

## Resection of a large benign pharyngo-oesophageal submucous mesenchymoma via laterocervical approach

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### Abstract

Two rare cases of benign submucosal pharyngo-oesophageal mesenchymoma are presented in this paper. One patient was treated by tumour removal via a combined thoracic and laterocervical approach and the other by resection through a laterocervical approach. The paper discusses the pathology and diagnosis of benign mesenchymomas. The authors suggest that for large tumours located in the pharynx and extending down the oesophagus without adhesion to the oesophageal wall, the laterocervical approach can avoid complications associated with the thoracic approach. The new technique caused less tissue damage and provided a quicker recovery and shorter hospitalization.

**Key words:** Mesenchymoma; Pharynx; Oesophagus; Surgical Procedures, Operative

### Introduction

The large benign mesenchymomas that originate at the pharyngo-oesophageal junction are rare and tend to be misdiagnosed. The diagnosis and treatment of the disease are difficult. It is generally accepted that a thoracotomy be performed for huge tumours in the chest cavity, even though this approach causes substantial injury to the patient. Presented in this paper are two cases of benign submucosal pharyngo-oesophageal mesenchymoma treated in our hospital, each case being treated differently.

### Case reports

#### Case 1

A 59-year-old male complaining of dysphagia over a six-month period was admitted to hospital. The patient said his discomfort had worsened progressively and was associated with a 25 day history of tarry stools. Physical examination revealed no abnormality in the cervical lymph nodes, heart, lungs or abdomen. The patient's height was 162 cm and weight 38 kg. He was extremely emaciated and anaemic. Laboratory tests yielded the following: RBC:  $2.53 \times 10^{12}/L$ ; HGB: 85 g/L; WBC:  $8.0 \times 10^9/L$  (N 0.57, L 0.42); ESR: 10 mm/h; GPT: normal; stool and urine test: normal; occult blood: (+). B-mode ultrasonic examination found no abnormality in the liver, spleen, or kidney. A barium swallow indicated a snowflake-like pattern in the middle-lower part of the oesophagus. The diameter of the dilated segment, extending from the thoracic inlet to the diaphragm was up to 7 cm. The cardia was narrowed. On the basis of the findings, a tentative diagnosis of cardiospasm was made. The fibre-optic gastroscope could not be introduced into the stomach due to the obstruction caused by a large tumour. Biopsy of the tumour suggested 'oesophageal leiomyoma'. The patient was then transferred to the Department of Thoracic Surgery. A thoracotomy and tumour resection was performed under

general anaesthesia. A cucumber-shaped mass with intact mucosa was found within the oesophageal wall. The mass was attached to the pharynx by a pedicle with no adhesion to the oesophageal wall. The endoscopy revealed a pedicle of 6 cm  $\times$  5 cm  $\times$  3 cm in size based in the left pyriform sinus of the pharynx and left entrance of the oesophagus, with dilated mucosal vessels.

A two step procedure was taken. First the tumour within the oesophagus, measuring 25 cm  $\times$  8 cm  $\times$  3 cm (Figure 1), was removed by thoracic surgery. Then the pedicle was resected using a YAG-laser through a lateral pharyngotomy approach (Figure 2). The post-operative pathological examination revealed that both the pedicle and body of the tumour were a benign mesenchymoma. While the pedicle contained more fibro-adipose tissue, the body showed polypoid degeneration. An immuno-histochemical study yielded negative results for myelin basic protein and S-100 protein, and positive staining for vimentin.

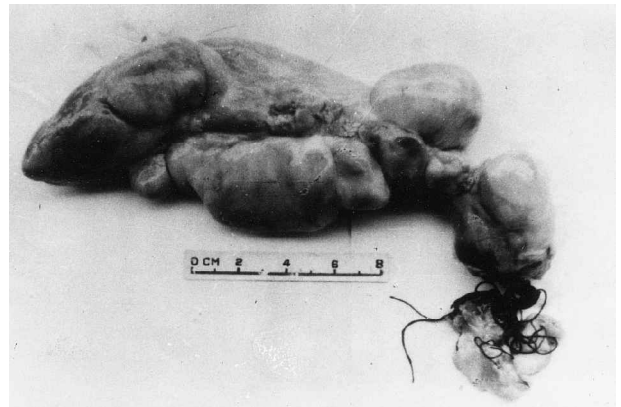


FIG. 1

The body of the large benign mesenchymoma, measuring 25 cm  $\times$  8 cm  $\times$  3 cm (from patient 1).

