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Isolated hypopharynx lipoma: a case presentation and literature review

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Abstract

Background: Lipomas are the most common benign mesenchymal tumors. They are defined as subcutaneous neoplasms of mature adipocyte cells that can occur wherever fatty tissue is. Lipomas are rare in the upper aerodigestive tract. Usually asymptomatic, they may be painful, uncomfortable, or even life-threatening especially if voluminous and located in the upper aerodigestive tract.

Case presentation: A 67-year-old female patient has presented with dyspnea on mild effort and chronic orthopnea. The physical examination was normal while the fiber optic endoscopy revealed a submucosal round-shaped mass rising from the left side of the post-cricoid region. CT scan revealed a well-circumscribed fatty mass of the left piriform sinus for which the patient underwent an endoscopic transoral approach for a complete removal with good results.

Discussion and conclusion: Pharyngeal lipomas are rare entities that might be life-threatening. Although clinical manifestations are not specific, imaging techniques, especially MRI, help set the diagnosis showing a fatty mass of the upper aerodigestive tract. However, pathology examination is crucial to rule out low-grade liposarcomas. Surgical management is not well-codified and has benefited from the development of endoscopic techniques.

Keywords: Lipoma, Hypopharynx, Pyriform sinus, Larynx, Endoscopy

Background

Lipomas are the most frequent mesenchymal benign tumors. They can develop in all fatty areas of the body, be solitary or multiple. They are primitive in the vast majority of cases and occur mainly in adults between 40 and 60 years of age [1]. Malignant transformation has never been described. Very rarely, in the head and neck region, they can be infiltrative [2, 3]. They do not regress spontaneously. Their prevalence is estimated at 0.6% of neoplasms [4, 5]. A scarcity is more marked in the ORL area, particularly the pharyngolaryngeal location.

We present the case of a pharyngeal lipoma involving the left piriform sinus diagnosed in a 67-year-old female **Case presentation**A female patient in her sixties with no significant medical history was referred to the ENT outpatient clinic by her cardiologist for chronic orthopnea and dyspnea on mild

patient presenting with mild exertional dyspnea and orthopnea, for which she underwent a complete endo-

scopic removal with a satisfactory clinical outcome.

physical effort associated with normal cardiac functioning. The patient reported a 05-year history of persistent pharyngeal foreign body sensation with recent snoring and sleep apnea. Otherwise, no other symptoms such as dysphagia, dysphonia, cough, or expectorations were described.

Physical examination disclosed no cervical lymphadenopathies and no goiter. Fiber optic endoscopy revealed a submucosal round-shaped mass rising from the left side of the post-cricoid region. The tumor was covered by normal mucosa and was moving up and down

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during swallowing and respiration. The mass filled the supraglottic region during inspiration and disappeared during swallowing (Videos 1 and 2).

For further characterization of the tumor, a cervical CT scan with contrast injection was ordered. It showed a low-attenuation mass that rose from the left aryepiglottic fold (Figs. 1 and 2) with very thin septa, scattered small areas of soft tissue density, and thin capsule which made it hard to distinguish from the surrounding air in the upper aerodigestive tract.

The patient underwent an endoscopic transoral approach under general anesthesia for the complete

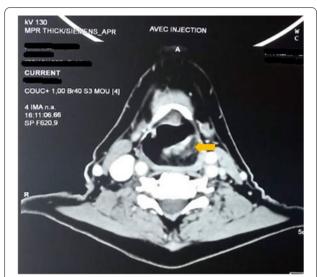


Fig. 1 Cervical CT scan axial view showing a mass of the left pharyno-epiglottic fold

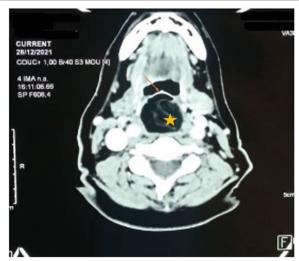


Fig. 2 Cervical CT scan C+ axial view showing upper aerodigestive tract obstructive lipoma



Fig. 3 Complete resection of the lipoma

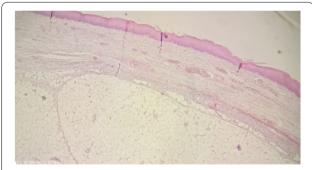


Fig. 4 Benign submucosal adipocyte proliferation (HES*40)

removal of the tumor and the exceeding overlying pharyngeal mucosa. The tumor has a small peduncle with a narrow base originating from the post-cricoid area (Fig. 3). No sutures were placed in the root of the peduncle. The postoperative course was uneventful. The patient received antibiotics and paracetamol for 1 week and anti-reflux medication until complete healing of the wound. A feeding tube was used for 3 days; then, oral feeding was allowed; and the patient was discharged from the hospital. However, non-spicy mixed food was recommended for 2 weeks.

