

Dysphagia Due to Mediastinal Tuberculous Lymphadenitis Presenting as an Esophageal Submucosal Tumor: A Case Report

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Mediastinal tuberculous lymphadenitis is rare in adults, and it is even rarer for dysphagia to be the presenting symptom of mediastinal tuberculous lymphadenitis. Mediastinal tuberculous lymphadenitis with esophageal symptoms has been presented as esophageal ulceration, mucosal or submucosal mass with ulceration, fistula or sinus formation, extrinsic compression, or displacement of the esophagus. An exaggerated form of extrinsic compression may be presented as a submucosal tumor, radiologically or endoscopically. A barium esophagography of a 34 year-old woman with painful dysphagia revealed a large submucosal tumor-like mass on the mid-esophagus. The symptom was spontaneously improved over a 3-week period together with reduction of the mass size. A computed tomography of the chest disclosed an enlarged subcarinal lymph node and histologic examination of the specimen obtained by thoracoscopic biopsy brought about a diagnosis of tuberculosis. We herein report a case of mediastinal tuberculosis with unusual manifestations.

Key Words: Tuberculosis, Mediastinal lymphadenitis, Esophagus, Dysphagia

Dysphagia can be caused by extrinsic lesions compressing the esophagus such as osteophytes, thyroid diseases, and lymphadenopathy at the level of the cervical esophagus and left atrial enlargement, aortic aneurysms, congenital vascular anomalies, and various mediastinal diseases at the level of the thoracic esophagus(Shay 1991). Mediastinal diseases which can compress the thoracic esophagus and cause dysphagia include sarcoidosis(Cook *et al.* 1970), neoplasia(lung cancer, lymphoma,

or metastases)(Le Roux 1962), or mediastinal lymph nodes from inflammatory processes such as histoplasmosis(Wiersema *et al.* 1994) or tuberculosis(Dow 1981; Ghimire and Walker 1985; Damtew *et al.* 1987; Gupta *et al.* 1992; Mokoena 1992).

Tuberculosis has also rarely been reported to cause dysphagia by involvement of the esophageal mucosae, primarily or secondarily (Damtew *et al.* 1987; Rosario *et al.* 1989; Tornieporth *et al.* 1991; Mokoena *et al.* 1992; Mönig *et al.* 1995; Tassios *et al.* 1995). Among the causes of secondary esophageal tuberculosis, mediastinal tuberculous lymphadenitis has been claimed as the main cause and presented as esophageal ulceration(Damtew *et al.* 1987; Rosario *et al.* 1989; Mönig *et al.* 1995; Tassios *et al.* 1995), mucosal or submucosal mass with ulceration(Dow 1981; Damtew *et al.* 1987; Tornieporth *et al.* 1991), fistula or sinus formation(Dow 1981; Rosario *et al.* 1989; de Silva *et al.*

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1990; Mokoena *et al.* 1992), extrinsic compression(Dow 1981; Gupta *et al.* 1992; Mokoena 1992), or displacement of the esophagus(Ghimire and Walker 1985).

We recently experienced a 34-year-old housewife with dysphagia, and whose initial esophagogram revealed a large submucosal tumor-like lesion almost obstructing the esophageal lumen. The mass was spontaneously decreased in size 3 weeks later and this finding led us to suspect that the mass would be inflammatory in origin. Thoracoscopic biopsy of the lesion brought about the confirmative diagnosis of mediastinal tuberculous lymphadenitis. We herein report a case of mediastinal tuberculous lymphadenitis with unusual manifestations.

CASE REPORT

A 34-year-old female was referred to our hospital for further evaluation of a submucosal mass-like lesion seen on the barium esophagogram taken at a local hospital. She had suffered from dysphagia and odynophagia for 1 month, which led her to seek medical counseling. Despite dysphagia and odynophagia, she tolerated a soft diet. She complained of weight loss(5 kg/one month) but denied other constitutional symptoms such as fever, and night sweating. The patient was told to be admitted to the hospital. However, her admission had been delayed for 3 weeks. On admission, she said that the degree of her dysphagia and odynophagia was much improved. There was no history of fever, cough, sputum, night sweating, anorexia, use of medications, allergy, tuberculosis or exposure to it.



Fig. 1. Initial esophagogram taken at a local hospital showed a large filling defect at the mid-esophagus. The surface was smooth and the angle between the esophageal wall and the margin of the mass was almost perpendicular. This finding was more consistent with a submucosal tumor rather than extrinsic compression.



Fig. 2. Esophagoscopy performed 3 weeks after initial presentation revealed a smoothly bulging mass on the mid-esophagus(28 cm from the anterior incisor teeth). Compared with previous esophagography, the mass was reduced in size and the luminal narrowing was not so significant.

