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CLINICAL

An unusual cause of dysphagia in a young adult

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ABSTRACT We report a case of a young woman who presented to our institution with recent-onset dysphagia. At oesophagogastroduodenoscopy an oesophageal ulcer was found. Biopsies revealed changes consistent with granulomatous inflammation. A diagnosis of oesophageal tuberculosis was made and, following anti-tuberculous medication, the ulcer and symptoms completely resolved.

KEYWORDS Anti-tuberculosis therapy, oesophageal tuberculosis, oesophagogastroduodenoscopy

DECLARATION OF INTERESTS No conflict of interests declared.

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CASE REPORT

A 25-year-old woman presented to us with a complaint of three months' progressive dysphagia. She was otherwise in good health and denied any weight loss, chest pain, cough, expectoration or fever. A clinical examination proved unremarkable. In particular, neither neck nodes nor hepatosplenomegaly were noted. Laboratory assessment showed a normal complete blood count, liver and renal function. Oesophagogastroduodenoscopy (OGD) showed an oesophageal ulcer with rolled, everted edges, starting at 25 cm from the incisors (Figure 1). The endoscope passed down the oesophageal lumen and into the stomach with ease. Biopsy revealed changes consistent with granulomatous inflammation (Figure 2). There was no evidence of dysplasia or malignancy. A Ziehl-Neelsen stain was negative for acid-fast bacilli.

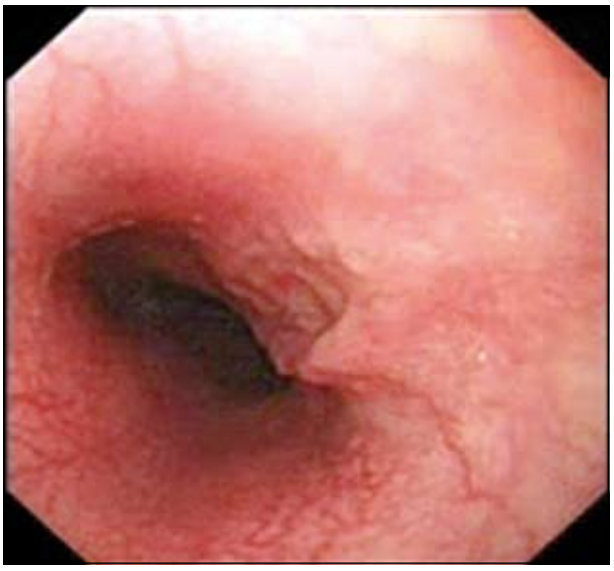

FIGURE 1 Image of an oesophageal ulcer with everted edges starting at 25 cm from the incisors, from a 25-year-old woman presenting with a three-month history of progressive dysphagia.

A diagnosis of probable oesophageal tuberculosis was made. In order to distinguish between primary oesophageal tuberculosis and secondary involvement of the oesophagus by a tuberculous process in the mediastinum or lung, a CT scan of the thorax was carried out (Figure 3). No evidence of cavitation or miliary disease was apparent on the lung images. A diagnosis of primary, as opposed to secondary, oesophageal tuberculosis was therefore made. At the time, we did not have facilities for culture and sensitivity testing for *Mycobacterium spp*. A test for HIV infection was negative. A decision was therefore made to treat the patient with anti-tuberculous therapy and to monitor the clinical situation closely to assess response to treatment.

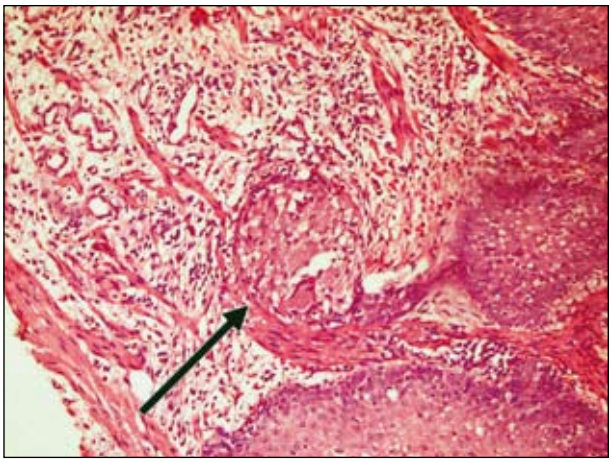

FIGURE 2 Section from the ulcer biopsy to show the necrotising granulomatous inflammatory nature of the lesion with no evidence of dysplasia or malignancy. Ziehl-Neelsen staining was negative for acid-fast bacilli.

