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Squamous carcinoma arising from a true tracheal bronchus: Management and case report

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ABSTRACT

A case of a squamous carcinoma arising in a "True" tracheal bronchus is described. The presentation and management of this case is discussed.

1. Introduction

Tracheal bronchus is a rare congenital anomaly with a published incidence of between 0.1% and 3% first described by Sandifort in 1785 and includes a variety of bronchial anomalies arising from the trachea and main bronchus supplying the upper lobes [1,4]. Malignant neoplasms arising in these bronchi are rare and may pose a diagnostic and management dilemma [3,6].

2 Presentation of case

A 56 year old male presented with a history of a dry cough, shortness of breath and a headache, he had no previous illness. Clinically he appeared well with a normal pulse rate and blood pressure. The jugular venous pressure (JVP) was markedly elevated and on auscultation of the chest there was decreased air entry on the right side. The chest X-ray showed a large right sided intra-thoracic mass (Fig. 1). His white cell count, erythrocyte sedimentation rate and haemoglobin were completely normal. A computerized tomogram (CT) of the chest showed a large mass arising from an anomalous tracheal bronchus with compression of the right main bronchus and obliteration of the right middle lobe bronchus. There was compression of the mediastinum with no lymphadenopathy (Fig. 2). A bronchoscopy was performed confirming a tracheal bronchus arising above the carina on the right side of the trachea. Endoscopic cytology and bronchial brushings were negative. Echocardiography was used to exclude thrombi in the heart as a result of SVC occlusion. The pre-operative staging was T3M0N0.

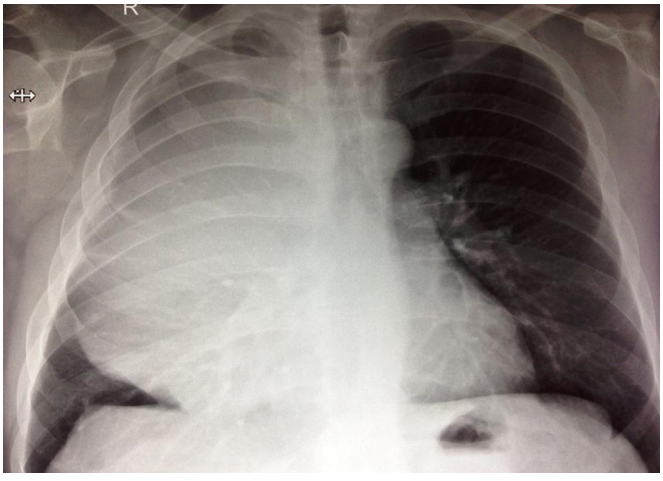

Fig. 1. X-ray chest showing a large intrathoracic mass.

A right thoracotomy was performed and a large mass was encountered occupying most of the intra-thoracic space with compression of the lung (Fig. 3). Access to the hilum was gained by retracting the mass posteriorly. It was intimately involved with the mediastinum, overlying the superior vena cava (SVC) which could not be visualised.

The pericardium was opened anteriorly to gain access to the SVC and blood supply. As this mass had involved and occluded the SVC, a clamp was applied at the atrial level and the SVC transected. The mass was then lifted superiorly and the SVC clamped at the confluence of the two brachiocephalic vessels. This allowed the main

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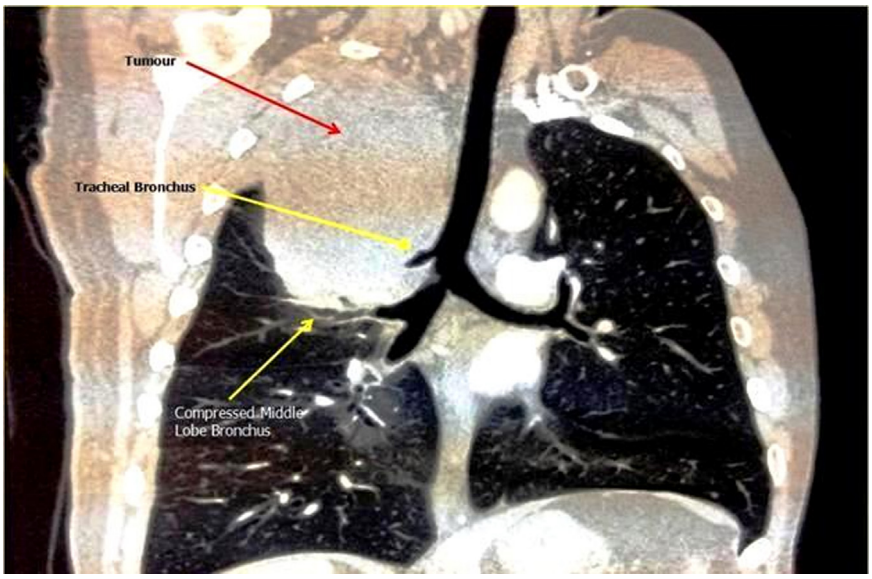

