THE FIRST REPORTED CASE OF BRONCHOLITHIASIS IN SINGAPORE

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Broncholithiasis is a lung condition characterised by the presence of one or more calculi in the bronchial tree. These calculi are referred to as broncholiths. The subject of broncholithiasis has been of great interest to the medical profession for centuries, although it is a condition that is rarely encountered. The purpose of this paper is to present the first case of broncholithiasis that has been seen in Singapore.

REVIEW OF LITERATURE

The first detailed description of broncholithiasis appeared in 1600 A. D. when Schenck a Grafenberg described 17 cases of broncholithiasis reported in the literature. In 1744 Boerhave described the case of a botanist who had coughed up 400 broncholiths over many years and died at the age of 54 years of "phthisis calculae". In the English medical literature Lloyd was able to find reports on only 18 cases of broncholithiasis from the period 1900 to 1930. He reported four cases of his own. Subsequently, in 1941 Van Ordstrand et alia found reports of 26 cases and reported 2 cases of their own. Zahn in 1946 was able to find reports of 71 cases in the literature. He added another case. His was the first case of broncholithiasis encountered in 4,000 cases of tuberculosis at Fitzsimons General Hospital. In 1950 Schmidt et alia publish- an excellent report on 41 cases seen at the Mayo Clinic. Baum et alia in 1958 presented 2 cases of expectorated broncholiths which were proved to be caused by histoplasmosis. Recently Coleman and workers at Mount Sinai Hospital have referred to 80 cases they have seen. In most of the recorded cases diagnosis was made by obtaining a history of having coughed up a broncholith, by finding a broncholith at the time of bronchoscopy, by surgical exploration, or by post-mortem examination.

PATHOGENESIS

One of the first persons to describe the pathological features of broncholithiasis was Poulalion (1891). He stated that broncholiths may be cartilaginous, calcareous or osseous and that they may arise anywhere in the pleuro-pulmonary tissues. Broncholiths may originate in the lumen of the bronchus, in the bronchial wall or around the bronchus subsequently eroding the wall of the bronchus.

The exact mechanism underlying the pathogenesis of broncholiths is still not entirely understood. But it is generally accepted that calcium deposition in inflamed, necrotic or degenerated tissue ultimately gives rise to the formation of broncholiths. Such calcification is now considered to occur usually in peribronchial lymph nodes involved by primary tuberculous infection or infection by histoplasma capsulatum. These calcified areas or lymph nodes may erode the bronchial wall and enter the bronchial lumen, partially or completely, to give rise to broncholiths.

Other conditions like pulmonary abscess, pulmonary cysts, pulmonary tumours may give rise to calcification and broncholith formation, but this is now thought to be unusual. In fact it is now felt by some that pulmonary disease is probably secondary to broncholith formation rather than vice versa.

It is generally accepted that metastatic calcification which occurs in conditions like hyperparathyroidism, renal rickets, osteomalacia and malignant bone tumours does not usually give rise to broncholiths.

The common sites of broncholith formation are at the lymph nodes situated in the angle between the middle and right lower lobe bronchus and at the lymph nodes at the origin of the anterior and lingular divisions of the upper lobes. Broncholiths vary in size from granules to the size of stones. They are usually greyish-white in colour, hard, rough and irregular. When coughed up or removed bronchoscopically they may get fragmented. Their main constituents are similar to normal bone composed of 85% - 90% calcium phosphate and 10% - 15% calcium carbonate.

A broncholith which is not coughed up promptly may cause collapse of lung distal to the obstruction. Subsequent to this obstruction secondary infection may occur and pneumonitis, bronchiectasis, lung abscess or empyema may follow.

CASE HISTORY

C. A. E., a female, aged 65 years, was seen at our chest outpatient clinic in February 1961. She gave a history of 2 years cough off and on productive of whitish sputum, and 2 weeks blood staining of sputum. There was no history of having swallowed or coughed up any foregin body.

On clinical examination she was found to be in good general condition. She was slightly febrile.

The pulse rate was 64 p.m. and the blood pressure was 140/86. There was no clubbing of fingers and no lymphadenopathy. The significant clinical findings were in the chest. The vocal fremitus was descreased over the right lower chest posteriorly . The breath sounds were diminished over the same area and there were crepitations with rhonchi. The vocal resonance was also decreased.