The patient was started on isoniazid, rifampicin, ethambutol and pyrazinamide, with pyridoxine. Her symptoms settled rapidly, and a follow-up OGD three months after starting treatment revealed some mucosal distortion at the site of the previous ulcer, but no luminal narrowing. Anti-tuberculous therapy was continued for a total of ten months. A further OGD at

14 months was normal with no scar or luminal narrowing evident (Figure 4). The patient is currently well, some 14 months post completion of treatment, without oesophageal symptoms or systemic symptoms of tuberculosis.

DISCUSSION

Dysphagia is an unusual symptom in a 25-year-old individual. It would be reasonable to be perplexed by such a patient in routine clinical practice, since she might be thought too old for the congenital causes of dysphagia, such as oesophageal atresia, Schatzki's ring, a vascular ring and oesophageal webs, and too young for some of the acquired ones, such as a peptic stricture or an oesophageal neoplasm. At our tertiary care cancer hospital in Pakistan, the mean age of presentation for oesophageal cancer is considerably lower than that seen in the West. A recent epidemiologic study from our own institution has shown a mean age at presentation of 56 years, compared with the 65–70 years commonly seen in the West.[1] Thus, one would be correct to consider unusual causes of dysphagia such as eosinophilic oesophagitis, scleroderma, herpes oesophagitis, tuberculous oesophagitis, dysphagia lusoria and muscular dystrophy, among others.

Oesophageal tuberculosis is a rare clinical entity. An early twentieth century literature review of 16,489 autopsies of individuals with tuberculosis revealed that only 0.15% had oesophageal tuberculosis.[2] A more recent review by Marshall showed only 0.3% cases of oesophageal tuberculosis out of 297 patients suffering from tuberculosis of the gastrointestinal tract.[3] Oesophageal tuberculosis can occur as a result of direct extension from adjacent infected structures, such as mediastinal or hilar lymph nodes, a pulmonary focus, vertebral bodies or the larynx, as well as by haematogenous dissemination, as in miliary tuberculosis.[2–5] It is clear that secondary tuberculosis of this kind usually occurs in the presence of tuberculosis elsewhere, and tends to occur in a patient with evidence of widespread tuberculous infection.

Primary oesophageal tuberculosis is a rare diagnosis. While most types of oesophageal tuberculosis will present with dysphagia, endoscopic appearances are quite variable, ranging from hypertrophy, granular and ulcerated form.[6–8] Oesophageal tuberculosis presenting in the form of a tumour-like growth with a stricture can be mistaken for a carcinoma.[9–10] The most common site of oesophageal involvement is in the mid-oesophagus near the bifurcation of the trachea.

Oesophagogastroduodenoscopy and biopsy are the diagnostic procedures of choice in oesophageal tuberculosis.[3–7] A CT scan of the thorax is mandatory for differentiating primary from secondary oesophageal

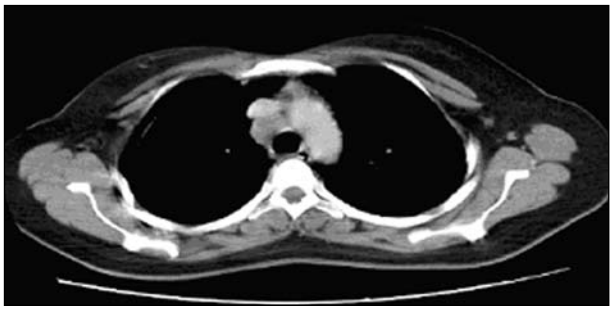

FIGURE 3 The pre-treatment CT scan of the thorax showed multiple subcarinal and peri-tracheal lymph nodes, the largest about 1.5 cm in size. However, none were in direct contact with the oesophagus, and there was no evidence of pulmonary disease.

