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(Breif Communication)

# Primary Tracheal Neurilemoma (A Case Report)

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= Abstract = Primary tracheal neurilemoma is a very rare neoplasm. Only 17 cases were reported in the literature during 1950-1985. We report a case in a 15-year-old Korean boy who presented with persistent productive cough and dyspnea. An endobronchial lobulated mass was demonstrated in the left main bronchus. He was treated successfully by left bronchial resection and carinal reconstruction. The tumor was diagnosed as primary tracheal neurilemoma. This is the second case of primary tracheal neurilemoma diagnosed in Korea.

Key words: ***Neurilemoma, Trachea***

## INTRODUCTION

Primary neurogenic tumors are rare in the lower respiratory tract, being more frequently reported in the lung and bronchi than the trachea (Straus and Guckien 1951; Miller 1969; Silverman *et al.* 1976; Ma *et al.* 1981).

Of the neurogenic tumors, neurilemomas form approximately one third of the group(Bartley and Arean 1965). The most common locations are the flexor surfaces of the extremities, neck, mediastinum, retroperitoneum, posterior spinal roots, and cerebellopontine angle(Oberman and Suller 1967).

To our knowledge, the total number of tracheal neurilemomas reported in the literature from 1950 to 1985 is seventeen(Kim *et al.* 1985). We report a new case along with a brief review of the previous reports of tracheal neurilemoma.

## CASE REPORT

A 15-year-old Korean boy was admitted to Seoul National University Children's Hospital on July 14, 1986, because of a one month history of difficult respiration that didn't respond to therapy which included bronchodilators and steroids, and was aggravated 3 days prior to admission. He had a history of persistent productive cough and intermittent hemoptysis of 5 months' duration, without other symptoms of systemic illness.

On physical examination, respiration rate was 28 per minute. Alae nasi flaring, suprasternal and subcostal retractions were detected, with wheezing and moderate aeration over both lung fields. Arterial blood gas values in room air were pH 7.48, $pO_2$ 41 mmHg, bicarbonate 32 mmol/L. A chest roentgenogram showed right lung herniation with left lower lobe collapse. Bronchogram revealed a round lobu-

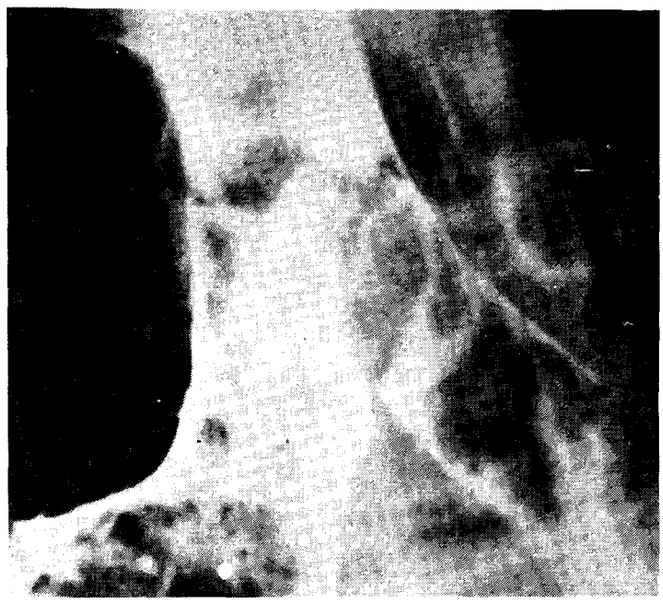

Fig. 1. On bronchogram, a round lobulated, 1.5 cm sized tumor mass arising from left tracheobronchial angle shows almost complete obstruction of the left main bronchus.

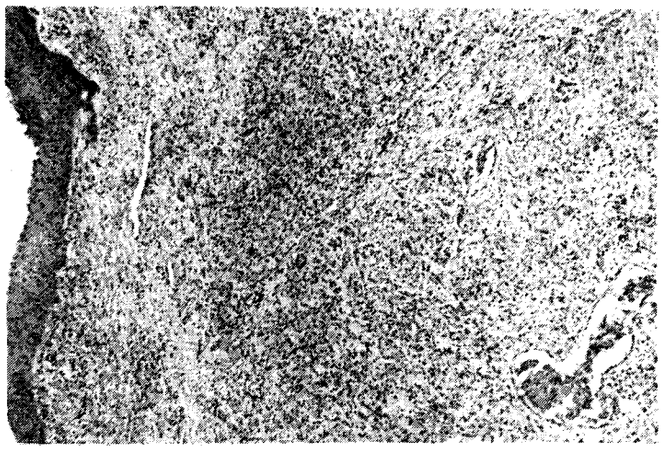

Fig. 2. Photomicrograph of the tumor, showing spindle cells in irregular arrangement and focal cystic degeneration.

