

Case report

## Benign mesenchymoma of the esophagus

Erkmen Böke<sup>a,\*</sup>, Ali Sarigül<sup>a</sup>, Arzu Sungur<sup>b</sup>, Bedri Uzunalimoğlu<sup>b</sup>

<sup>a</sup>Department of Thoracic and Cardiovascular Surgery, Hacettepe University Medical School, 06100 Ankara, Turkey

<sup>b</sup>Department of Pathology, Hacettepe University Medical School, 06100 Ankara, Turkey

Received 22 May 1996; revised 25 May 1996; accepted 4 July 1996

### Abstract

Benign mesenchymoma is an extremely rare neoplasm mostly located in or about the kidney and is composed of a haphazard mixture of adult fat, fibrous tissue and tangled blood vessels, scattered nests or masses of smooth muscle cells, occasionally islands of cartilage, bone, and lymphoid tissue as well as other mesenchymal elements. Only two cases of benign mediastinal mesenchymomas were reported in the literature. In this report we describe a benign mesenchymoma of the mediastinum which presented itself with symptoms and signs of the traction diverticula of the esophagus. Copyright © 1997 Elsevier Science B.V.

**Keywords:** Esophagus; Mediastinal tumor; Mesenchymoma

### 1. Introduction

Mesenchymal neoplasms account for about 6% of primary mediastinal mass lesions. They occur in either sex and in both adults and children. Practically all histological types of mesenchymal neoplasms may be seen in the mediastinum and it is estimated that 55% of these tumors may have the chance to undergo malignant transformation [1,4]. Benign mesenchymoma of the mediastinum is an extremely rare neoplasm and only a few cases were documented in the literature [5,7].

### 2. Case report

An 18 year old woman was admitted to the hospital with complaints of cough, dyspnea and dysphagia of 6 months duration. Physical examination revealed no abnormalities, except for hypochromic microcytic anemia with a haemoglobin level of 8.8 g/dl. The laboratory findings were within normal limits.

Chest radiography showed widening of the mediastinum particularly towards the upper right region. Contrast study of the esophagus revealed a diverticula in the upper third of the esophagus. Bronchography revealed a normal bronchial tree with downward displacement in the upper lobe bronchi. Esophagoscopy confirmed the diagnosis of diverticulum of the sac which was full of food remnants and the mucosa was noted to be intact. Computed tomography revealed a large right sided mediastinal mass extending from the apex to the level of the carina with a traction diverticulum of the esophagus localised in the inner side of the mass (Fig. 1).

A right posterolateral thoracotomy through the fifth intercostal space was performed on November 21, 1988. A 7 × 8 × 5 cm mass lesion in the posterior mediastinum was identified and found to be confluent with the upper third of the esophagus, 2/3 of which was covered with esophageal muscle fibres. The mass was in the esophageal wall. There was a traction type diverticulum inside the mass, containing a full thickness of esophageal wall including the muscularis layer. The tumor was not too necrotic. The neck of the diverticu-

\* Corresponding author. Tel.: +90 312 3117377; fax: +90 312 3110233.

lum was closed and the mass was excised with an 8 cm stapler which was placed longitudinally just beneath the mass with a 36 French size esophageal rigid bougie within the lumen to prevent constriction.

A nasogastric tube was kept in place for 7 days. At the end of this period esophagography showed that the passage was completely intact without any filling defects or diverticulae.

After 24 months the patient was symptom free and without any clinical complaints.

### 3. Pathological findings

Macroscopic examination of the excised specimen consisted of a well capsulated mass measuring  $8 \times 7 \times 5$  cm in its largest diameters. It was slightly firm, rubbery in consistency. A cut surface revealed a grayish-white, slightly trabecular appearance with randomly distributed irregular soft yellow areas.

Microscopic examination of the sections taken from different parts of the mass appeared to be composed of mainly fibrous tissue and vascular elements with areas of mature fat tissue (Fig. 2). There were frequent cells with eosinophilic cytoplasm randomly distributed throughout the lesion. Although cross striations were detectable in H + E sections phosphotungstic acid-hematoxylin (PTAH) stain demonstrated the presence of this striated muscle cells within the lesion.

