Case Report

Angiolipoma of Hypopharynx

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Abstract

Pedunculated angiolipoma is a rare benign tumor of upper aero digestive tract. We report a very rare case of pedunculated angiolipoma arising from hypopharynx which presented as a mass protruding outside the oral cavity. As the mass came out of mouth, patient was unable to close her mouth and suffered from difficulty in taking food. The mass was removed per orally under general anaesthesia and the patient was discharged in excellent condition. She is still under follow up and no recurrence since May 2012. Few cases are reported in literature but such a presentation is uncommon.

Key words: Angiolipoma, Hypopharynx;

Introduction

Lipoma is the one of the common benign tumors accounting for 4% to 5% of all benign tumors. Angiolipoma is a variant of lipoma according to WHO classification of soft tissue

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tumors. ² When vascular component is prominent in lipomatous lesion, the term angiolipoma is used. Angiolipoma is not common in head neck region. According to authors on 2010 only 38 cases were reported.³

We are presenting a case of huge pedunculated angiolipoma arising from hypopharynx and protruded outside the mouth.

Case report

A 48 years old Bangladeshi female of low socioeconomic status presented with a mass coming out of her mouth for two days which suddenly assumed a big size. She noticed a small mobile mass inside her mouth for about 10 days. When the mass protruded out of mouth she became unable to close her mouth; eating and drinking was difficult. She was suffering from throat discomfort and foreign body feeling for near about one year and was treated with antihistamines, antibiotics and anxiolytic drugs but did not get relief of symptoms. She had no history of pain, dysphagia, respiratory distress,

dysphonia, per oral bleeding or weight loss. She was nonsmoker with unremarkable medical history.

On examination, the mass was firm, smooth, non tender and reddish in color. It was freely mobile due to its large stalk arising from hypopharynx. No attachment was seen in the oral cavity. Oral cavity was normal. There was no palpable lymphnode in the neck. Fiber optic laryngoscopy and CT scan revealed the origin of mass from posterior wall of rt. piriform fossa. Right Supraglottic region was found edematous. Vocal cords and other parts of larynx were normal. Air way was adequate. FNAC report, "Smears show hemorrhagic material containing scattered inflammatory cells. No malignant cell is seen."

During the preoperative period we introduced Nasogastric tube and started I/V antibiotics. Patient was prepared for surgery. Per oral exicision of mass was done under general anesthesia with endotracheal intubation. Endotracheal intubation was not difficult. The pedicle was clamped with 'Negus Forceps', cut and legated. Per operative finding was that pedicle of the mass was attached to the posterior part of medial surface of right piriform sinus adjuscent to arytenoid cartilages. Other part of piriform fossa, aryepiglottic fold and larynx was normal. Length of the mass with stalk was 16 cm. Postoperative state was uneventful. Nasogastric feeding was started 3 hours later and Nasogastric tube was removed after 24 hours. Patient was discharged in excellent condition.

Discussion

Lipomas are the most common neoplasm arising from fat tissue. 13% of all lipomas occur in head and neck, including cheek, tongue, palate, parotid gland, neck and larynx.⁴ There are some variants of lipoma which are often called 'fibrovascular polyp'. A qualifying prefix "fibro" can be used for

lipomas possessing an unusually prominent connective tissue component. The term 'fibrovascular polyp' has been recommended by WHO for polyps termed as fibroma, fibromyxoma, fibroepithelial polyps, fibrolipoma or angiolipoma.⁵

If the vascularity of the lesion is much greater than that of simple lipoma, the designation 'angiolipoma' is appropriate.⁶

Approximately 85%–90% of the polyps of upper aero-digestive tract arise from the proximal end of the esophagus where the mucosa is thin and pliable.^{7,8}

They are usually pedunculated. The remaining 16% had their pedicle based in the hypopharynx.9 In our case was the pedicle arose from hypopharynx. Fibro vascular polyps are postulated to originate from two areas of lower resistance in the pharyngeal musculature. One is between the superior and inferior cricopharyngeal muscle called Killian's dehiscence, the other is between the inferior cricopharyngeus muscle and the proximal end of the esophagus called the Laimer's triangle where the muscular support of the esophageal wall is relatively poor. In these areas, nodular submucosal thickenings of submucosal folds may evaginate through the mucosa.10

According to WHO, histologically angiolipoma consist of two mesenchymal elements: mature adipocytes and branching capillary sized vessels which are thin walled and usually contain fibrin thrombi. The relative proportion of adipocytes and blood vessels varies. Angiolipoma is encapsulated tumor. A non-encapsulated tumor mass which extend into the surrounding tissue which are previously called 'infiltrating angiolipoma'^{3,11} such as Spinal angiolipoma and intramuscular haemangioma are now regarded as different lesions according to WHO².