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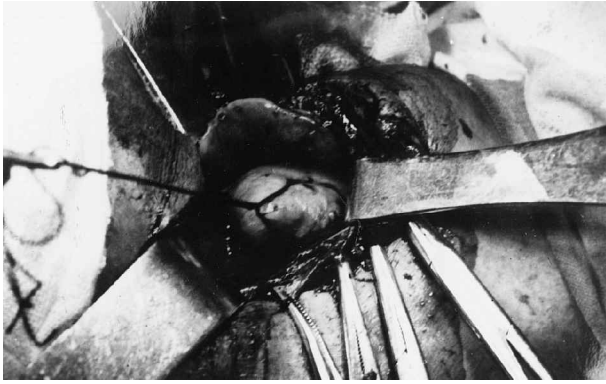


FIG. 2

The pedicle of the large mesenchymoma was exposed via the path of lateral pharyngotomy (from patient 1).

Two weeks after surgery, the patient could eat without assistance and recovered gradually. Forty-eight days later, he was discharged with a net weight gain of three kg. A six-year follow-up did not indicate any recurrence. The patient's body weight had increased from the original 38 kg to 52 kg.

#### Case 2

A 63-year-old man was admitted with an eight months' history of dysphagia, that had become much worse in the 15 days before admittance. Physical examination showed that the patient at a height of 170 cm and weighing only 45 kg was extremely emaciated. Indirect laryngoscopy found a pinkish, thumb-sized neoplasm in the pharynx. Contrast radiography of the oesophagus revealed that the oesophagus was dilated with a strip-like tumour extending from the top of the oesophagus to the cardia. A chest magnetic resonance image (MRI) showed an oval-shaped smooth-edged mass, measuring 7.5 cm at the widest part. The tumour was located within the posterior mediastinum from the level of the sixth cervical vertebra level to the upper edge of the diaphragm. The trachea and aortic arch were shifted laterally (Figure 3(a) and (b)). All of these findings suggested a benign oesophageal tumour. Fibre endoscopy found a massive, pink-coloured, soft, tumour from the pharynx down to the cardia and without adhesion to the oesophageal wall. A biopsy demonstrated a benign mesenchymoma. After one week of supporting treatment, resection of the tumour was undertaken via a laterocervical approach. The surgery confirmed that the tumour pedicle arose from the left wall of the laryngopharynx and extended to the entrance of the stomach. The entire tumour was lobulated, measured about 40 cm in length and was not adherent to the oesophagus. The tumour, weighing 695 g, was completely removed (Figure 4). After histopathological examination, a final diagnosis of a huge benign pharynoesophageal mesenchymoma was established (Figure 5). One week after the operation, the patient was able to swallow food and achieved a quick recovery. He was discharged with no discomfort 15 days after the operation. There was no recurrence during two and a half years of follow-up.

#### Discussion

Benign mesenchymomas are tumours with a high degree of differentiation. The tumour tissues mimic the original tissues in structure, cellular morphology and colour. The tumour is usually free of integument, or has only incomplete integument, and tends to adhere to adjacent

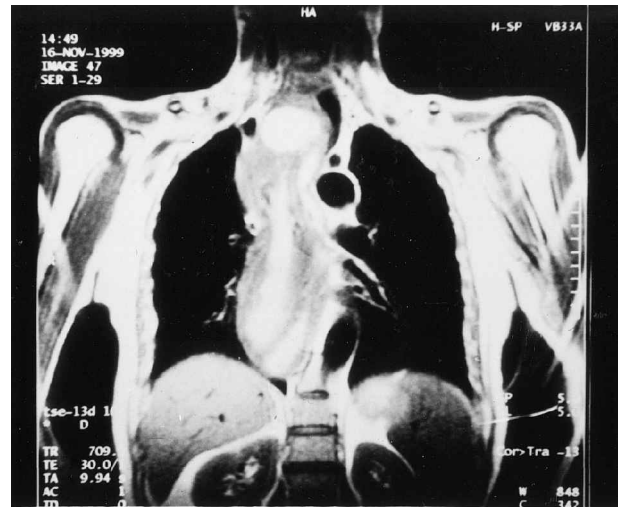


FIG. 3

Magnetic resonance imaging (MRI) from patient 1. MRI shows a large tumour in the oesophagus.

tissues. A cross-section of the tumour appears yellowish or dark red and greyish in colour. The tumour can consist of adipose cells, smooth muscle, blood vessels, striated muscle, cartilage, bone, mucous tissue, lymph tissue, and haematopoietic tissues. Usually, a mixture of two or more of these components is present in different proportions and clinically no two structurally identical benign mesenchymomas have ever been found.<sup>1-3</sup>

The tumour in *Case 1* measured 31 cm × 8 cm × 3 cm, and its pedicle was based in the laryngopharynx and the oesophageal junction. The tough pedicle was yellow-white in colour and rich in fibrous connective and adipose tissue. It presumably originated from the submucosal fibrous and adipose tissues of the laryngopharynx. Rupture and bleeding of mucosal vessels on the tumour surface, caused by inflammation and compression, contributed to the presence of blood in the faeces.