She also denied smoking, alcohol consumption, and taking contraceptive medication. Her family history was unremarkable. Upon physical examination, the patient appeared not so ill. Vital signs were stable and no lymphadenopathy was found. The examination of the lungs, heart, and abdomen disclosed no specific abnormalities.

The urine was normal. The white-cell count was 6,300/mm³ with normal differential counts. The corrected ESR(erythrocyte sedimentation rate) was 20 mm per an hour(Wintrobe method). All other routine laboratory tests revealed no abnormal findings. An x-ray film of the chest did not show any lung parenchymal change or mediastinal pathology. The barium swallow film taken three weeks before admission revealed a large protruding mass lesion on the right lateral aspect of the mid-esophagus(Fig. 1). Because the surface of the mass was smooth and the angle between the

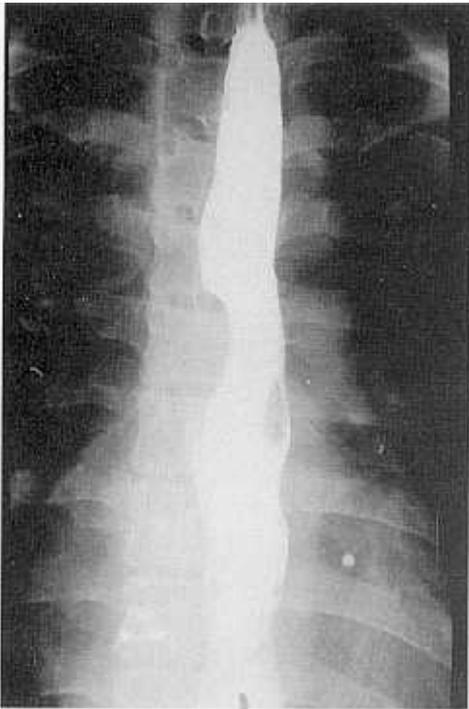


Fig. 3. A follow-up esophagography also revealed less degree of luminal protrusion and narrowing.

esophageal wall and the margin of the mass was almost perpendicular, the mass was considered a submucosal tumor of the esophagus. On the second hospital day, an upper gastrointestinal endoscopic examination revealed only a smoothly elevated lesion with firm consistency on the mid-esophagus(28 cm from the anterior incisor teeth) (Fig. 2). Compared with the barium esophagogram, the mass size and the degree of luminal narrowing seemed to be decreased. A follow-up barium swallow film obtained on the 4th hospital day also demonstrated much reduction of the mass size and luminal narrowing(Fig. 3). These dramatic changes seen on endoscopic and radiologic examinations led us to suspect this mass to be extrinsic and inflammatory. A computed tomography of the thorax taken on the 6th hospital day disclosed an enlarged subcarinal lymph node measuring 1.5 cm in diameter(Fig. 4). The esophageal wall, hilar lymph nodes, and lung parenchyme did not show any abnormalities. To obtain a tissue diagnosis, a thoracoscopy was performed on the 11th hospital day which revealed a subcarinal mass firmly adherent to the right side of the esophagus. Thoracoscopic mass excision was done with some difficulty due to surrounding fibrosis. Histological examination of the specimen showed chronic granulomatous inflammation

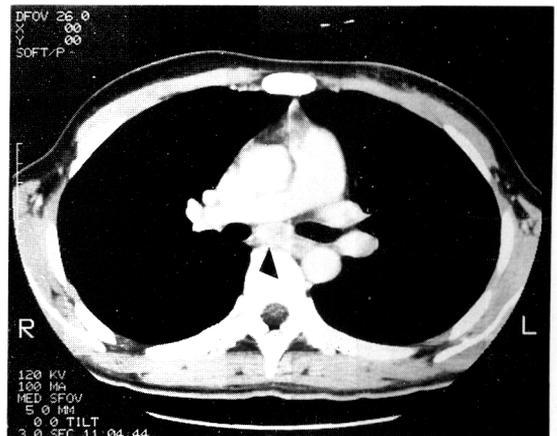


Fig. 4. A computed tomography the chest(contrast-enhanced) disclosed an enlarged subcarinal lymph node with a low-attenuation center(arrow).



Fig. 5. A follow-up esophagogram taken on the 7th postoperative day (thoracoscopy) revealed no longer a filling defect at the same level.

with caseous necrosis. Ziehl-Neelsen stain of the specimen, however, failed to disclose acid-fast bacilli. Thoracoscopic excision of the subcarinal lymph node almost abolished her symptoms of dysphagia and odynophagia.