Pathogenesis of angiolipoma is not clear. There are different theories and possible causes include trauma, familial incidence², fatty metamorphosis of a central hemangioma, hyperplasia of fat with increased proliferation of vascular component, vascular proliferation in a congenital lipoma, hormonal change during puberty. It might be a true neoplasm. It is suggested that Mast cells may have a role in angiogenesis as they produce VEGF (vascular endothelial growth factor) and found at a greater amount in angiolipoma than that of lipoma. ^{2,3,12}

Fibrovascular polyp or angiolipoma may remain asymptomatic for years until it reaches a large size. The most common complaints include dysphagia and sensation of a mass and no other symptom. Often they are treated as globus as in case of our patient. Other symptoms are odynophagia, dysphonia and choking sensation which is relieved by swallowing a soft structure. Patient may give history of regurgitation of polyp into mouth which disappear on swallowing. When the polyp twists it leads to haemorrhage and necrosis of the lesion. In our case regurgitated polyp came out of mouth and attained a large size within short time probably due to twisting and internal haemorrhage. 13

Diagnosis of angiolipoma in hypopharynx or fibrovascular polyp of esophagus is difficult. Fibre optic laryngoscopy and CT Scan was used to confirm the origin of the pedicle in our case. Barium contrast study and esophagogastroscopy are very commonly used and are useful. Unless regurgitated, the presence of such a lesion is difficult to diagnose, and 30% of the patients may die without a correct diagnosis.¹⁴

Surgical removal of the polyp is recommended treatment option. These should be removed as soon as possible because of the progressive nature of the lesion and the

underlying risk of asphyxiation and sudden death specially in case of polyp arising from hypopharynx. Many approaches have been used to remove such polyps until now considering the mass, pedicle, origin and extension. They are per oral, lateral pharyngotomy, cervical esophagostomy, Endoscopic resection and resection using CO2 laser. Per oral removal for polyps in oropharynx and hypopharynx was found common in literature. Jun W et al suggest that In general, small polyps less than 2 cm in diameter and with a thin pedicle can be removed by ligation or electrocoagulation. Polyps larger than 8 cm long or those with a thick, richly vascularised pedicle can be excised by cervical incision. When the polyp is too large to be removed through pharyngotomy, removal via gastrostomy is recommended. 13

Conclusion

Pedunculated angiolipoma is a rare benign tumor of hypopharynx. Local excision is curative and recurrence is rare.

References

- 1. Wakely C. Sommerville P. Lipomas. Lancet 1952; ii: 995-9.
- Fletcher C.D.M., Unni K.K., Mertens F. (eds.): World Health Organization Classification of tumors. Pathology and Genetics of Tumors of Soft Tissue and Bone. IARC Press: Lyon 2002. Available from:http://www.iarc.fr/en/publications/ pdfs-online/pat-gen/bb5/bb5classifsofttissue.pdf
- Arenaz Búa J, Luáces R, Lorenzo Franco F, García-Rozado A, Crespo Escudero JL, Fonseca Capdevila E, et al. Angiolipoma in head and neck: Report of two cases and review of the literature. Int J Oral Maxillofac Surg. 2010;39: 610–5.

- Altug HA, Sahin S, Sencimen M, Dogan N, Erdogan O. Non-infiltrating angiolipoma of the cheek: A case report and review of the literature. J Oral Sci. 2009;51:137–9. [PubMed]
- Jass JR, Sobin LH, Watanabe H. The World Health Organization's Histologic Classification of Gastrointestinal Tumors. A cinnentary on the 2nd ed. Cancer 1990; 66: 2162-2167.
- Batsakis JG.ed. Soft tissue tumors of the head and neck: unusual forms. In: Tumors of the head and neck. 2nd ed. Baltimore: W B Saunders Co. 1979: 360-4.
- 7. Patel J, Kieffer RW, Martin M, Avant GR. Giant fibrovascular polyp of the esophagus. Gastroenterology 1984; 87: 953-956.
- 8. Vrabec DP, Colley AT. Giant intraluminal polyps of the esophagus. Ann Otol Rhinol Laryngol 1983; 92: 344-348.
- Caceres M, Steeb G, Wilks SM, Garrett
 HE Jr. Large pedunculated polyps
 originating in the esophagus and

- hypopharynx. Ann Thorac Surg 2006; 81: 393-396.
- Paczona R, Ivan L, Jori J, Ivanyi B. Peroral endoscopic removal of a regurgitated giant polisegmented fibrovascular polyp of the esophagus. Diagn Ther Endosc 2001; 7: 197-201.
- 11. Gonzales-Crussi F, Enneking WF, Arean V. Infiltrating angiolipoma. J Bone Joint Surg. 1966;48:1111–4. [PubMed]
- Ida-Yonemochi H, Swelam W, Saito C, Saku T. Angiolipoma of the buccal mucosa: A possible role of mast cell derived VEGF in its enhanced vascularity. J Oral Pathol Med. 2005; 34:59–61. [PubMed]
- Jun W, De-min H, NI Xin, Li-jing M, Jingying Y,Yang X. Fibrovascular polyp of the hypopharynx and esophagus. Chinese Medical Journal 2011; 124(19): 3182-3184.
- 14. Timmons B, Sedwitz JL, Oller DW. Benign fibrovascular polyp of the esophagus. South Med J 1991; 84: 1370-1372.