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Localized tracheopathia osteoplastica of the subglottis

A. R. BIRZGALIS, F.R.C.S., W. T. FARRINGTON, F.R.C.S., L. O'KEEFE, F.R.C.S., J. SHAW, F.F.A., R.C.S.Eng*

Abstract
Tracheopathia osteoplastica is a benign dysplasia of the tracheobronchial tree which rarely presents clinical problems especially when localized. We present a case which caused an unexpected difficulty in intubation and was subsequently removed endoscopically using the CO_2 laser.

Key words: Tracheal diseases, tracheopathia osteoplastica; Intubation, intratracheal, complications

Introduction

Tracheopathia osteoplastica is a benign progressive dysplasia of the tracheobronchial tree which is characterized by the presence of cartilagenous and bony nodules. The condition, first described by Wilks in 1857 (Martin, 1974), is usually asyptomatic but may cause increasing dyspnoea, haemoptysis and a dry cough. Incidental autopsy findings and improved bronchoscopy techniques have shown the condition to be commoner than once thought (Way, 1967). Most cases show a florid, diffuse involvement of the lower trachea and major bronchi with long stenosing segments. Localized involvement is unlikely to present clinical problems and is rarely reported.

A case of localized tracheopathia osteoplastica of the subglottis is presented which was diagnosed and treated endoscopically following a difficult endotracheal intubation.

Case report

A 57-year-old man was admitted for coronary artery by-pass surgery following a three-year history of increasing angina and angiographic evidence of three-vessel disease. There were no symptoms or signs of pulmonary disease and no relevant past otolaryngological history.

Following anaesthetic induction, conventional endotracheal intubation was found to be impossible due to subglottic obstruction from a hard, non-ulcerating lesion occupying the posterior two-thirds of the tracheal lumen. Via the rigid bronchoscope, a bougie was passed anterior to the lesion, over which a size 4.0 microlaryngoscopy tube was guided. Although the subsequent cardiac procedure was uneventful, extubation was felt to be unsafe without endotracheal surgery or tracheostomy.

The patient was returned to theatre the following day and formal rigid bronchoscopy performed. A hard ridge of tissue was found with normal overlying mucosa. It appeared to project from a narrow base on the posterior tracheal wall 1 cm below the vocal cords (Fig. 1). Attempted biopsy using traditional instruments was unsuccessful due to the solidity of the lesion. Using the CO_2 laser (Sharplan 1030 CO_2 Laser system) on continuous wave mode at a power of 25 watts, several puncture holes were made at its periphery, allowing piecemeal removal from the tracheal wall (as shown in Fig. 1). The remaining tracheobronchial tree was entirely normal and the patient was successfully extubated.

Subsequent histological examination demonstrated normal lamellar bone with no evidence of malignancy or metaplasia and a diagnosis of tracheopathia osteoplastica was made. A small residual projection of bone was found at subsequent bronchoscopy but the airway was satisfactory. A lateral neck X-ray demonstrated the residual rim of bone (Fig. 2). The patient remains well six months later.

Discussion

The exact aetiology of tracheopathia osteoplastica is uncertain although several theories have been proposed. Many believe that the condition is due to ecchondroses and exostoses as originally suggested by Virchow (1863), whereas Aschoff (1910) felt that elastic tissue metaplasia was the initiating process. Jepson and Sorensen (1960) found a relationship with primary ozaena and

