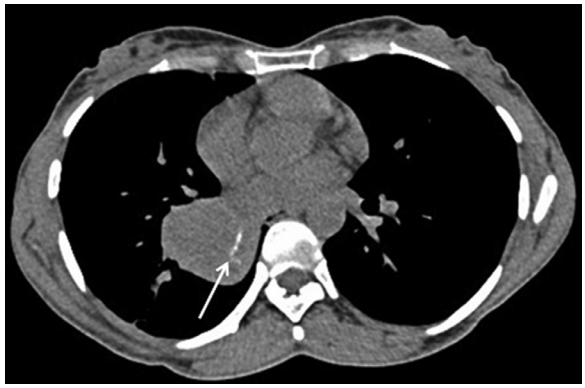

Fig. 2 – Noncontrast axial CT chest showing a central right lung mass with eccentric calcification (arrow).

extending up to the dome of diaphragm. It did not show opacification with contrast, thus excluding an arteriovenous malformation or Scimitar vein. The right lower lobe showed irregular bronchiectatic cysts with air trapping. Fibrobronchiectatic changes were present in the right upper lobe and apical segment of right lower lobe (Fig. 4). A provisional CT diagnosis of bronchial carcinoid with a distal bronchocoele was made; with sequelae of pulmonary tuberculosis in right lung.

Bronchoscopy revealed an endobronchial growth obstructing the right lower lobe bronchus. It was highly vascular and bleeding on biopsy was controlled by adrenaline and cold saline. Microscopy showed a tumor of small cells, with moderate pink to clear cytoplasm and round nuclei around a blood vessel. No mitotic figures were seen. A group of spindle cells with some mucinous intercellular material were also seen. Findings were consistent with bronchial carcinoid. Since the patient is asymptomatic for the last 6 months, she has not undergone surgery due to the ongoing Covid-19 pandemic.

Discussion

Bronchial carcinoid tumors are indolent malignant tumors of variable histological features varying from low to high grade tumor arising from Kulchitsky cells of bronchial epithelium [3]. The age of presentation is usually between third and seventh decades. Clinical presentation depends on its location within the lung. Peripheral tumors are usually asymptomatic and detected incidentally [3]. Central tumors usually present with symptoms of airway obstruction such as breathlessness, wheezing, recurrent pulmonary infections, fever and hemoptysis. They may be asymptomatic in about 30%-40%, as in our case. Kulkarni et al had reported a central left lung carcinoid detected incidentally in a road traffic accident patient who had no pulmonary symptoms [4].

On plain radiography, a round to oval hilar mass may be seen with features of bronchial obstruction. Sometimes a peripheral nodular opacity is seen. Some lesions may have lobulated margins and eccentric calcifications. Due to their central location, only findings related to bronchial obstruction may be seen on radiography initially, which include

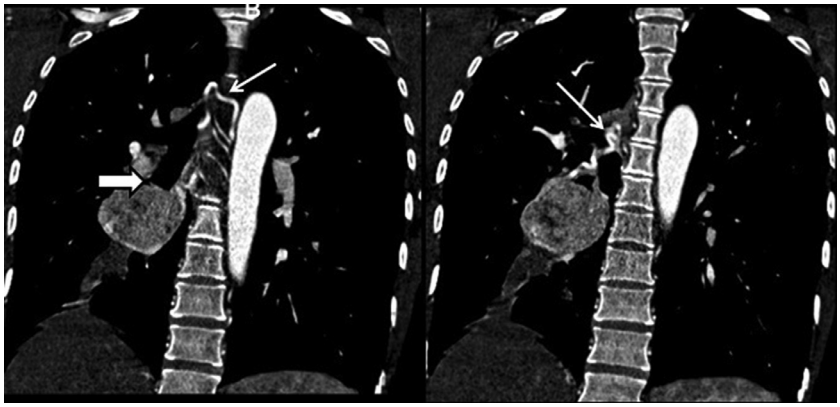

Fig. 3 – – Contrast-enhanced coronal CT Chest showing the enhancing mass occluding the right lower lobe bronchus (block arrow) and supplied by hypertrophied bronchial arteries (thin arrow). A distal bronchocele of fluid density identified, which appeared as a "scimitar"on the chest radiograph.

