

Dyspnea and Dysphagia as First Sign of Hypopharyngoesophageal Lipoma

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ABSTRACT

We present a rare case of a 72-years old male with a big lesion coming out from his mouth suddenly. Examination showed a mucosal lesion seen outside of the mouth, saliva running outside, he was not able to swallow, it was hard to speak and breathe for him. At the time of arrival the patient was stable, later developed a distress and dyspnoea because of the part of tumor inside of his mouth was moving in and out on breathing and was obstructing his airway. This situation required an urgent tracheostomy to secure the patient's airway. After tracheostomy we continued with endaural approach technique to remove the tumor, the origin of lesion was identified within the postcricoid area. Lesion was removed safely, sent to histology with no complication after. Results came back as a lipoma pedunculated. Literature search did not reveal any cases of limb lipoma presenting with a sudden hanging tumor causing airway compromise and dysphagia, all cases were diagnosed during routine examination. Our case has proven that hypopharyngoesophageal lipoma can present as an acute condition and we have to be able to save and manage patients like this.

Keywords: Lipoma, limb lipoma, pedunculated lipoma, acute distress syndrome, dysphagia

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INTRODUCTION

Gastrointestinal tract inflammatory fibroid polyp is an uncommon, benign, submucosal lesion. Ward "lipoma" first was mentioned by Oslen in 1709. The „gastrointestinal tract lipoma " was described in 1949 by Vanek. „Oral lipomas" were used by Roux in 1884, he described it as a „yellow tumor" [1,2]. Lipoma is a benign tumor derived from adipose tissue. It is a common mesenchymal tumor typically coming from the fat tissue, most commonly from the subcutaneous tissue. Microscopically: well-defined, encapsulated floating, on touch brittle tumor. Tumor can have a lobulated shape, which is then appears huge. Lipoma can be classified as superficial or deep. Variation of the superficial lipoma - „lipoma pendulum" - a floating, polypoid tumor typical for the digestive tract and skin [3,4].

Benign tumors of the esophagus are very rare, about 0.5-0.8 % of all esophageal neoplasms. Approximately, 60 % of benign esophageal neoplasms are leiomyomas, 20% are cysts, 5% are polyps, and less than 1 % are lipomas [5]. The upper third of the esophagus is the most common site of origin for digestive tract lipoma. Men present with this lesion more frequently than females. Lipoma can be a random finding on imaging or endoscopic examination. More than 85 % are asymptomatic [3]. Huge size lipoma can cause airway obstruction, dysphagia, foreign body sensation, regurgitation and even pain [6]. Endoscopic techniques, images - CT, MRI can be used for diagnosis. Gastroesophageal examination may be complicated by insufficient esophageal lumen, which can be partially or completely filled with tumorous mass [7]. On CT, the

lipoma appears as a homogeneous hypodense well-defined mass. CT is beneficial for lipoma diagnosis, but for soft tissue structures MR imaging is a gold standard [8]. Indication for surgical intervention is: symptomatic patient, suspicious of malignancy or possible transformation, but it is very rare [5,7].

CASE PRESENTATION

During on-call duties, we had a phone call from ENT colleagues from another ENT department about 72- years old patient with a tumor hanging out from his mouth, this patient has an intermittent stridor. An urgent CT was performed and showed a pedunculated polypoid mass size about 41x33 mm, 22x14 mm out of mouth, other part of mass within the oral cavity about 30x20 mm. Mass origin is likely from the oro-hypopharyngeal wall, or esophageal wall. Esophageal lumen was dilated, the epiglottis was depressed by mass anteriorly partially obstructing glottis (Figures 1, 2 and 3).

Patient arrived within an hour by ambulance, was asymptomatic, stable. Patient has a history of not being well last night, starting to cough, to drool, to vomit. We suppose that during vomiting mass has come out from his mouth and he got worse with his breathing. On examination a polypoid mass was hanging out from mouth (Figure 4), saturation started to drop, sequently, patient transferred to the intensive unit care to control his breathing. Patient