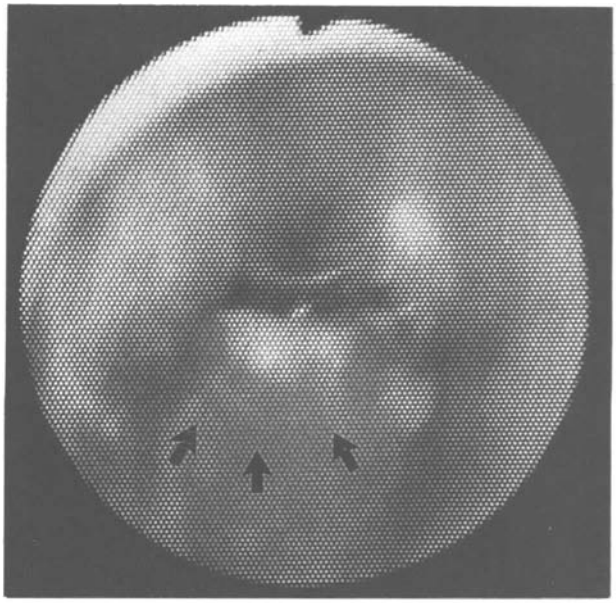

FIG. 1
Endoscopic view of the lesion with the endotracheal tube placed anteriorly. The sites of puncture holes created by CO_2 laser are shown (arrows).

From University Department of Otolaryngology, *Department of Anaesthesia, Manchester Royal Infirmary, Oxford Road, Manchester.
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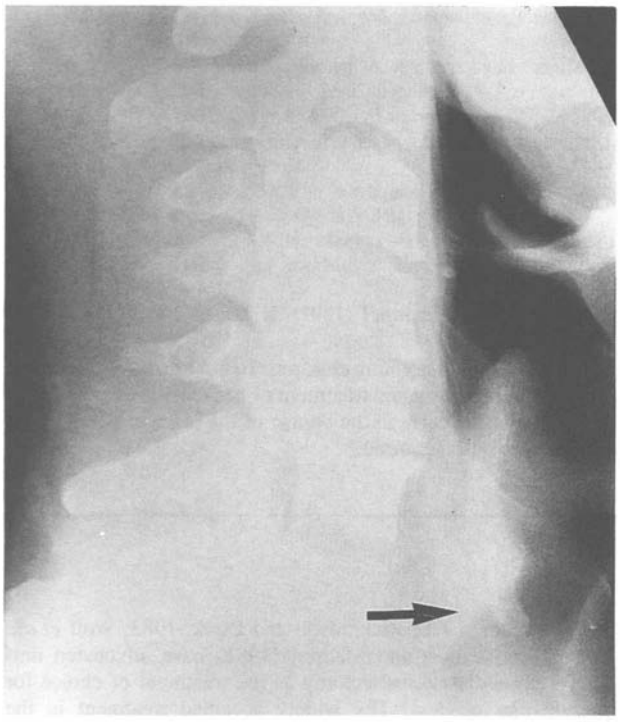

FIG. 2

Lateral neck X-ray. Note the bony remnant of the lesion projecting into the subglottis (arrow).

suggested that chemical or mechanical irritation was the initiating factor. This theory has been supported by Harma and Suurkari (1977) who reviewed 30 cases and confirmed an association with atrophic rhinitis and pharyngitis. They suggest that chronic inflammation leads to an accumulation of calcium salts within the tracheal mucosa around which cartilage and bone gradually develop.

Most patients with tracheopathia osteoplastica are asymptomatic with a reported incidence of 1 in 400 autopsies (Pounder and Pieterse, 1982). The introduction and widespread use of modern bronchoscopic methods has made clinicians more aware of the condition, although most sympotomatic lesions are both diffuse and extensive. Typically such patients have a history of chronic irritating cough associated with the production of blood-streaked crusts and intermittent hoarseness, sometimes many years before diagnosis (Martin, 1974). Localized lesions rarely present clinically and have been reported in association with difficult intubation on only two occasions (Lundgren and Stjernberg, 1981; Smith *et al.*, 1987) both of which were caused by long stenosing tracheal segments. In our case, the lesion was localized, arose from a narrow base and the remaining tracheobronchial tree was normal.

Increased awareness of this condition will prevent unnecessary anxiety, investigation and surgical intervention. Although the bony nature of the disease makes an adequate biopsy and excision difficult (Carr and Olsen, 1954), we found laser bronchoscopy very useful in allowing piecemeal removal of such a solitary lesion.

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Address for correspondence:
A. R. Birzgalis,
University Department of Otolaryngology,
Manchester Royal Infirmary,
Oxford Road,
Manchester M13 9WL.