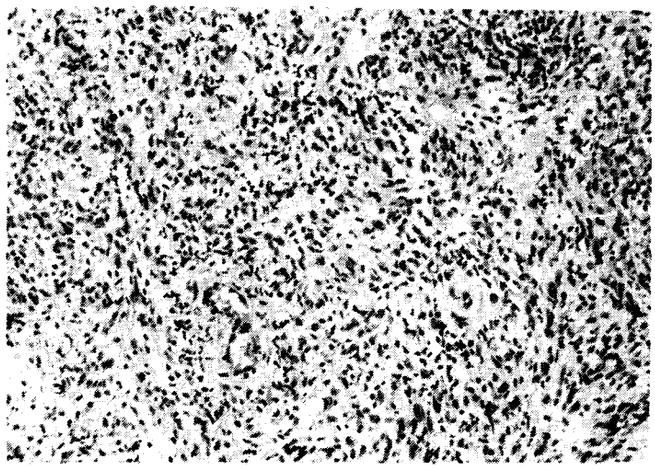

Fig. 3. Microscopic picture showing nuclear palisading in an area of tumor.

lated mass arising from the left tracheobronchial angle which completely obstructed the proximal left main bronchus (Fig. 1). At bronchoscopy, an erythematous movable mass was found covered with whitish necrotic material.

On August 1, 1986, left bronchial resection and carinal reconstruction were performed. At postoperative bronchoscopy, complete removal of tumor and patency of both main stem bronchi were confirmed. He was discharged on the 12th postoperative day, and the patient is doing fine as of the day of this report.

**Pathology**: The polypoid tumor was a reddish round smooth-surfaced friable mass, 1.5 cm in diameter, covered with whitish brown exudate. Microscopically, the moderately cellular tumor mass consists of spindle cells, arranged mostly in haphazard pattern with many areas of microcystic change, hemosiderin deposits and collagen fibrosis. Vessels were prominent. In a few areas, however, the tumor cells were arranged in a palisading fashion or in an organoid pattern. Occasional large hyperchromatic nuclei were present. However, mitotic figures were absent (Fig. 2, Fig. 3).

## DISCUSSION

The first report of a primary tracheal neurilemoma was in 1951(Straus and Guckien). Until 1985, a total of 17 cases were reported. They occurred in a wide age range from 6 to 71 years in both sexes, at all levels of the trachea(Kim *et al.* 1985). The symptoms may be insidious in onset and the tumor may occlude 3/4 of the lumen before symptoms are reported. The most common symptom is wheezing. Cough, dyspnea and hemoptysis are frequent. Pain, weight loss and malaise are unusual (Cleveland *et al.* 1977; Parrish *et al.* 1983). With progressive growth and more complete bronchial obstruction, secondary pulmonary changes occur-(Noya *et al.* 1970; Nass and Cohen 1979; Horovitz *et al.* 1983).

Because primary tracheal tumors are rare, the inclusion of tumors in the differential diagnosis of adult-onset dyspnea and wheeze would be non-practical. However, if the asthmatic patient's progress is less than satisfactory, the physician should consider obstructing lesions of the major airways-(Karlan *et al.* 1973; Horovitz *et al.* 1983; Parrish *et al.* 1983). High-penetration chest radiography, fluroscopy, tomograms and computerized tomography of the trachea(Horovitz *et al.* 1983; Parrish *et al.* 1983), pulmonary function tests(Parrish *et al.* 1983), and bronchogram(Noya *et al.* 1970) are helpful adjuncts for this. Bronchoscopy should be carried not only for diagnosis but also for planning the surgical resection(Kim *et al.* 1985).

Because of the possibility of severe hemorrhage and incomplete excision with early recurrence and due to technical difficulties in removal, endoscopic removal of tumors is a controversial approach, and tracheal resection with primary anastomosis, lobectomy, or pneumonectomy have been used (Abudallo *et al.* 1976; Nass and Cohen 1979; Ma *et al.* 1981; Horovitz *et al.* 1983). The prognosis is excellent following surgical excision, and postoperative recurrence is rare(Harkin and Reed 1969). However, malignant von Recklinghausen's disease was reported(Hanada *et al.* 1982). Because the tumor is characterized by slow growth and later recurr-

ence is possible, annual follow-up physical examinations, including chest X-ray films are recommended(Nass and Cohen 1979).

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=국문초록=

## 원발성 기관 신경섬유초종 1례


서울대학교 의과대학 소아과학교실·흉부외과학교실*·병리학교실**·방사선과학교실***

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저자들은, 5개월간의 지속성 기침과 기관지 확장제 및 부신피질 호르몬 등의 치료에 반응없는, 1개월간의 진행성 호흡관란을 주소로 내원한 15세 남아에서 기관종양을 발견하고, 기관지 절제술 및 단단문합을 실시하였던 바 이 종양의 조직검사상 원발성 기관 신경섬유초종을 나타내었다. 이는 1951년부터 1985년까지 전세계적으로 17례만이 보고된 매우 드문 종양이고 국내에서는 두번째 증례로 사료되어 보고하는 바이다.