The chest P. A. x-ray showed the presence of an opacity in the right lower zone which on the right lateral view appeared to be mainly an anterior segmental consolidation—collapse of the right lower lobe. There was also an opacity in the right upper zone merging with the mediastinum. The latter was considered to be a soft tissue opacity.

Blood examination showed the E. S. R. to be 54/92 (1st & 2nd hour), the haemoglobin to be 70% (Sahli), the leucocyte count to be 12,500 per c.mm. and the differential cell count to be P86, L8, M2, E4.

The sputum was negative for tubercle on direct smear examination. Tuberculin test by the Heaf Multiple Puncture method was positive 1°.

A diagnosis of pneumonia of the right lower lobe was made but underlying bronchiogenic carcinoma was considered as a possibility. The patient was given a course of Tetracycline 250 mgms 6 hourly for one week. Re-x-ray of the chest after this course showed some clearing of the right basal opacity but this was not complete. Subsequently a one week course of Chloramphenicol 250 mgms 6 hourly was given but with no further change in the chest x-ray appearance. As bronchogenic carcinoma had been considered as a possible underlying pathology initially, in view of the persistence of some of the basal opacity, a bronchoscopy was done.

At bronchoscopy done under local anaesthesia, the trachea was found to have much light brownish-yellow pus. The left main bronchus and its branches were normal. The pus was seen to be coming from the right main bronchus. The whole right bronchial tree was inflamed. In the right lower lobe bronchus just distal to the opening of the apical branch of the right lower lobe, a mass was seen with necrotic tissue, pus and blood around it. The mass was removed in fragments with necrotic tissue. Among the fragments was a fairly large greyish gritty hard mass 12mm x 6mm x 6mm in size. On close examination it looked like a broncholith.

The fragments on chemical analysis were found to contain calcium and magnesium phosphate consistent with that of normal bone.

The patient made a fairly uneventful recovery after bronchoscopy. During the recovery period she coughed up 2 more broncholith fragments. The physical signs in the chest resolved and there was complete clearing of the chest x-ray shadow in the right lower zone with re-aeration of the anterior segment of the right lower lobe. She is still attending our out-patient clinic for follow-up and remains in good health and free of symptoms.

DISCUSSION

The presence of a broncholith may or may not give rise to symptoms. When it does give rise to symptoms these are usually the result of the secondary effects of the broncholiths. The common symptoms are recurrent cough, fever associated with lung infection distal to the broncholith and haemoptysis which may sometimes be massive. The clinical picture may often be indistinguishable from a case of bronchogenic carcinoma. Postero-anterior, oblique and lateral chest x-rays, tomographic x-rays and bronchograms may help in showing up the broncholith and its location. A final diagnosis sometimes can be made at bronchoscopy as it was in this case, but often can only be made at thoracotomy with surgical exploration, resection and biopsy of the resected part.

The treatment of broncholiths is bronchoscopic removal where possible. This is possible in about 25% of cases. In the others, if symptoms are persistent or severe, like recurrent lung suppuration or massive haemoptysis, then thoracotomy with surgical exploration and resection of the affected area may be necessary. Such resection may sometimes be hazardous and has to be quite extensive, often much more extensive than the segment involved due to the often marked inflammatory changes that take place at the lung root from infection. For this reason surgical intervention should be withheld as long as possible and undertaken only when absolutely necessary.

In this patient the prevailing symptoms were blood staining of sputum and cough. On the clinical, laboratory and radiological findings a diagnosis of pneumonia was made without excluding the possibility that it could be a bronchogenic carcinoma. It was only at bronchoscopy that the final diagnosis was made when the broncholith was removed.

It is now generally accepted that broncholiths are due to calcified lymph nodes adjacent to bronchi eroding through the bronchial wall into the lumen. The calcified lymph nodes are usually due to primary tuberculous infection or infection by histoplasma capsulatum.

