

## Case Report

### **Esophageal Haemangioma: A case report and review of literature**

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#### **Abstract**

A 28-year-old female presented with the history of dysphagia to liquids. Initial evaluation through endoscopy was unremarkable and biopsy was reported negative for malignancy. Re-biopsy revealed a stricture like esophageal growth at 23cm from upper incisors, causing narrowing of esophagus that raised suspicion of malignancy. Microscopic examination revealed dilated blood vessels lined by flattened endothelial cells. Diagnosis of esophageal cavernous haemangioma was made. Immunohistochemistry (CD34) confirmed the diagnosis. Endoscopic resection of tumor is being planned for the patient.

#### **Introduction**

Esophageal Haemangioma is a very rare benign tumor of esophagus, which can mimic malignancy. Even though it is benign, it is very important because it can result in fatal complications, such as severe haemorrhage.<sup>1</sup> Fewer than hundred cases have been reported in the literature. To date and to the best of our knowledge this is the first case report from our region.

#### **Case Report**

A 28-year-old young female patient presented with complaints of dysphagia for last 6 months. Patient stated that dysphagia was only present for liquids; there was no dysphagia to solids. Patient belonged to a rural area, and was assessed by local General Practitioner and Endoscopy with multiple biopsies was done in a nearby hospital. Reports were negative for malignancy but no further details were provided. Due to deteriorating general health and progressively worsening symptoms, patient had a re-biopsy done after a few days. She did not report any appreciable change in appetite, odynophagia or melena. Weight loss was documented as 2kg in last 3 months. Patient's past medical history and family history was unremarkable. Initially her barium swallow was done which revealed a lesion obstructing esophagus around the middle part in the form of stricture. Endoscopic examination was performed later; which revealed a stricture like submucosal, esophageal growth, measuring 2.4 x 1.3cm in the esophagus, located at 23cm from the upper incisors. It was bleeding profusely to touch. Biopsy specimen was taken and sent to Shifa International Hospital for histopathology.

Biopsy specimen consisted of three, grey tan, soft

tissue fragments that measured 0.8 x 0.5 x 0.3cm in aggregate. Microscopic examination was done; sections showed fragments lined by stratified squamous epithelium. Lamina propria showed irregular dilated blood vessels lined by flattened endothelial cells. Focal chronic infiltrates were also seen. No evidence of atypical mitosis or malignancy was seen. Diagnosis of "Esophageal cavernous haemangioma" was made. CD 34 was done later, which confirmed the diagnosis.

#### **Discussion**

Esophageal Hemangioma is a rare benign tumour of Esophagus; fewer than hundred cases have been reported in the literature.<sup>1</sup> It has been described in literature for more than 5 decades.<sup>2</sup> Patterson et al,<sup>3</sup> reported one case of esophageal Haemangioma. Out of 19982 autopsies performed in New York Medical College over 10 years time, only 3 cases of esophageal Haemangioma were noted.<sup>3</sup> Review of 106 cases of vascular gastrointestinal tumours from the Mayo Clinic revealed only two cases of haemangioma of esophagus.<sup>3</sup>

Osler-Weber-Rendu is a hereditary disorder, that can result in formation of multiple esophageal haemangiomias<sup>4</sup> but our patient did not have such a family history. Blue rubber bleb nevus syndrome is another vascular disorder affecting gastrointestinal tract and can present with bleeding. It was initially thought to be related to haemangioma, but later it was considered as a venous malformation.<sup>5</sup>

Regarding age of the patients, most of them being diagnosed with cavernous esophageal haemangioma, were above 40 years of age. Our patient was 28 years old; which is considerably young age in comparison to the average age of presentation of disease. The range of patient's ages usually ranges from newborn to 72 years, peaking in the fourth decade for both sexes; males reach another peak in the sixth decade.<sup>6</sup>

Esophageal Haemangioma is particularly common in Japan due to unknown reasons. Up to a decade back 67 cases were reported in Japan alone on esophageal haemangioma. Three cases of pediatric post-cricoid and esophageal haemangioma have also been identified, but the primary lesion was in post-cricoid region, extending into the upper esophagus.<sup>7</sup>

Clinical presentation in patients includes obstructive symptoms such as dysphagia and odynophagia. Dysphagia can be both to liquids as well as solids. The most common complaints are dysphagia (45.2%); followed by haematemesis in 25.8%, malaena in 12.9% and retrosternal pain 12.9%.<sup>6</sup> As patients usually present in middle or old age, it can strongly mimic malignancy as well. If left undiagnosed, patient may present with complications such as severe haematemesis.<sup>1</sup> Therefore, timely diagnosis and treatment is of utmost importance. Esophageal carcinoma can also rarely co exist with Haemangioma;<sup>8</sup> which further highlights the need of early diagnosis.

Araki and associates<sup>1</sup> have shown that esophageal haemangioma can be biopsied without serious consequences;<sup>1</sup> nevertheless the possibility of major haemorrhage still exists. Esophageal biopsy is usually not recommended; unless advanced technology such as CT scan is not available. Computed Tomographic findings of an intramural mass; with marked enhancement, following intravenous contrast enables the diagnosis to be suggested pre-operatively.<sup>9</sup> But it is advanced technology requiring highly skilled radiologists for interpretation, which is difficult in remote areas.

As stated earlier, biopsy is not a recommended test for diagnostic evaluation of esophageal haemangioma due to risk of bleeding, but in our patient biopsy was done twice due to a high suspicion of malignancy and unavailability of computerized tomographic scan. She did not suffer any complications but if bleeding would have occurred, it could result in fatal consequences.

Another study based on 30 case reports of esophageal haemangioma revealed that mostly haemangiomas favour upper portion of esophagus.<sup>6</sup> In our patient it was located at the junction of upper and middle esophagus.

For the treatment, many options are available but

minimal surgical techniques are usually appreciated.<sup>6</sup> First choice of treatment for esophageal haemangioma should thus be an endoscopic resection; if the tumour is located within the mucosal or submucosal layer.<sup>1</sup> However, if it is impossible to resect endoscopically, then either endoscopic injection sclerotherapy (EIS) or a surgical resection should be considered.<sup>1,7</sup> Another, study suggests that superficial tumour sizes less than 2.5 cm in diameter, with a thin pedicle may be suitable for endoscopic resection. Surgical resections are still the main option for most physicians.<sup>6</sup> Newer advanced techniques include fulguration using potassium titanyl phosphate/yttrium aluminum garnet laser.<sup>10</sup> Our patient was a good candidate for endoscopic resection of the haemangioma and the procedure was performed. Her post operative course was unremarkable.

## References

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