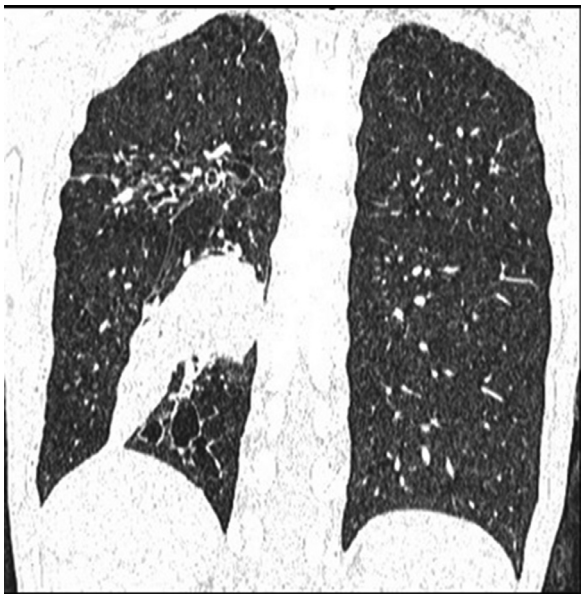

Fig. 4 – Coronal CT Chest lung window shows post obstructive bronchiectatic cysts in the right lower lobe and sequelae of pulmonary tuberculosis in right upper lobe.

bronchocele, bronchiectasis and air trapping as seen in this case. On chest radiography, bronchocele is seen as a well marginated elliptical or branching radio-opacity. In our case, mucus retention distal to the tumor resulted in a vertically oriented bronchocele in the right lower lobe, with a narrow tail visible below the dome of diaphragm. This radiographic appearance of a bronchocele in the right lower lobe (associated with a bronchial carcinoid) mimicking Scimitar vein, has not been described in literature.

Scimitar syndrome is a rare congenital anomaly characterized by partial anomalous pulmonary venous connection (PAPVC) and ipsilateral lung hypoplasia, almost exclusively on the right side. The pulmonary venous supply from the right lung particularly the middle and lower lobes drains via an anomalous vein into the systemic circulation (most commonly inferior vena cava), instead of left atrium [5]. It is classified into 2 forms: infantile and adult. Adults forms are usually asymptomatic [5]. On plain radiography, the anomalous vein is seen as a vertical tubular opacity paralleling AV the right cardiac border and directed towards the right cardiophrenic angle. Associated findings such as ipsilateral volume loss, mediastinal shift, dextrocardia and small hilum (due to hypoplastic ipsilateral pulmonary artery) may be seen [6], none of which were seen in our case. On CT angiography, it was confirmed that the "scimitar" was actually a bronchocele distal to bronchial carcinoid as it showed no contrast opacification.

CT is useful in the diagnosis of bronchoceles and also in finding the underlying cause. Bronchoceles are seen as non-enhancing, fluid attenuation structures, especially in the lower lobes. Contrast enhanced CT can easily differentiate between the obstructing tumor and distal bronchoceles except in cases of occult small lesions, where nuclear imaging may be useful.

On contrast enhanced CT chest, carcinoid tumors are seen as well-defined, round to oval hyperenhancing nodules, which may be lobulated. Eccentric calcifications are characteristically seen on non-contrast scan. They show intense contrast enhancement due to their high vascularity. Bronchial carcinoid tumors may sometimes be confused with pulmonary varix or pulmonary artery aneurysm due to this intense enhancement. Trachea and main bronchi are supplied predominantly via bronchial artery [7]. In our case, hypertrophied bronchial arteries were noted supplying the carcinoid.

Carcinoids have a characteristic bronchoscopic appearance – endobronchial polypoidal/ nodular lesion with smooth surface and cherry red colour. The histologic diagnosis can be made with bronchoscopic biopsy, with an increased tendency to bleed [2], as was observed in our case.

Surgical resection is considered the gold standard for treatment of bronchial carcinoid. However, in selected cases for example, where the patients' condition does not allow resection or in lesions that are entirely endobronchial, transbronchial resection may be an alternative [8]. Bronchial artery emboliza-

tion prior to transbronchial resection may be a feasible option to reduce bleeding [9].

Conclusion

In an asymptomatic patient, a central bronchial carcinoid can rarely present on the chest radiograph as a bronchocoele on the right side, mimicking Scimitar syndrome. This appearance warrants further evaluation using a contrast CT, so that the underlying tumor is not missed and the patient can be managed appropriately.

Authors' Contributions

The manuscript has been read and approved by all the authors, the requirements for authorship have been met, and each author believes that the manuscript represents honest work.

Patient Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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