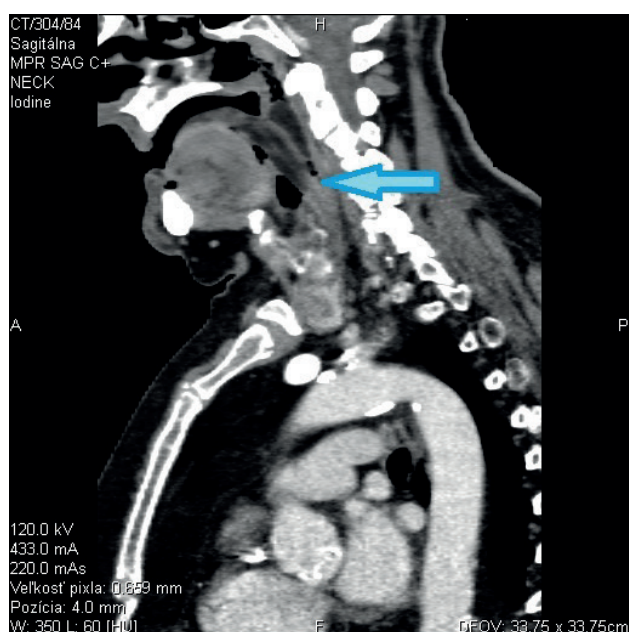


Figure 1. Sagittal CT scan of the tumor (marked by arrow).

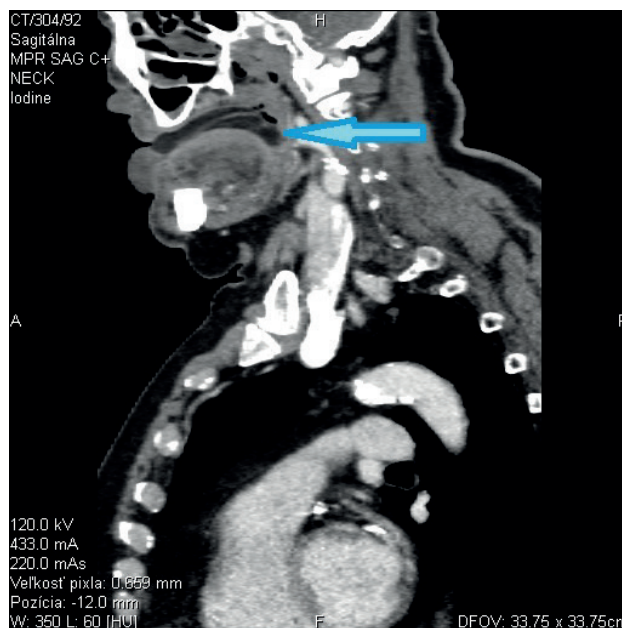


Figure 2. Sagittal CT scan of the tumor (marked by arrow).

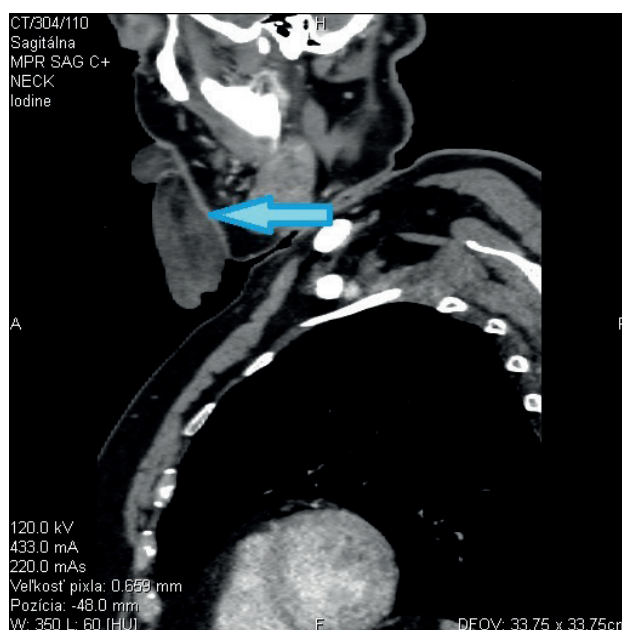


Figure 3. Sagittal CT scan of the tumor (marked by arrow).

became symptomatic, breathless, saturation went down. The floating mass in the oral cavity was seen, lesion was pulled out from the mouth manually. Patient was transported to operation room still holding the mass manually, then an urgent tracheotomy was performed.

Following it, under general anesthesia we identified a huge lobular tumor with origin from the mucosal surface of the posterior wall of the hypopharynx postcricoid area, (Figure 5). It completely fills the oral cavity, and then the tumor was removed safely



Figure 4. Patient with polypoid mass hanging out from mouth (after tracheostomy).

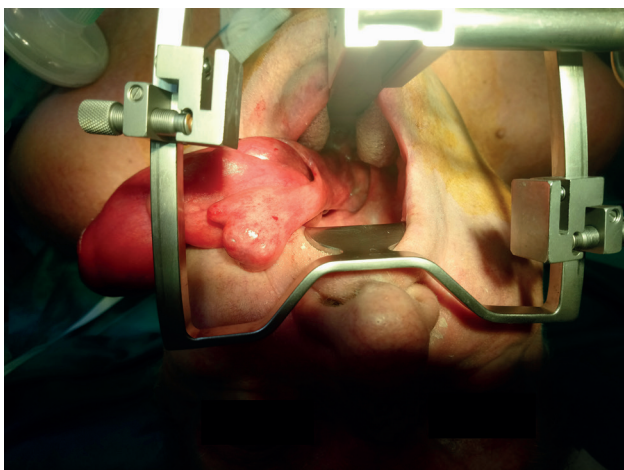


Figure 5. Intraoral view helping by Kasperbauer's spreader.



