

# Spontaneous contained transmural oesophageal rupture clinically resembling intramural rupture

## A case report

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### Summary

Spontaneous rupture of the oesophagus may occur either transmurally or intramurally. The symptoms, signs, clinical course, treatment and prognosis differ in the two types. Transmural rupture is generally regarded as a serious condition, usually requiring operative treatment and having a high morbidity and mortality. Intramural rupture is a much more benign condition, is treated non-operatively and has a good prognosis.

A few cases have been documented in which a transmural rupture was contained within the mediastinum; an additional case is described. Under these circumstances the transmural rupture has the clinical features of an intramural rupture. It is suggested that transmural ruptures should be subdivided into those which are not contained (the vast majority) and those which are.

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Spontaneous transmural rupture of the oesophagus is a well-defined clinical entity in which a longitudinal tear occurs, usually in the left posterolateral wall of the distal third, presumably due to a sudden increase in intra-oesophageal pressure.<sup>1</sup> Among the characteristic symptoms are severe substernal and epigastric pain, rapid respiration and shock.<sup>1,2</sup> The early signs include epigastric tenderness and rigidity, evidence of hydropneumothorax, usually on the left, and subcutaneous emphysema in the neck.<sup>1,2</sup> Pleural aspiration may reveal food particles or gastric juice in the aspirate.<sup>1</sup> Septic mediastinitis and septicaemia may follow. Chest radiography usually shows a hydropneumothorax and mediastinal emphysema,<sup>1,2</sup> while a barium contrast swallow demonstrates the rupture with dye tracking into the mediastinum or pleural cavity. Transmural rupture has been variously described as an ominous<sup>3</sup> or catastrophic<sup>4</sup> event, with a continuing high mortality,<sup>5</sup> requiring operative treatment in the vast majority of cases.<sup>4,6</sup> A few patients recovering without thoracotomy have been described but these should be regarded as rare exceptions.<sup>1,2,7</sup>

Spontaneous intramural rupture of the oesophagus differs in many respects from the transmural variety.<sup>3,8-11</sup> In this condi-

tion a barium contrast swallow shows a tiny tear extending into the wall, with longitudinal band-like defects caused by intramural dissection. No actual penetration of the wall or extravasation of dye occurs. In 1980 Kerr<sup>3</sup> found that 15 cases of spontaneous intramural rupture and intramural haematoma had been documented, and described 5 new cases. In the same year 2 further cases were reported<sup>9</sup> and subsequently 2 more.<sup>11</sup> The dominant symptom in all 24 cases was severe retrosternal or epigastric pain, with concomitant dysphagia in all and haematemesis in some. None of the severe signs of transmural rupture were present, and the chest radiographs showed no evidence of pneumothorax, pleural effusion or surgical emphysema in the neck or mediastinum. The definitive diagnosis was by means of a barium contrast swallow.<sup>3,8</sup> Endoscopy was advocated by some but it was felt that it should be avoided in the acute stages.<sup>11</sup> All but one of the patients with intramural rupture recovered with non-operative, conservative treatment (*vide infra*).

Occasionally a case is encountered in which the clinical features differentiating transmural from intramural rupture are less clearly defined, as in the following one.

### Case report

A 48-year-old man first experienced a continuous, gnawing, non-radiating lower retrosternal pain while walking about at home. He was not eating at the time and there was no retching, vomiting or coughing. Swallowing liquids or solids aggravated the pain. His general practitioner found him to be in good general health and not distressed. Blood pressure and the cardiovascular and respiratory systems were normal. Oesophageal or gastric ulcer was diagnosed and treatment instituted accordingly.

Both the retrosternal pain and dysphagia increased, necessitating admission to hospital 10 days later. Again no obvious distress was present. Systematic clinical examination revealed no abnormality. The pulse rate, blood pressure, electrocardiograph and differential blood count were normal.