Histopathology examination (Fig. 4) confirmed the diagnosis of lipoma characterized by the presence of lobulated, mature adipocytes with minimal connective tissue stroma, enclosed in a thin, fibrous capsule with no cellular atypia.

The patient's episodes of care timeline is reported in Fig. 5.

Discussion

Lipomas are the most common benign mesenchymal tumors [1]. They are usually presenting as subcutaneous neoplasms of mature adipocyte cells. However, they can occur wherever fatty tissue is found. In 80% of cases, they are typically subcutaneous lipomas with no particularities [1, 2]. Head and neck lipomas count for 13% of all cases, mainly located in the nuchal area [6]; less frequently in the anterior cervical region, infratemporal fossa, oral cavity, and parotid gland; and even more rarely in the upper aerodigestive tract [5]. In our case, the lipoma originated from the anterior wall of the left pyriform sinus.

Lipomas are usually described as painless, slow-growing soft tissue tumors of different sizes that are characterized by an insidious evolution. In fact, they usually remain silent and are often discovered fortuitously during a clinical examination or a radiology test. Nevertheless, lipomas of the upper aerodigestive tract may go unnoticed on CT scans due to the similarity of

density between the air and the fatty portion especially in the pedunculated forms. They are most often sporadic in isolated cases. However, in 5–15% of patients, lipomas are multiple, thus described as lipomatosis, and approximately a third of these will be familial [7] as well as associated with other syndromes and diseases such as Gardner's syndrome, Bannayan-Zonana syndrome, Dercum syndrome, Cowden syndrome, Proteus syndrome, and Madelung's disease [8].

Clinically, lipomas may remain asymptomatic for a long period until reaching a considerate size; consequently, they are discovered at an advanced stage. On the other hand, for the symptomatic forms, and more precisely in the pharyngeal ones, clinical features depend on the location and the impact of these lipomas on the adjacent structures. The patient may describe a simple throat discomfort, a pharyngeal foreign body sensation or heaviness, dysphagia, and swallowing difficulty. Furthermore, some may experience life-threatening dyspnea, especially in the pedunculated forms, due to laryngeal obstruction, a complete externalization through the oropharynx as the case described by Gilberto et al. [9], or in the huge compressive forms. In this context, one case of death has been reported following asphyxia which was secondary to a voluminous and obstructive hypopharyngeal lipoma [10]. On the optic endoscopy, the lipoma appears as a well-limited, round-shaped, submucosal mass. It may be pedunculated or sessile.

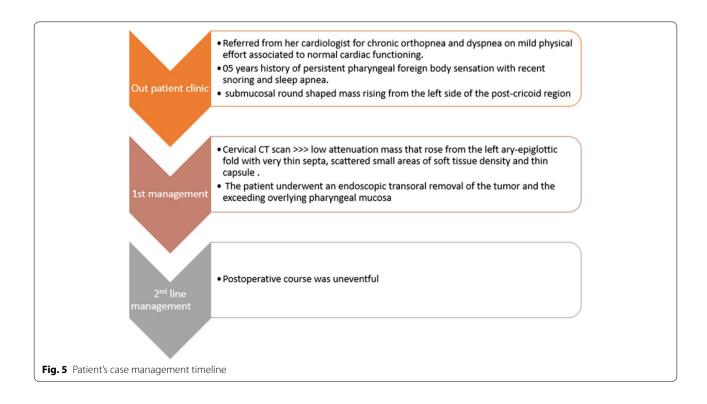


 Table 1
 Chart summarizes all hypopharyngeal lipomas reported in the literature to date