Figure 6. Tumor after removal.

(Figure 6). Nasogastric tube was inserted. Tumor was sent for histological examination, result was - lipoma pendulum, limb lipoma.

After the patient has recovered from the procedure, we could get more history from the patient's relatives. Patient has had a family celebration. He was in a good condition before, without any serious problem with digestive tract, no vomiting, no coughing, with no previous gastrointestinal problems or examinations. Tumor comes up suddenly without any warning signs. He also suffered from high blood pressure, ischemic heart disease, AV block, benign prostatic hyperplasia.

No complication was seen during hospitalisation, the patient was decannulated on the 5th postoperative day, nasogastric tube was removed on the 9th postoperative day. Patient safely discharged home. Follow up after two weeks, patient remain asymptomatic, on endoscopic examination tumor free.

DISCUSSION

Lipomas have been found in all parts of the digestive tract, most often in the intestine. Esophageal lipoma is a very rare tumor, asymptomatic, mainly identified accidentally, it has never been described as an acute condition or as a life-threatening. In literature we haven't found lipoma coming out of mouth as a first sign in diagnostics.

In the differential diagnosis in the esophagus, we mainly think of Zenker's diverticulum. As far as the fatty tissue tumor, we start to think about liposarcoma, which is one of the most common sarcomas in adulthood. Well-differentiated liposarcomas are clinically low-malignant only rarely metastatic sarcomas, with less-to-poorly differentiated liposarcomas often being high-malignant sarcomas. There are several histological subtypes of lipomas such as simple lipoma, fibrolipoma, mixed lipoma, chondroid lipoma, angiolipoma, angiomyolipoma, myelolipoma, spindle lipoma, sialolipoma, pleomorphic lipoma and atypical lipoma [9]. In a differential diagnosis in dyspneic disorders, it may be a lipoma that grows directly from the larynx, which is also rare; only a few cases are described in the literature [10,11].

Mayo et al. present 4,000 clinical cases of benign neoplasia. From these benign neoplasia, lipomas formed 4.1 % , esophageal lipomas 0.4 %. Nora presents 17 oesophageal lipomas, of which 16 were

located intraluminal. Akiyama et al. documented 10 esophageal lipomas, 7 of which were in the cervical and 3 in the thoracic region. Moerscha and Harrington study included 7,459 drinks, where 44 benign esophageal tumors were randomly found and in only two cases it was a shy lipoma [5,12].

The choice of surgical technique depends on the localization, size and accessibility of the tumor. Endoscopic techniques are more often popular, as it shortens length of hospitalisation and recovery of the patients. It is associated with a lower risk of complications (such as, pulmonary atelectasis, dystelectasis and pleural effusion) [12]. Open surgical techniques are used for better clarity and easier ligation of blood vessels. It is beneficial for the impossibility of removing enormous mass through the esophageal lumen. The use of a transoral, transthoracic, or transcervical approach depends on the size of the tumor, the location of the intraluminal mass and the origin of tumor /from the hypopharyngeal or esophageal wall/, the risk of airway injuries and large vessels [7,12].

In the literature, the tumor of the upper third of the esophagus is presented through a right-handed mini-thoracotomy with a video-assisted thoracoscopic technique [5]. Weigel et al. present a case of resection of the giant stemmed lipoma of the lower third of the esophagus through the transgastric laparoscopic approach [12]. In our case a transoral approach was used. Tumor's origin was identified on the low part of the posterior

pharyngeal wall near the postcricoid area; the tumor was removed safely with no complications.

Gastrointestinal tract lipomas are rare, but esophageal lipomas even more. They represent 0.03 % of all esophageal benign neoplasia. Presentation is often asymptomatic, however, sometimes it can be associated with breathing difficulties and can lead to apnoea. In our case, lipoma was huge. The patient suddenly became breathless and went into acute distress syndrome. We would like to emphasize that lipoma coming out of the oral cavity can be a life-threatening. We have to be aware about this rare tumor, we need to be ready to face it if a situation like this comes to our practise.

Author contribution

All authors reviewed the results and approved the final version of the manuscript.

Ethical approval

It is not necessary to case report.

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Conflict of interest

The authors declare that there is no conflict of interest.

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