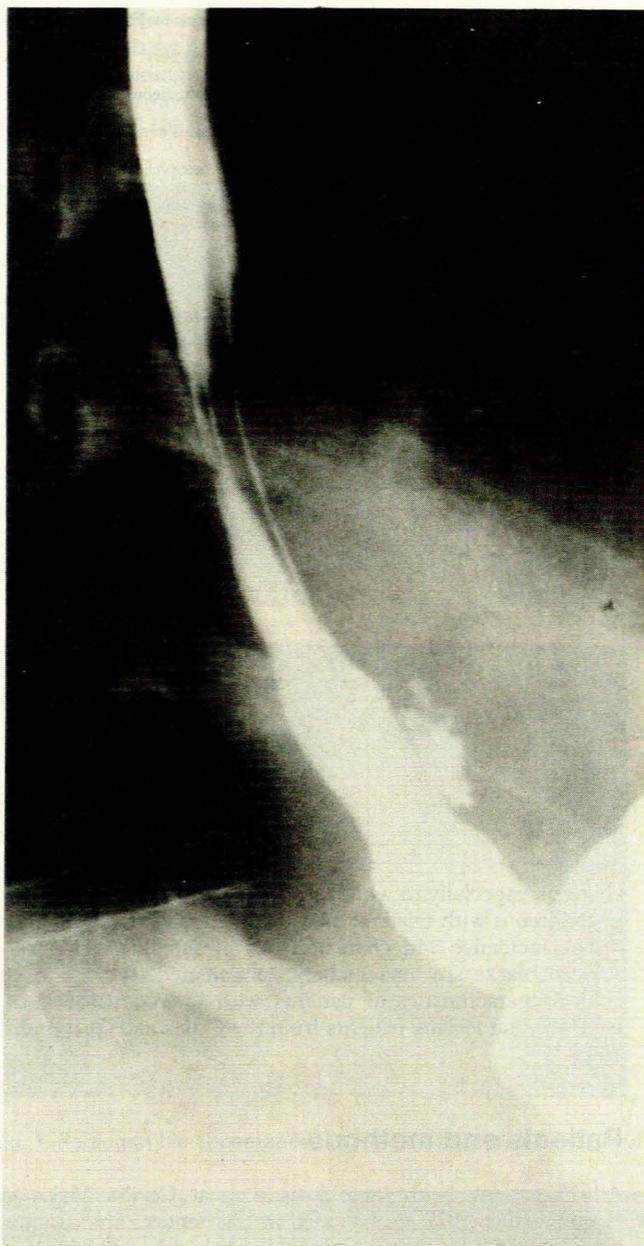
A barium swallow showed a transmural rupture of the left posterolateral border of the lower thoracic oesophagus, approximately 3 cm above the diaphragmatic hiatus with no intramural dissection (Fig. 1). The contrast medium eventually tracked to the left and upwards into the mediastinum for a distance of 3 cm from the oesophagus, but did not enter the pleural or pericardial spaces. Chest radiographs showed pleural thickening around the base of the left lung (Fig. 2). There was no pneumothorax or mediastinal or subcutaneous emphysema. Subsequent pleural puncture confirmed the absence of fluid at the left base. The condition was diagnosed as a spontaneous transmural rupture of the oesophagus, probably localized or contained by previous pleural thickening and adhesions. In view of the absence of shock, septicaemia and other thoracic signs, non-operative treatment, consisting initially of broad-spectrum antibiotics and parenteral feeding, was instituted.

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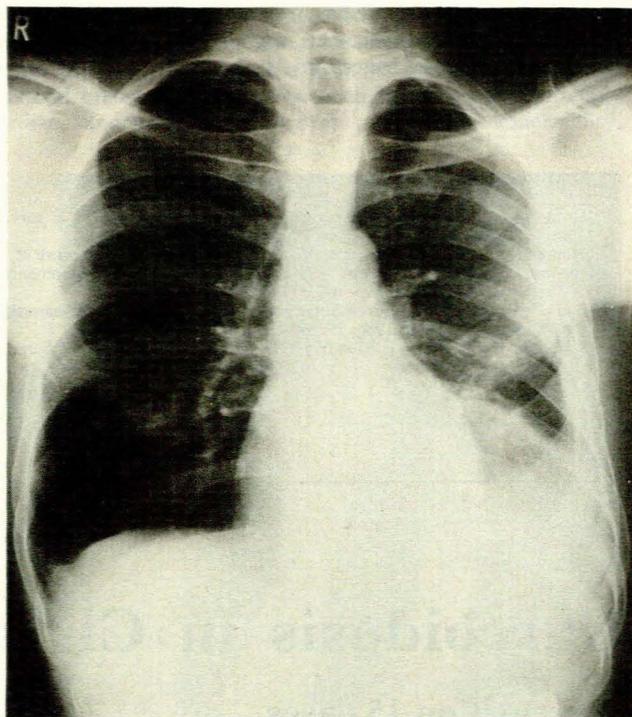
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**Fig. 1.** Transmural rupture of the left lower thoracic oesophagus. The barium eventually tracked to the left for a distance of 3 cm from the oesophagus.

Oesophagoscopy 8 days after admission failed to show the rupture or any other oesophageal lesion. While steady improvement occurred during the patient's hospitalization, barium swallows were repeated at 2, 6 and 14 weeks. At all examinations the transmural, contained rupture was seen with progressively less contrast medium outside the lumen. At 8 weeks he had become symptom-free, i.e. at a time when the rupture was still evident radiologically. Eventually, at 8 months, it sealed off.