It is significant that in a place like Singapore

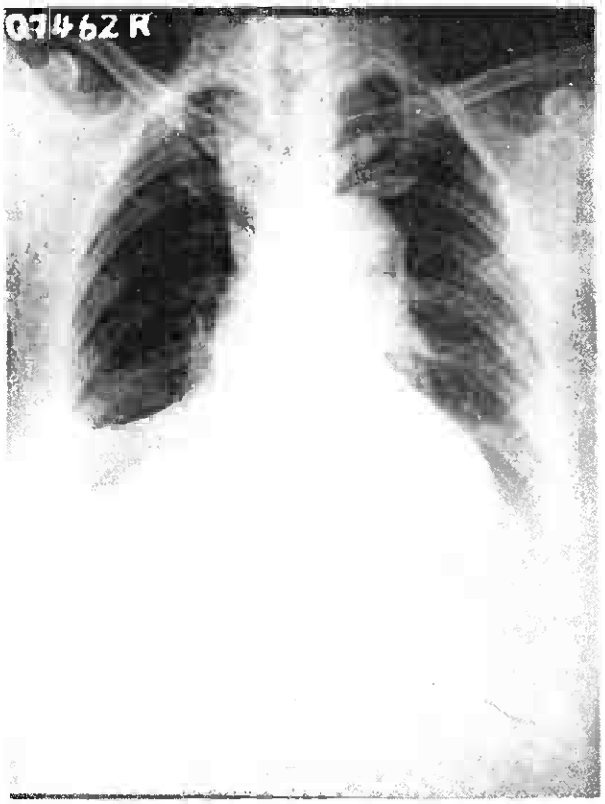

Fig. 1. Chest P.A. x-ray of patient when first seen at our outpatient chest clinic showing opacity in the right lower zone.

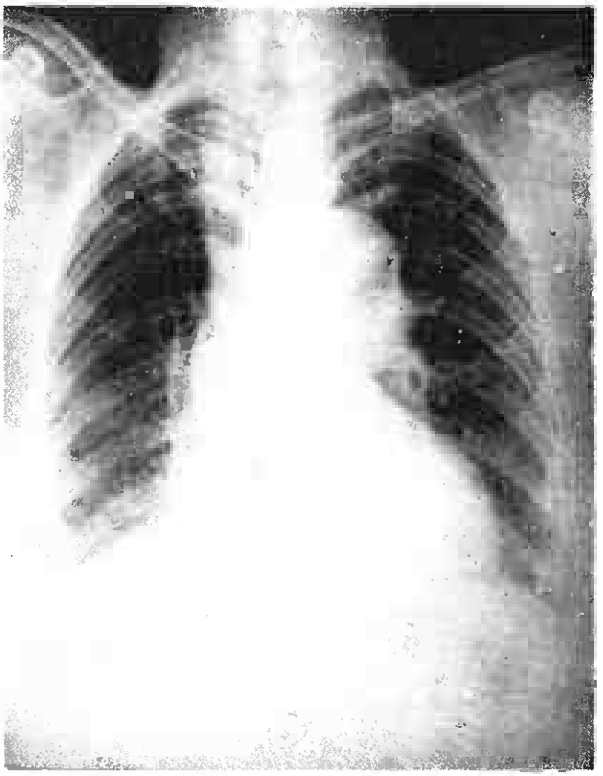

Fig. 3. Chest P.A. x-ray of patient after bronchoscopic removal of broncholith. The opacity in the right lower zone has cleared completely.