Authors Year Kramer [17] 1934 Max et al. [18] 1952	Year Country	No. of cases	Age/sex Location	_ocation	Symptoms	Treatment	Outcome
	1 New York	36 ^a	_ 0.0	Hypopharynx and esophagus (n = 6), hypopharynx (n = 26), doubtful (n = 4)	Lump or pressure in the throat, intermittent extrusion of the mass into the mouth, dysphagia, and dyspnea	Most of the cases endorally, some operated upon externally	-1
		d L	1 W/25	Right pyriform fossa	Tickling while swallowing	Resection	Second episode after 02 years
	1952 New York	-	52/M	Left pyriform sinus	Intermittent dyspnea and foreign body sensation in the throat	Endoscopic resection using Michel clips, anchored with long black silk thread around the mass	Symptom-free
David et al. [19] 1987	7 USA	_	I W/99	Hypopharynx	Intermittent dysphagia for solids and coughing	Laser excision	Symptoms' resolution
Nash et al. [20] 1989	- 6	_	58/M	Hypopharynx	Dysphagia	Endoscopic resection	Symptoms' resolution
Fyfe et al. [10] 1991	l USA	_	17/M	-eft aryepiglottic fold	Sudden death	I	ı
lwasaki et al. [21] 1992	2 Japan		44/M	Right edge of the epiglot- tis	Sudden death	I	I
			51/F	Hypopharynx	Asphyxia	1	1
Gutsch et al. [22] 1993	3 Poland	_	56/F	Hypopharynx and larynx	Expiration dyspnea and change of the voice	Per os resection	Complete cure
Hellin et al. [23] 1994	1 Spain	_	40/M	Parapharyngeal space	Obstructing the airway	I	ı
Eckel and Jungehulsing 1994 [11]	4 Germany	m	_	-eft pyriform sinus	Intermittent dysphagia for solids	Endoscopic resection by CO_2 laser	Uneventful
Zbaren et al. [24] 1995	5 Switzerland		47/M	Larynx	I	I	I
Wenig [12] 1995	5 USA	m	28/F 51/F 51/M	Supraglottic larynx and piriform sinus	Airway obstruction, dysphagia, piriform throat discomfort, and a sensation of excessive secretion in the throat	1	I
Jack et al. [25] 1996	5 USA	_	36/M	Retropharynx	OSAS	Transoral resection	Symptom-free
Welinder et al. [26] 1996	5 Denmark	·		Vallecula epiglottica	Fatal airway obstruction	ı	ı
Gao et al. [27] 1997	7 China	-		Hypopharynx	Dysphagia, throat discomfort, and airway obstruction	ı	I
Nwaorgu et al. [28] 1997	7 Nigeria	-	25/M	Pharynx	Intraoral mass and respiratory obstruction necessitating tracheotomy	Left lateral pharyngotomy	ı
Nishimura [29] 1998	8	1	61/M	Pharynx	OSAS	1	_

Complete recovery

Endoscopic complete resection

Progressive dysphagia

Left cricopharyngeal region

W/99

2012 Colombia

Pena-Valenzuela et al. [49]

Authors	Year	Country	No. of cases	Age/sex	Location	Symptoms	Treatment	Outcome
Barry et al. [30]	2000	France	5	M and F	Pharynx and larynx	Dysphagia, throat discomfort, and airway obstruction	I	1
Jungehülsing et al. [31]	2000	Germany	7	M and F	Hypopharynx and larynx	1	1	I
Maged and Riad [32]	2000	Scotland	_	50/M	Larynx	Hoarseness and dyspnea	1	I
Srinivasan and Davies [33]	2000	Ž	-	57/M	Pharynx and larynx	Snoring and intermittent choking	I	I
Nishiyama et al. [34]	2001	2001 Japan	-	82/F	Hypopharynx	Wheezing and intermit- tent breathlessness	I	1
Cantarella et al. [35]	2001	2001 Italy	-	77/F	Right pyriform sinus and the right arytenoid	Dysphagia and difficulty swallowing and occasional nasal regurgitation of food	Complete resection during suspension microlaryngoscopy	Full recovery
Hockstein et al. [36]	2002	USA	-	64/M	Retropharyngeal	OSAS	Not operated	No growth during the 5-year follow-up
Grützenmacher et al. [37]	2002	Germany	-	42/M	Left aryepiglottic	Throat discomfort and airway fold obstruction	ı	I
Miloudi et al. [38]	2005	Morocco	-	57/F	Epiglottis	Dyspnea and dysphagia	I	1
Singhal et al. [39]	2005	India	-	56/F	Epiglottis	Fatal airway obstruction	I	I
Namyslowski et al. [40]	2006	Poland	-	40/M	Retropharynx	Increasing snoring and apnea during sleep, morn- ing headache, daytime sleepiness, and swallowing difficulty	External cervical approach	Symptoms' amelioration
Dereköy et al. [41]	2007	2007 Turkey	-	63/F	Tonsil	Confusion, disorientation, and difficult intubation	ı	I
Mitchell et al. [42]	2007	CK CK	_	62/F	Oropharynx	Stridor	I	1
Mattioli et al. [43]	2008	Brazil	_	58/M	Larynx	Hoarseness and dyspnea	1	1
Minni and al [44]	2008	Italy	_	48/M	Paraglottic space	Hoarseness and soft voice	I	1
Silvia et al. [45]	2010	Italy	-	44/M	Hypopharynx	Sudden death during sleep	ı	ı
Eyigor et al. [46]	2011	Turkey	-	W/09	Vocal fold and vocal process of the arytenoids	Hoarseness	ı	1
Evcimik et al. [47]	2011	Turkey	-	51/F	Left pyriform sinus	Mass in the throat and difficulty in swallowing	Complete resection using suspension microlaryngoscopy	Full recovery
Nader et al. [48]	2012 Iran	Iran	-	63/M	Right aryepiglottic fold	Intermittent stridor and snoring	I	I

