Chronic Tracheopathia Osteoplastica
A Case Report

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Tracheopathia osteoplastica as defined by Carr and Olsen,¹ is a rare
disease of the trachea and bronchi. It is characterized by growths of
cartilage and bone within the wall of the trachea or bronchus that project
into the lumen. These may be in the form of sessile polyps or plaques.
If the growths become excessive they may partially obstruct the lumen
of the tracheobronchus. The clinical symptoms are cough, expectoration,
hemoptysis, fever and episodes of pneumonitis. The symptoms vary in
intensity, depending upon the extent of involvement and the degree of
obstruction. In 1887 Wilks² first described this disease. Many case re-
ports have been presented since that time. Dalgard³ had reviewed the
literature and established 90 authentic cases in 1947. Carr and Olsen⁴
in 1950 presented seven cases in Mayo Clinic.

The purpose of this report is to establish another case diagnosed by
bronchoscopy and present several features of this case which we feel are
unusual.

This 41 year old white man states he was well until the middle of March, 1964,
when he had chest pain on the left side, cough, expectoration of semipurulent material
and fever and chills. He was a student at one of the local colleges and was diagnosed
by the school physician as having pneumonia. An x-ray film dated March 31, 1964,
revealed atelectasis of the lingula. He was advised to seek bronchoscopy and admission
to the Missouri State Sanatorium. On April 17, 1964, bronchoscopy was performed,
as an out-patient. At the superior margin of the trachea on the left side a projection
of firm irregular tissue began and extended through the entire length of the trachea.
This tissue presented itself in shelf-like fashion. A biopsy taken from the inferior
portion of this was noticed to be hard and partly calcified. Visualization of the lingula
bronchus failed to reveal any obstruction which would account for the atelectasis. The
remainder of the tracheobronchial tree was normal.

He was then admitted to the Missouri State Sanatorium April 22, 1954. At this
time the atelectasis in the lingula division of the left lung had cleared but since no
positive diagnosis had been made, rebronchoscopy was advised. On May 25, 1954,
bronchoscopy was repeated. The same pathological findings were present. The pathological diagnosis of chronic tracheopathia osteoplastica was established by biopsy.
Because no specific therapy was indicated he was discharged and instructed to have
recheck bronchoscopy every six months.

On August 9, 1954, bronchoscopy was repeated and again no additional findings
noted. He developed a severe upper respiratory infection in September, 1955, with
cough, hemoptysis, fever and chills and was re-admitted to the Missouri State San-
otrium on October 3, 1955. Bronchoscopy was repeated on October 5, 1955, when the
lesion previously noted was again observed but further calcification and ossification
had occurred with almost complete fixation of the left side of the trachea. Purulent
secretions were noted in both bronchial systems. These were aspirated and cultures
for sensitivity made.

At the time of this admission he was placed on alevaire inhalation every four hours
with vaponephrine drops included in the inhalation. Nasal oxygen was given Pro ve

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FIGURE 1: Shows atelectatic area in the lingula of the lung.

FIGURE 2: Tracheogram showing thickening and rough edges in trachea.
Penicillin was administered twice daily for three days. On this routine he improved remarkably and was discharged October 13, '55.

He has had two episodes of obstructive pneumonitis since discharge. For the first of these which occurred in February, 1956, he was treated with broad spectrum antibiotics and prednisolone, 5 mgm. three times a day for five days. His symptoms of this episode completely abated and he had no further pulmonary symptoms until July 10, 1957. At this time he developed upper respiratory infection, cough, productive sputum, and a feeling of tightness in the upper anterior chest. Broad spectrum antibiotics were again prescribed for five days. On August 9, 1957, bronchoscopy was again repeated at St. John's Hospital, Springfield, Missouri. The previously described calcified growths along the left side of the trachea were again noted. For the first time, however, the calcified polyp-like growths were noted in the right primary bronchus and the bronchus intermedius of the posterior wall. Biopsy of these again was performed and the previously established diagnosis supported. He has now returned home with instructions to take postural drainage and ammonium chlorides during the winter months. He is to receive antibiotics from his local physician for three days each month rotating the antibiotics to prevent the emergence of resistant organisms.

We wish to express our appreciation to Dr. David F. Gorelick and Dr. Fred C. Coller, consultant pathologists of the Missouri State Sanatorium. We also wish to express our appreciation to Dr. William E. Taylor of the Southwest Missouri State College.

COMMENT

Carr and Olsen¹ discussed the hypothesis as to the pathogenesis of tracheopathia osteoplastica and state that none of these have been proved. They considered the most acceptable of these hypotheses to be Dalgaard's concept, in which the undifferentiated connective tissue cells in the wall of the trachea and bronchi develop into cartilage cells that grow into islands of cartilage, eventually becoming calcified. The invasion of the connective tissue is rich in blood vessel formation which results in the formation of marrow cavities lined with osteoblasts. This may account for the transformation into bone. It should be noted that for three years the disease in this patient was noted to be confined to the trachea. In

![Diagram of trachea](image)

**FIGURE 3**: (1) Schematic drawing of trachea. (1A) As seen through bronchoscope, 1955. (2) Schematic drawing showing extension into the right primary bronchus, 1957.
six bronchoscopies, all performed by the same individual (J.W.P.), no lesion had been found extending beyond the trachea. On the seventh bronchoscopy of August 9, 1957, involvement of the right primary bronchus and bronchus intermedius was noted. At the time of first bronchoscopy on April 17, 1954, the patient had an atelectasis of the lingula demonstrated by chest x-ray film but no obstruction could be found in the lingula bronchus.

The patient has now received broad spectrum antibiotics and prednisolone therapy on two occasions. An accurate evaluation of the benefit of the cortisone cannot be ascertained but symptomatic relief was prompt and effective. We feel that it is entirely possible, however, that the prednisolone and antibiotics might relieve some of the inflammatory reaction throughout the tracheobronchial tree and that in extremely severe cases of this disease its use would be well warranted. We would also stress the use of postural drainage, expectorants and bronchodilators in the treatment of this disease.

REFERENCES