Four regimen anti-tuberculous chemotherapy (isoniazid, ethambutol, rifampicin, and pyrazinamide) was started. A repeat barium swallow film taken on the 7th post-operative (thoracoscopy) day no longer revealed a filling defect (Fig. 5). On the 22nd hospital day, the patient was discharged uneventfully.

A follow-up barium esophagography performed on two months after discharge showed no evidence of recurrence and the patient is now in good health (1 year after discharge).

DISCUSSION

Mediastinal tuberculosis may, albeit rare,

present with esophageal symptoms (Amorosa *et al.* 1978; Ghimire and Walker 1985). Previous reports suggest that pain and difficulty in swallowing are usually due to direct esophageal involvement (Fahmy *et al.* 1969; Wales *et al.* 1976; Dow 1981; Damtew *et al.* 1987; Rosario *et al.* 1989; Mokoena *et al.* 1992). Also extrinsic compression of the esophagus without mucosal changes has been reported to produce dysphagia (Dow 1981; Gupta *et al.* 1992; Mokoena *et al.* 1992). Interestingly, Ghimire and Walker (1985) reported a case of mediastinal tuberculous lymphadenopathy who had painful dysphagia without significant involvement of the esophagus endoscopically and radiologically. They assumed that the contiguous inflammation of paraesophageal tissues resulted in disturbed esophageal motility of the patient.

Our case presented here is interesting in that her initial esophagogram showed a submucosal tumor-like intraluminal protruding mass and the mass was spontaneously reduced in size over 3-week period. Initially, we considered this mass to be a submucosal tumor rather than extrinsic compression because the angle between the esophageal wall and the margin of the mass was almost perpendicular (Fig. 1) and subsequent spontaneous reduction of the mass size made us believe that this mass would be inflammatory in origin and extrinsic in location. Cases with prominent extrinsic compression on a barium esophagography were illustrated by Dow (1981), and Gupta *et al.* (1992). However, their cases were different from ours in that the angle between the esophageal wall and the margin of the mass was wide and this finding easily suggested that these masses would be of extrinsic compression rather than intrinsic masses. Spontaneous improvement of dysphagia was also noted in the case presented by Gupta *et al.* (1992). In their case, a repeat endoscopic examination revealed an ulcerated nodular lesion which had initially been found to be only of extrinsic compression. They postulated that the caseous lymph node causing esophageal compression extruded their contents by invading the esophagus, resulting in the partial spontaneous relief of dysphagia. Also, they asserted that their case illustrated one of the

ways of esophageal involvement by tuberculosis; namely, spread from mediastinal lymph nodes. On the contrary, spontaneous relief of dysphagia in our patient was assumed to be due to the central necrosis which is a frequent finding in tuberculous lymphadenitis (Im et al. 1987) and subsequent shrinkage of the mass. If an affected lymph node ruptures into the mediastinum, fibrous mediastinitis may ensue (Goodwin et al. 1972; Dukes et al. 1976; Dines et al. 1979). Also esophageal fistulas may be produced in a similar way (de Silva et al. 1990). Confirmative diagnosis can be made by the histologic or microbiologic examinations of the specimen obtained by flexible endoscopic biopsies in cases with mucosal lesion (Dow 1981; Damtew et al. 1987; Rosario et al. 1989; Tornieporth et al. 1991; Gupta et al. 1992; Mokoena et al. 1992; Tassios et al. 1995), by mediastinoscopic (Cameron 1978) or thoracoscopic (as in our case) biopsies without mucosal lesion. There have been cases in which operation was necessary to procure the specimen (Mokoena et al. 1992). Endosonography and endoscopic fine-needle aspiration biopsy can play a role in diagnosing mediastinal masses that produce esophageal symptoms (Wiersema et al. 1994).

Most patients have been reported to be successfully treated with a three-drug anti-tuberculous chemotherapy regimen (Dow 1981; Damtew et al. 1987; Gupta et al. 1992; Tassios et al. 1995). However, four-drug regimen (isoniazid, ethambutol, rifampicin plus pyrazinamide or streptomycin) is strongly recommended in such a country as Korea which has high prevalence of primary drug resistance. The present case has been put on four-drug regimen for a year and is cured uneventfully. Surgical intervention is reserved for those patients in whom complications demand surgery, for example a mediastinal abscess seem to be unresponsive to non-operative management (Mokoena et al. 1992).

Although mediastinal tuberculous lymphadenitis is rare, it is increasing in adults (Amorosa et al. 1978; Liu et al. 1978; Bloomberg and Dow 1980). It should be included in the differential diagnoses of dysphagia and it should be borne in mind that it can present with various endo-

scopic and radiological findings even as a submucosal tumor. High index of suspicion is necessary to diagnose and treatment in time will completely cure mediastinal tuberculous lymphadenitis with or without tuberculous esophagitis.

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