The size of the tumour in *Case 2* was 40 cm × 7.5 cm × 5 cm, and its pedicle, like that in *Case 1*, was attached to the laryngopharynx and oesophageal entrance. Structurally, it grew outward and was lobulated with a soft and fibropolypus-like texture. A number of plasmocytes and adipose cells were observed on microscopy, along with smooth muscle cells and vessels. The tumour growth was irregular with no signs of nerve fibres. The number of plasmocytes, adipose cells, and amount of fibrous tissue was greatest at the furthest part of the tumour away from



FIG. 4

The tumour, 40 cm × 7.5 cm × 5 cm, its form appears lobulated, and its tail presents black-brown colour because of gastric acid corrosion (from patient 2).

the pedicle. The compression by the tumour caused the smooth muscle fibres to become less dense and resulted in the relaxation and enlargement of the oesophageal cavity. The non-invasive growth of the tumour left the oesophageal mucosa intact.

A benign mesenchymoma appearing in the area of the laryngopharynx and the oesophagus is clinically rare. This kind of tumour originates from the muscular layer or the submucosa and is generally single, strip-like, and lobulated. By the time the symptoms such as difficulty in swallowing, nausea, dyspnoea, emaciation, and anaemia become apparent, the size of the tumour is already remarkable.<sup>5,8</sup> X-ray examination is helpful in the identification of the disease and a pathological biopsy is necessary for confirmation of the diagnosis. Because the disease is clinically uncommon, it tends to be misdiagnosed. In *Case 2*, the enlarged oesophagus and smooth interior wall as revealed by X-ray examination, led to the misdiagnosis of 'mega-oesophagus'. An accurate diagnosis is not difficult, but differential diagnosis should be carefully conducted to exclude such diseases as leiomyoma, lipoma, and malignant mesenchymoma, which microscopically often shows abnormal karyokinesis.<sup>7,9</sup> X-ray examination presents the benign mesenchymoma as a smooth mass without a filling-defect in the oesophageal wall.<sup>6</sup> In *Case 1*, a soft fibre-scope was unsuccessfully used for examination of the oesophagus and a satisfactory biopsy was not obtained, thereby resulting in a wrong diagnosis of oesophageal leiomyoma. With a correct procedure, which includes a rigid endoscope for the examination of the oesophagus, plus other examinations, a correct diagnosis can be made at an early stage of the treatment. If that had been achieved, a thoracotomy could have been avoided.

Generally speaking, a huge tumour located within the oesophagus is removed via an open thoracotomy approach, which causes significant morbidity and involves unwanted complications that leave the patients feeling very uncomfortable.<sup>10,11</sup> A benign massive mesenchymoma with a pedicle based in the laryngopharynx or the oesophageal entrance can be completely removed via a cervical approach as long as the tumour body has no attachment to the interior of the oesophageal wall. The pharyngostomy exposes the tumour pedicle, but the recurrent laryngeal nerve should be protected. Separation and exposure of the cervical oesophagus should be performed as posteriorly as possible so that the cervical oesophagus can be dissected vertically without damaging the other laryngeal nerve. While the tumour is being stripped and removed, strong force should be avoided,

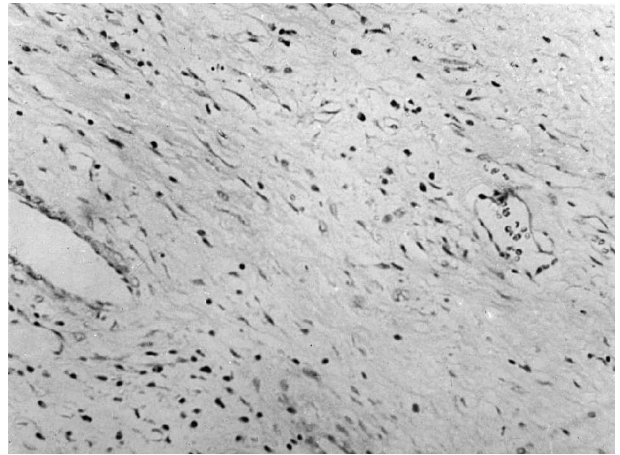


FIG. 5

Histological sections from patient 2. There are smooth muscle tissue, vessel and lipocytes in the tumour (H & E; ×100)

otherwise, the tumour might rupture. Despite the tumour pedicle having no integument, its morphology and tissue texture are still different from that of normal tissue.<sup>1,4,8</sup> Completely removing a tumour of this type is the key for reducing the likelihood of recurrence.

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Lisi Liu, M.D. takes responsibility for the integrity of the content of the paper.

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