### 4. Discussion

Any mass composed of at least two unrelated mesenchymal elements other than fibrous tissue is referred to as mesenchymoma. The benign variant of the mesenchymal tumors is most often located in or about the



Fig. 1. Computed tomography revealed a large right sided mediastinal mass extending from the apex to the level of the carina with a traction diverticulum of the esophagus localised in the inner side of the mass.



Fig. 2. Benign mesenchymoma composed of fibrous, mature fat and muscle tissue (H + E  $\times$  230).

kidney and is composed of a haphazard mixture of adult fat, fibrous tissue and tangled blood vessels through which are scattered nests or masses of smooth muscle cells, occasionally islands of cartilage, bone, and lymphoid tissue as well as other mesenchymal elements. Because all are extremely mature, these lesions have been interpreted by some as hamartomas rather than neoplasms [5]. As the presented tumor contained more than two unrelated mature mesenchymal elements other than fibrous tissue (fat, blood vessels and striated muscle) it was classified as benign mesenchymoma. Irregular distribution of the tissue elements, particularly the muscle cells suggest that this could also be a hamartomatous lesion composed of mature mesenchymal elements most probably representing a developmental abnormality rather than a true neoplasm. These two terms are used interchangeably in the literature.

No morphological evidence of malignancy was found in our case. The malignant counterpart is referred to as malignant mesenchymoma [2,5]. These rarely encountered tumors are seen in soft tissue, mainly in the extremities and occasionally in the heart, liver and esophagus. Both adults and children are affected and extremely aggressive infiltrative lesions occasionally metastasize and are directly responsible for death in over 50% of adults and about 40% of children. The basic pathological characteristics of malignant mesenchymoma is the presence of different degrees of cellular anaplasia of the different mesenchymal components making up the neoplasm such as liposarcoma, rhabdomyosarcoma, fibrosarcoma, chondrosarcoma and so forth [5].

Two cases of benign mediastinal mesenchymoma were reported previously. One case merits mention. He died because of the widespread infiltration of the tumor into the pleura, the esophagus, the lungs, and beneath the diaphragm in the retroperitoneal area to the level of the renal arteries, and was too extensive for complete excision by the time of surgery. Although the tumor was histologically benign, by virtue of its local infiltra-

tion, biologically it must be considered locally malignant [5].

Symptoms of the mediastinal mass lesions are frequently nonspecific in nature [3,6]. Our patient presented with signs and symptoms referring to esophageal function, and barium studies misled to the diagnosis of the mass but not the diverticulum. Computerized tomography showed that there was a paraesophageal solid mass with a traction diverticulum localised in the inner side of the mass.

Benign mesenchymoma is a rather rare lesion in this location, and a benign mesenchymoma of the mediastinum presenting itself as a diverticulum of the esophagus has not previously been documented. This well capsulated mass and the diverticulum were totally excised and the patient was free of any complaints 24 months after surgery.

## References

- [1] Everett C, Volberg F, Ulshen M, Murray G. Intramural esophageal hamartoma. *Gastrointest Radiol* 1980;5:317–319.
- [2] Maratake J, Jimi A, Morie A, Inokuma T, Ohno M. Malignant mesenchymoma of the esophagus. *Acta Pathol Jpn* 1994;34:925–933.
- [3] Mardy JD, Ewing MP. The mediastinum. In: Glenn WWL, editor. *Thoracic and cardiovascular surgery*, fourth ed. Norwalk, Connecticut: Appleton Century Crafts, 1983:181–208.
- [4] Oakley RM, Carty M, Cudmore RE. Multiple benign mesenchymomata of the chest wall. *Pediatr Radiol* 1985;15:58–60.
- [5] Pachter MR, Lattes R. Mesenchymal tumors of mediastinum. *Cancer* 1963;16:74–94.
- [6] Payne S. Esophageal diverticula. In: Shields TW, editor. *General thoracic surgery*, second ed. Philadelphia: Lea and Febiger, 1983:859–870.
- [7] Weidner N, Cruz DS. Phosphaturic mesenchymal tumors. *Cancer* 1987;59:1442–1454.