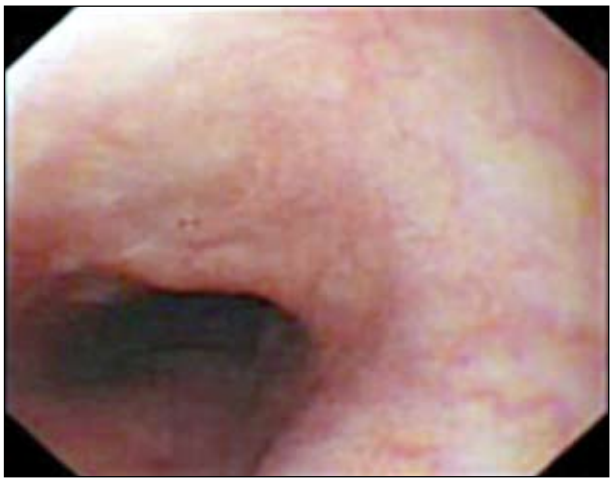

FIGURE 4 A follow-up image showing the same region of the oesophagus as in Figure 1, 14 months later, following nine months of antituberculous treatment.

infection. In the patient we present, enlarged mediastinal nodes were seen but were not felt to be contiguous with the oesophagus. Mediastinal tuberculosis involving the oesophagus may present with features similar to those seen in the patient described, but may also be associated with with fistula or sinus formation.[10] Endoscopic ultrasonography may be of further value in distinguishing between primary and secondary oesophageal tuberculosis, since it may help to define more clearly whether the oesophageal lesion is secondary to the direct extension of mediastinal lymphadenopathy.

Confirmation of the diagnosis of oesophageal tuberculosis requires histological demonstration of caseating granulomata and acid-fast bacilli from the endoscopic biopsies or isolation of M tuberculosis from tissue specimens. However, the presence of these diagnostic features is highly variable.[3–5] Nevertheless, histology, Ziehl-Neelsen staining and *Mycobacterium* cultures should be routinely performed in suspected cases of oesophageal tuberculosis in order to maximise the diagnostic yield. Cytology and PCR have also proven to be useful in cases where the initial standard biopsies showed non-specific changes.

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In cases of secondary oesophageal tuberculosis, the diagnosis can be facilitated by confirming tuberculous infection in neighboring structures. A Mantoux or tuberculin test would be of negligible value in this situation. Given the high background prevalence of tuberculosis in our community, the positive predictive value in confirming tuberculosis would be very low. Similarly, a negative test would be unhelpful in confidently excluding tuberculosis as a diagnosis.[11]

Oesophageal tuberculosis is usually treated with anti-tuberculous drugs. A six- to nine-month course of anti-tuberculous chemotherapy is sufficient for immuno-competent patients treated with a regimen consisting of four first-line drugs, namely isoniazid, rifampicin, ethambutol and pyrazinamide for the initial two months, then continuing with isoniazid and rifampicin for another four to seven months.[3,4] Surgery is usually reserved for complications including a non-healing tracheo-oesophageal or broncho-oesophageal fistula, a recalcitrant stricture or bleeding from an aorto-oesophageal fistula.[3–5]

CONCLUSION

The patient suffered from primary oesophageal tuberculosis. Although we were unable to prove this beyond doubt, by positive culture, we feel that the histological appearances, the rapid response to treatment with anti-tuberculous medication and the patient's continued excellent health since the completion of treatment, all confirm this as the diagnosis. The patient in question was HIV negative, which serves as a timely reminder that tuberculosis often affects the immunocompetent, may crop up in unusual places and needs to be constantly borne in mind when confronted by a common symptom in an unusual setting.

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