Two years previously the patient had been admitted because of the sudden onset of sharp, stabbing epigastric and left-sided pleuritic pain and dyspnoea following a long history of chronic productive cough. The pain at that time had a different distribution and had not been associated with dysphagia. Examination had revealed a swinging temperature, a right-sided pleural effusion and a large left pyopneumothorax. There had been no mediastinal or subcutaneous emphysema, and no



**Fig. 2.** Dense pleural thickening at the base of the left lung due to a previous pyopneumothorax.

gastric or oesophageal contents had been aspirated from the pleural cavity. Although two sputum cultures had been negative for tubercle bacilli, the condition had been diagnosed clinically as a tuberculous pyopneumothorax. It had responded well to closed pleural drainage and antituberculosis therapy, with complete clinical recovery but with residual pleural thickening at the left base.

## Discussion

The few reported patients with 'free' (i.e. not contained) transmural rupture of the oesophagus who recovered without thoracotomy can be regarded as rare exceptions. The present case is in a different category since a transmural rupture occurred which was contained or localized within the mediastinum, presumably due to pre-existing pleural adhesions following earlier pyopneumothorax. It shows that, where containment occurs, a transmural rupture may present clinically as an intramural rupture without any of the severe symptoms and signs of a transmural rupture. Like intramural rupture, it may also be treated non-operatively.

A similar case has been reported previously by Hammerschmidt *et al.*<sup>2</sup> Cameron *et al.*<sup>4</sup> reported 8 cases in which contained transmural intrathoracic oesophageal disruptions were treated non-operatively (none of these ruptures being spontaneous). The criteria for conservative management were determined as a disruption contained in the mediastinum, a cavity draining back into the oesophagus, minimal symptoms and minimal signs of sepsis. It appears that the present case falls into this category.

It can be concluded that transmural ruptures should be subdivided into those which are not contained (the vast majority) and those which are (a small minority). Provided the latter conform to the criteria laid down by Hammerschmidt *et al.*,<sup>2</sup> non-operative therapy may be considered.

It should be pointed out that while intramural rupture is 'benign' in comparison with the transmural variety, a single

case of intramural rupture has been reported in which extensive dissection of the wall occurred, and this led to necrosis which required a total oesophagectomy.<sup>12</sup>

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# Sarcoidosis in Ciskei

## A report on 15 cases

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### Summary

Since 1977 15 patients with sarcoidosis have been seen at the Cecilia Makiwane Hospital, Ciskei. Three patients were 13 years of age or less and 5 were under 20 years; 6 patients had phalangeal osteolytic lesions, 5 with concomitant skin involvement. Only 3 patients had respiratory symptoms.

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In the largest study of sarcoidosis in South Africa, involving 110 patients seen at Groote Schuur Hospital between 1969 and 1975,<sup>1</sup> it was noted that uveitis, skin lesions and lymphadenopathy were more frequently found in black patients than in white patients, while respiratory symptoms occurred far less frequently in blacks. None of the patients had bony involvement. In 1974, 10 patients with radiological evidence of sarcoidosis in the hands were reported from Pretoria.<sup>2</sup> Characteristic features in these patients included cyst formation and destruction of the bones and joints.

Involvement of bone is considered an uncommon manifestation of the disease and is typically found in patients with skin sarcoidosis. Sarcoidosis appears to be relatively uncommon in

children, especially in whites, and the most usual presentation in children is with bilateral hilar lymphadenopathy.<sup>3</sup>

The incidence and characteristics of sarcoidosis in South African blacks are inadequately documented. We report the clinical characteristics of patients with sarcoidosis seen at a large hospital serving patients from the Ciskei and surrounding areas.

### Patients and methods

All 15 patients with sarcoidosis seen at Cecilia Makiwane Hospital since 1977 are included in this report. The diagnosis was supported by histological evidence in 12 of the 15 cases; the clinical picture and course were characteristic in the other 3. The liver, skin and lymph nodes were the usual biopsy sites. In most cases haematological, biochemical and radiographic studies were performed. Both the Heaf and Mantoux methods were used for tuberculosis skin testing. Measurements of vital capacity and forced expiratory volume in 1 second were made with a dry spirometer (Vitalograph). The Kveim skin test was not performed in any of the patients.

### Results

The usual female-to-male ratio of 2:1 was observed in our small series, in which 11 out of the 15 patients with sarcoidosis were females. There were 5 patients under the age of 20 years, and 3 of these were 13 years old or younger. The commonest clinical feature at the time of diagnosis was the incidental finding of an abnormal chest radiograph in 9 patients with no respiratory complaints in 6; 5 of these had bilateral hilar lymphadenopathy and 1 an interstitial lung pattern.

The most striking feature in our series was the large number of patients (6) with osteolytic phalangeal lesions; 5 of the 6 had

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