Fig. 2. CT-chest showing a large right upper lobe soft tissue mass arising from a tracheal bronchus compressing the middle lobe bronchus and mediastinum.

pulmonary artery trunk to be exposed and the pulmonary vessels to the upper lobe identified and transected. The superior pulmonary vein supplying the right upper lobe was oversewn and transected intra-pericardial. The abnormal bronchus which supplied the right upper lobe could then be transected close to the trachea and the entire mass with the upper lobe removed and sent for histology. The bronchial stump was closed with interrupted 2/0 Vicryl(Ethicon) sutures. An 18 mm Dacron was sewn into position with 4/0 prolene replacing the SVC and re-establishing flow. The histology confirmed a poorly differentiated squamous carcinoma arising from the tracheal bronchus. The resected SVC was encased in tissue accounting for its occlusion but was not invaded by the tumour. No spread to any lymph glands were documented and all resection lines were clear. The tumour had extensive necrosis present within it (Fig. 4).

The patient has been followed up for 18 months and is asymptomatic with no evidence of tumour recurrence.

3. Discussion

The embryological development of the tracheobronchial tree begins at twenty four days as a ventral bud in the laryngo-tracheal groove. At twenty eight days the lung buds start evolving and continue to elongate developing the bronchial tree. It is thought that the anomalous bronchi develop around this stage although, the precise mechanism is controversial. There is a spectrum of anomalous bronchi and in a large series, 56% arose from the trachea, 4.8% from the carina and 38% from the bronchial tree [1,3]. They are seven times more common on the right side and are further classified as displaced when a branch of the upper lobe is missing or supernumerary when the upper lobe has its normal anatomical branching. A true tracheal bronchus known as a “pig bronchus”, is where the right upper lobe bronchus is absent and is “replaced” by the abnormal bronchus (Fig.5). It may be associated with other congenital anomalies and the importance of knowing and understanding this anomaly is for diagnostic and therapeutic purposes especially in fields of chest surgery, radiology and anaesthetic management [1–3]. The majority of patients with anomalous bronchi are thought to remain asymptomatic but numerous reports have been published with complications such as recurrent infections, bronchiectasis, haemoptysis, malignancy, chronic cough as well as acute respiratory distress and stridor in the paediatric population [7]. Reports of malignant tumours arising from tracheal bronchi

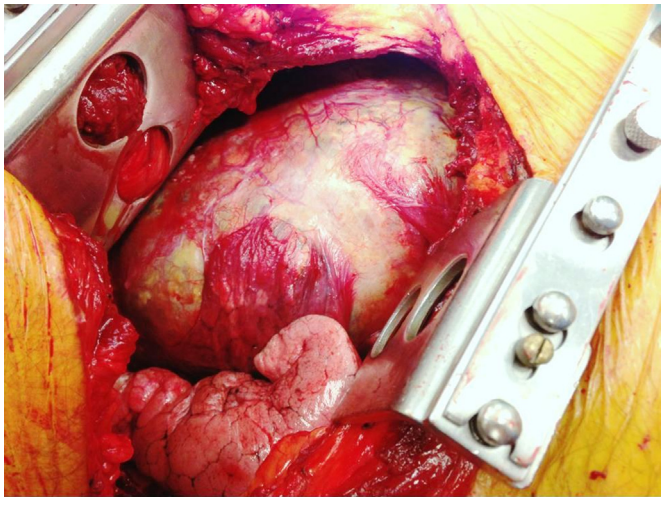

Fig. 3. Operative view of the tumour.