Table 1 (continued)

Authors	Year	Country	No. of cases	Age/sex	Location	Symptoms	Treatment	Outcome
Lee et al. [50]	2013	South Korea	_	53/M	The lateral wall of the right pyriform sinus	Mild dyspnea on exertion	Transoral robotic approach	Good results
D'Antonio et al. [51]	2013	2013 Italy	-	65/M	Left vocal cord	Hoarseness, choking spells, stridor, and dyspnea	Endoscopic complete resection	Complete recovery
Balasundaram [52]	2013	USA	-	M/97	Hypopharynx	Obstructive sleep apnea	Complete resection	Episodic sleep apnea and snoring resolved
Kazunori et al. [53]	2015	2015 Japan	-	64/M	Hypopharynx	Snoring and daytime sleepiness [obstructive sleep apnea syndrome (OSAS)]	Complete resection	Improvement of apnea- hypopnea index (AHI index)
Wolf-Magele et al. [54]	2015	Austria	-	52/M	Larynx	Acute stridor and dyspnea	Complete resection with the carbon dioxide laser	Full recovery
Al Abdulsalam et al. [55]	2016	Saudi Arabia	-	38/M	Left pyriform sinus	Progressive dysphagia	Complete resection with the carbon dioxide laser	Full recovery
Aydin et al. [56]	2016	2016 Turkey	—	24/M	Retropharynx	Progressive dysphagia and obstructive sleep apnea	Transoral surgical excision	Symptom-free
Acquaviva et al. [9]	2016	2016 Italy	-	63/F	Right pyriform sinus	Voluminous mass, partially protruding from the mouth	Endoscopic resection	Full recovery
Jabbr et al. [57]	2017	2017 United Arab Emirates	_	58/M	Right pyriform sinus	Progressive voice change and paroxysmal coughing spasms with swallowing difficulty and foreign body sensation in the throat	Endoscopic complete resection	Absence of recurrence with good follow-up results
Jia et al. [58]	2018	ÜK	-	63/M	Hypopharynx	Food regurgitation, dys- phagia, and weight loss	ſ	ı
Yoseph et al. [59]	2018	USA	-	W/09	Left pyriform sinus	Throat irritation and a pharyngeal foreign body sensation	Transoral robotic surgical (TORS) excision of the mass	Full recovery
Cukic et al. [60]	2019	Serbia	-	W/99	Left posterolateral pharyngeal wall and obliterating the left piriform fossa	Foreign body sensation in the throat	Trans-cervical excision	Full recovery
Liang et al. [61]	2021	2021 China	-	56/M	From the left pyriform to the oral cavity	Vomiting out of a smooth and giant mass in the oral cavity	Resection using transoral suspension laryngoscopy	No recurrence was found after 5 months of follow-up

^a Literature review ^b Case report

Histologically, simple lipomas can be distinguished, based on their stroma, from the other benign variants including myolipoma, chondrolipoma, angiomyolipoma, adenolipoma, myxolipoma, and spindle cell lipoma [11] on the one hand. On the other hand, it is also important to rule out some malignant histology types such as liposarcoma in particular the well-differentiated cell form [12].

Concerning radiology features, lipoma is typically a well-circumscribed, round-shaped mass with homogeneous characteristics corresponding to a fat imaging with a thin capsule, very thin septa (< 2 mm), and some scattered small areas of soft tissue density. On ultrasounds, they are mostly isoechoic (28-60%) and hyperechoic (20-50%), yet they are hypoechoic in about 20% of the time [1] with no acoustic shadowing and no or minimal color Doppler flow [13]. If encapsulated, the capsule may be difficult to identify sometimes and to be distinguished from the air around in the pharyngolaryngeal area [7]. Calcification may also be present in up to 11% of cases, although more commonly associated with well-differentiated liposarcoma [7]. Moreover, avidly enhancing, thick/ nodular septa or evidence of local invasion in addition to heterogeneous echotexture, more than minimal color Doppler flow, suggests malignancy.

