Oesophageal moniliasis causing fistula formation and lung abscess

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Oesophageal moniliasis is not a rare disease; more than 100 cases have been reported in the world literature in the last decade, and evidence indicates that the incidence may be much higher. It is becoming more common with increasing use of long-term antibiotics in those who have debilitating disease (Kaufman, Scheff, and Leviene, 1960) and in long-term survival patients who have had chemotherapy for malignant disease. Oesophageal moniliasis may occur during the course of malignant disease (Jensen et al., 1964), haematological disorder (Sanders et al., 1962), Plummer-Vinson syndrome (Watanabe, Tamaoki, and Uno, 1967), or multiple operations (Gaines and Remington, 1972). A possible association of intramural oesophageal diverticulosis and moniliasis has been described (Troupin, 1968) but Holt (1968), in a review of the literature, reported several cases with no predisposing cause. Our case probably falls into this group.

Oesophageal perforation due to oesophageal moniliasis has not been reported so far as we can trace. We report a case with pathological proof of oesophageal moniliasis with fistula formation into the apical segment of the right lower lobe and abscess formation but no pleural or mediastinal involvement at the time of operation.

CASE REPORT

A 28-year-old farmer was admitted to the Soraya Medical Centre, Esfahan University in June 1973 with a six-month history of dysphagia, shortness of breath, right chest pain, and productive cough, all symptoms becoming progressively worse; he had lost approximately 20 kg in weight. There was no complaint of postural aggravation, acid taste in the mouth, or heart-burn. Past history was completely negative and there was no history of having taken antibiotics. The patient had been healthy until six months before admission. On admission he was depressed, ill-looking, and dyspnoeic.

Routine laboratory tests were unremarkable; the erythrocyte sedimentation rate was 87/mm in the first hour; sputum culture showed Klebsiella.

Examination of the chest revealed rales throughout the right side and diminished breath sounds in the right lower hemithorax. The chest radiograph showed a large cavity with a fluid level in the right lower hemithorax (Fig. 1).

Barium swallow showed an irregularity of the entire oesophageal wall with pseudodiverticula
formation; no contractions could be seen in the oesophagus. The contrast media passed through a long, narrow fistula from the junction of the upper and middle thirds of the oesophagus to a large cavity in the right hemithorax (Fig. 2).

**FIG. 2.** Irregular oesophageal wall with a narrow fistula to a large cavity in the right hemithorax.

Oesophagoscopy showed an oedematous, reddish mucosa for the first 7 cm of the oesophagus, and below that the mucosa was irregular and shaggy with whitish necrotic tissue covering it as a slough. The oesophageal lumen was narrowed, and the oesophagoscope could not be passed beyond the level of the aortic arch. The mucosa was haemorrhagic and friable and bled easily during the examination.

The histopathological diagnosis of the biopsy material was 'pseudoepitheliomatous hyperplasia'.

The oesophageal washings showed the presence of mycelium and yeast-like cells, suggesting a *Candida* infection. Tissue samples from the oesophageal mucosa stained by Gram's method showed the presence of yeast-like cells. Culture of the same sample on propagation and differential media revealed the growth of typical chlamydospores of *Candida albicans* (Fig. 3).

The preoperative diagnosis was oesophagopulmonary fistula with lung abscess, most likely due to moniliasis. The patient underwent a right thoracotomy, and a large abscess in the right lower lobe, primarily in the apical segment, with some pleural adhesions was found. The abscess contained about 300 ml of putrefied material sur-

**FIG. 3.** Typical yeast-like cells and chlamydospore of *Candida albicans* cultured from tissue taken from within the oesophagus (Coton blue, ×200).
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rounded by necrotic lung tissue. A fistula, 3 mm in diameter, from this abscess to the junction of the upper and middle thirds of the oesophagus was demonstrated; the oesophageal wall was thickened in the upper and middle thirds.

A right lower lobectomy and total oesophagectomy were performed. The patient was left with a gastrostomy and an oesophageal fistula in the neck. A long loop colon transplant from cervical oesophagus to stomach is planned in six months' time.

Macroscopic examination of the oesophagus revealed whitish-grey patches on the mucosa which produced a complete false membrane over the whole oesophageal mucosa; a fistula, 3 mm in diameter, from the junction of the upper middle thirds was confirmed.

Microscopic examination revealed thickening of the mucosa, desquamation of the squamous epithelium, and lymphocytic infiltration in the submucosal and muscularis layers. Periodic acid-Schiff stain was used and revealed numerous mycelial threads and blastospores in the submucosa and situated more deeply in the muscularis layer (Fig. 4).

Preoperatively the patient received penicillin, streptomycin, gentamycin, oral nystatin, and amphotericin B. With the exception of nystatin, these were continued after surgery.

DISCUSSION

Candida albicans is normally a saprophytic organism. It can become a significant pathogen in the circumstances previously noted. The most common sites of involvement are the mouth, skin, vagina, and respiratory tract. Gaines and Remington (1972) reported eight cases of gastrointestinal tract (18%) and 12 cases of lung involvement in 33 surgical patients with systemic candidiasis confirmed by necropsy; others report similar findings. The usual sites of monilial involvement in the gastrointestinal tract are the mouth, pharynx, and lower gastrointestinal tract. The oesophagus, however, is an uncommon site for the disease. The first description of oesophageal moniliasis as a radiographic finding was by Andrén and Theander (1956). Troupin (1968) reported the first pathologically proved association of diverticulosis with moniliasis. In spite of the fact that perforation and fistula formation with mediastinitis are potential complications of oesophageal moniliasis, the incidence of fistula formation is extremely rare. If oesophageal perforation occurs acute mediastinitis

FIG. 4. Deep mucosa of the oesophagus showing mycelial threads and blastospores (Periodic acid-Schiff X355).
and empyema are potential complications but in our case no mediastinal involvement was present, the fistula opening directly into the apical segment of the right lower lobe.

In a review of 282 cases of oesophageal perforation collected from Neolon, Cuddy, and Gibbon (1961); Foster et al. (1965); Groves (1966); Hardin, Hardy, and Conn (1967); Gerard et al. (1968); Johnson, Schwegman, and MacVaug (1968); Wichern (1970); and Benny and Ochsner (1973) no record of oesophageal perforation, fistula formation, and lung abscess due to moniliasis is available.

REFERENCES


Requests for reprints to: Dr. Siavoush Sehhat, Esfahan Medical School, 160 Shah Avenue, Esfahan, Iran.
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