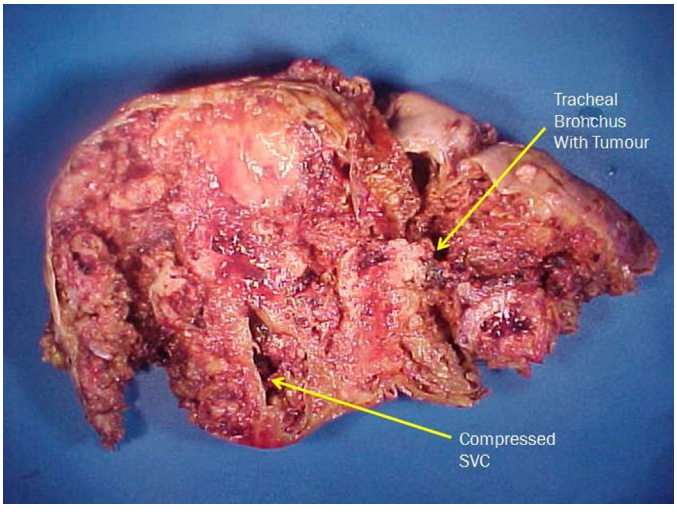

Fig. 4. Surgical specimen.

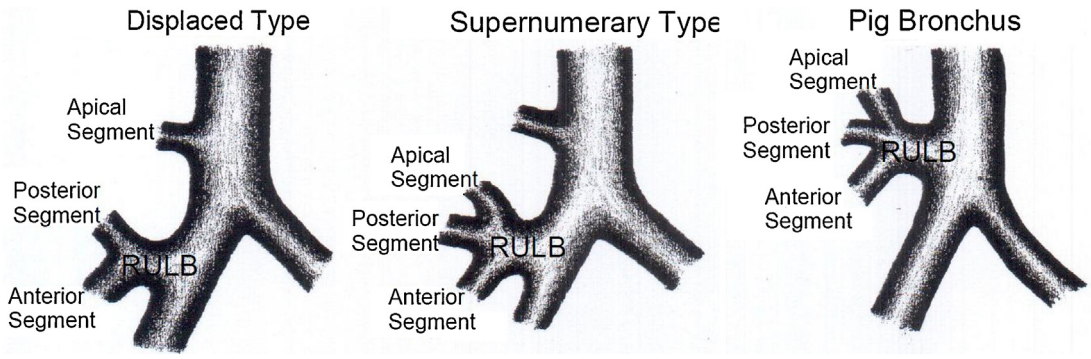

Fig. 5. Schematic diagram showing the different types of tracheal bronchi (RULB-right upper lobe bronchus).

have been published and this patient appears to be the third documented case of a squamous carcinoma arising in a "True" tracheal bronchus [4–6]. Management of these cases is surgical removal and pre-operative evaluation is critical in assessing operability. Knowledge of abnormal anatomy as well as anomalous vascular supply is important.

The complications following SVC replacement should also be stressed which may include stenotic anastomotic lines, kinking, thrombosis and or infection which is a serious complication. In cases of thrombosis, conservative management may result in a good outcome. Infection on the other hand may necessitate removal of the graft. Post-operative kinking may require immediate re-exploration. Contraindications to SVC replacement is essentially unresectable tumours that have involved the SVC. In our case there was no necessity for a bypass graft as the SVC was already occluded. In non-occlusive cases clamping of the SVC may result in haemodynamic changes which may include hypotension, cerebral oedema, intra-cranial bleed and transient cerebral dysfunction [8,9].

4. Conclusion

Malignant change in abnormal bronchi have been reported in the literature from time to time but squamous tumours appear to be very rare. To our knowledge, presentation with such a large tumour and a SVC syndrome has never been previously reported. This case presented a technical challenge due to the tumour's size and position. Although these anomalous bronchi are rare they may result in major difficulties if undiagnosed pre-operatively especially when using double lumen endotracheal tubes in thoracic surgery. If found incidentally patients should be informed of this anomaly in case there is need for future surgical management.

Conflict of interest

None.

Sources of funding

None.

Ethical approval

Patient consent – Yes.

Authors contribution

Dr N. Nicolaou: Participated in the conceptions and design of the paper. Drafted the manuscript and did literature review. Was the surgeon who performed the procedure.

Dr A. Du Plessis: Was the diagonistic radiologist and participated in drafting the Manuscript.

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