The diagnosis is usually indicated by clinical features and ultrasound results. However, in the upper aerodigestive tract, CT scan and MRI imaging may be helpful for a better evaluation of the mass and the surrounding structures. On CT scan, lipomas presented as fatty, homogeneous, low-attenuation masses with minimal internal soft tissue component occasionally. It may also show some areas of fat necrosis, blood vessels, and muscle fibers whereas a liposarcoma is eliminated firstly [7]. MRI can also be used as a diagnosis tool and show a high-signal mass on both T1 and T2 with saturation on fat-saturated sequences. In fact, MRI represents the main imaging tool for lipoma diagnosis with or without atypical features. As a matter of fact, when no suspicious features are present, MRI is 100% specific regarding the diagnosis of lipoma [14]. In the opposite case, if suspicious features of malignancy are present, the specificity of MRI is lower since some masses with atypical features will nonetheless be simple lipomas, while the sensitivity is still 100% [14].

Well-differentiated liposarcomas, which represent the main and most dangerous differential diagnosis of lipomas, have high chances of local recurrence and a possibility of delayed dedifferentiation after the initial treatment [15]. Because of the differences not only in the treatment's modalities, but also concerning the prognosis and the follow-up protocols, it is very important to distinguish simple lipomas from well-differentiated liposarcomas. In fact, immunohistochemistry describes the liposarcoma subtypes disclosing different morphologies, genetics, clinical behavior, pattern of disease progression, response to treatment, and 5-year survival rate [15, 16].

In the upper aerodigestive tract, surgical management presents a challenge regarding the security of the upper airways, the possibility of intubation, the possibility of jet ventilation use, and endoscopic surgery sittings. Since this is a rare entity, each case should be considered unique and have to be managed individually. Nonetheless, it seems that hypopharyngeal lipomas tend to rise from the post-cricoid region [15, 16]. Therefore, securing the upper airways might be challenging if the tumor is large and has no peduncle which implies a transitory tracheostomy. Also, the lent of the peduncle base might condition the necessity or not to put mucosal sutures or surgical glue in order to prevent salivary fistula. Surgical excision might be performed using endoscopic cold instruments (micro scissors, sickle). However, CO₂ laser offers better ergonomics especially regarding bleeding control.

Table 1 summarizes all hypopharyngeal lipomas reported in the literature to date.

Conclusion

Pharyngeal lipomas are rare entities that might be lifethreatening. Although clinical manifestations are not specific, imaging techniques, especially MRI, help set the diagnosis showing a fatty mass of the upper aerodigestive tract. However, pathology examination is crucial to rule out low-grade liposarcomas. Surgical management is not well-codified and has benefitted from the development of endoscopic techniques.

Patient's perspective

I recently had breathing difficulty so I went seeing my cardiologist. After routine work up, he advised me to see an otolaryngologist as my results were normal. In the ORL outpatient clinic, doctor performed a fibroendoscopy and discovered a tumor. I was scared of being diagnosed with cancer. I must say that I had swallowing difficulties that I didn't took seriously. I had a CT scan and the ORL told me I had a fatty mass that he can remove pretty easily. I got the surgery. Post operatively, I had mild pain but my swallowing and breathing difficulties were resolved. I was discharged from the hospital within 3 days. I went back to see my doctor within 10 days for pathology results. He appeased my worries as the tumor was benign. Since surgery my breathing and swallowing symptoms were completely resolved.

Supplementary Information

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Additional file 1: Video 1. Round shape mass filled the supraglottic region during inspiration.

Additional file 2: Video 2. The mass disappear during swallowing.

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Authors' contributions

NO was involved in the diagnosis, surgery procedure, and manuscript drafting. OQ was involved in the literature review and drafting of the manuscript. NH was involved in the pathology study and reviewed the manuscript. OA was involved in the surgery procedure and reviewed the manuscript. MNA reviewed the manuscript for insightful remarks. The authors read and approved the final manuscript.

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Availability of data and materials

The datasets generated and/or analyzed during the current study are not publicly available due to patient's data confidentiality but are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

An informed consent for publication purposes was obtained from the patient.

Competing interests

The authors declare that they have no competing interests.

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