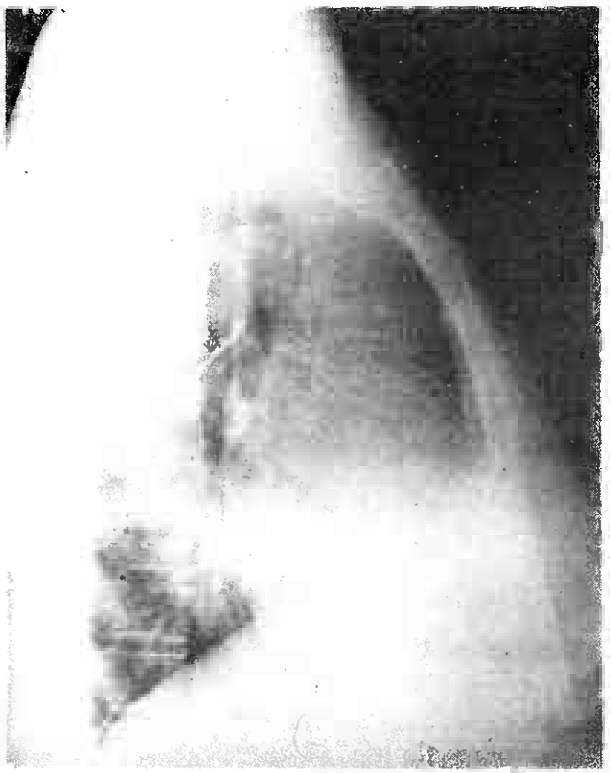

Fig. 2. Right lateral x-ray of patient when first seen at our outpatient chest clinic showing consolidation-collapse of mainly anterior segment of the right lower lobe.

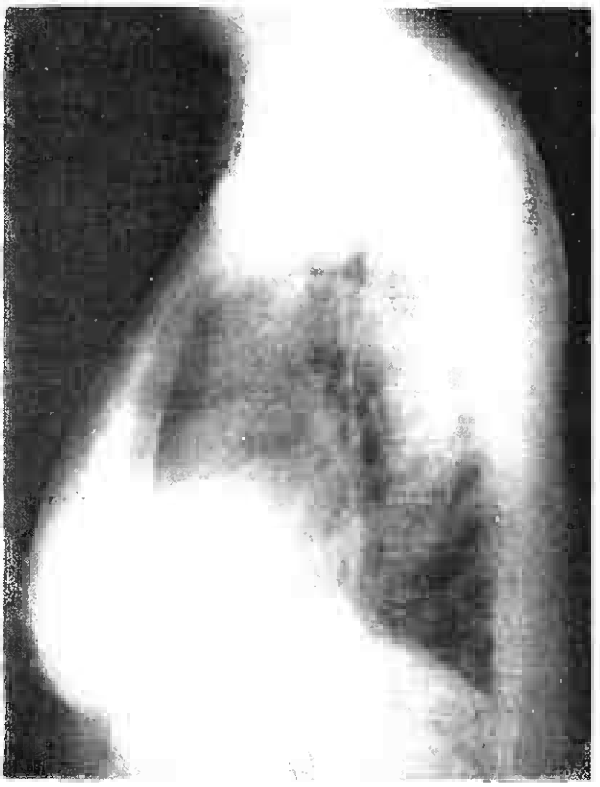

Fig. 4. Right lateral x-ray of patient after bronchoscopic removal of broncholith. The consolidation-collapse appearance of the anterior segment of the right lower lobe has cleared completely.

where the incidence of primary tuberculous infection is high and lymph node calcification common, in our experience broncholithiasis is rare. It has been so rare that this is the first reported case of it here.

SUMMARY

A case of broncholithiasis is presented as the first reported case of its kind in Singapore.

A review of some of the literature on the subject is given and the probable pathogenesis of the condition is discussed.

The symptomatology, diagnosis and treatment are briefly discussed.

REFERENCES

Schenck a Grafenberg, Joannes: Vol I Frankfurt. 1600. J. Rhodii pp 351-355.

Boerhave, quoted by Legry (1892): Arch gen. Med., 169, 337.

Lloyd, J.J. (1930): "Broncholithiasis with report of four cases", Amer J. Med. Sci., 179, 694-699.

Van Ordstrand, H.S., Moore, P.M., and Harris, H.E., (1942): "Broncholithiasis; Report of two cases. Cleveland Clin. Quart.; 9, 36-44.

Zahn, D.W. (1946): "Broncholithiasis", Amer. Rev. Tubec, 54, 418-423.

Schmidt, H.W., Claggett, O.T., and McDonald, J.R. (1950): "Broncholithiasis" J. Thoracic Surg., 19, 226-245.

Baum, G.L. Bernstein, I.L., and Schwarz, J. (1958): Amer. Review Tuberculosis, 77, 162.

Poulalion, S.A.M. (1891) "les pierras du poumon de la plavre et des bronches, et la pseudophthisis pulmonaire d' origine calculeuse", Thesis, Paris, G. Steinheil 240 pp.