

# Upper airway study should always come before any sleep study in OSAS evaluation: a giant parapharyngeal lipoma behind OSAS

M. CASALE<sup>1</sup>, F. SALVINELLI<sup>1</sup>, C.A. MALLIO<sup>2</sup>, V. FRARI<sup>1</sup>,  
B. VINCENZI<sup>3</sup>, F. BRESSI<sup>1</sup>, C.C. QUATTROCCHI<sup>2</sup>

Areas of <sup>1</sup>Otolaryngology, <sup>2</sup>Radiology, <sup>3</sup>Oncology; Campus Bio-Medico University, School of Medicine, Rome, Italy

**Abstract. – BACKGROUND:** The parapharyngeal space (PPS) is a rare site for neoplasms in the head and neck and lipoma represents 0.5% of all head and neck tumors.

**CASE REPORT:** We describe a case of a giant parapharyngeal lipoma in obese adult patient causing anatomic pharyngeal obstruction with severe obstructive sleep apnea (OSA) syndrome. The patient was successfully operated with trans-cervical approach.

**CONCLUSIONS:** In patients presenting with symptoms suggestive of OSA, it is essential to rule out any physical cause and perform a comprehensive ear, nose and throat examination including fibro-endoscopic upper airway examination before referral for sleep study and management. The ideal management for OSA involves treating the underlying cause. If no definitive cause is identified, management should begin with conservative measures such as lifestyle changes and a weight loss program; if these are unsuccessful, continuous positive airway pressure (CPAP), oral appliances or upper airway surgery could be considered. Better diagnostic methods should be developed to identify the main OSA causes and improve therapeutic outcomes.

*Key Words:*

Parapharyngeal lipoma, Obstructive sleep apnea, Fibro-endoscopic upper airway examination, Cervical MRI.

## Introduction

Lipoma in parapharyngeal space is rare benign tumour and it can extend from nasopharynx to the glottis. It has a mesenchymal origin, only 13% of which are located in the head and neck region representing 0.5% of all head and neck tumors<sup>1,2</sup>. They typically occur between the fourth and sixth decades of life with male preponderance. The parapharyngeal lipoma (PPL) is often

asymptomatic and the diagnosis may be difficult due to the slow growth rate of this neoplasm; they are usually not diagnosed before they become larger in size when the most common symptoms are typically respiratory disturbances and dysphagia. The MRI can supply adequate information with regard to the structure and extension of the tumour, although final histological confirmation is essential. Surgery is well established as the primary treatment modality for lipomas. A number of approaches have been proposed including transoral, transcervical, transparotid, transmandibular, and combinations thereof<sup>3</sup>. Obstructive sleep apnea syndrome (OSAS) is caused by intermittent collapse of the pharyngeal wall during sleep, which is typically associated with arousal and respiratory pause. Direct consequences are sleep disturbances, such as fragmentation and recurrent hypoxemia and hypercapnia episodes. When lipomas are localized in the retropharyngeal space obstructing the pharyngeal lumen, they become symptomatic for OSA<sup>4</sup>. We describe a rare case of OSA secondary to retropharyngeal lipoma.

## Case Report

A 70 year-old obese (BMI = 38) man presented at the Area of Otolaryngology of Campus Bio-Medico University, Rome, Italy, in November 2011 with worsening symptoms occurring since August 2009 characterized by hypersalivation, changes in voice tone, difficulty in swallowing, sensation of foreign body in the throat, severe sleep disturbances with snoring and respiratory apneas.

An ear, nose, throat (ENT) examination revealed a palpable mass in left side of the neck and a fibroscopic examination showed a big round and tender mass localized in the left oro- and hypo-

pharyngeal wall obstructing the pharyngeal lumen. The patient underwent polysomnography (PSG) that revealed a severe OSAS (AHI: 65) with Epworth sleepiness scale (ESS) score of 13.

To better define the extent and quality of this mass, a MRI exam was performed that confirmed a solid huge (90 x 60 mm) neoplasm, with a signal intensity consistent with fatty content localized in the retropharyngeal space with compression of the oro and hypo-faryngeal lumen; a dislocation of the carotid and jugular axis, parotid and submandibular salivary glands, hyoid bone and supra- and sub-hyoid muscles was also observed (Figure 1).

The patient directly underwent a transcervical radical surgical excision performed by FS. One month after surgery, the patient reported a considerable improvement of his symptoms with a ESS of 5 and AHI (Apnea-hypopnea index) of 31 despite unchanged BMI (body mass index).

Histopathological assessment confirmed the diagnosis of lipoma; no atypia or malignant features were present.

## Discussion

The parapharyngeal space (PPS) is a rare site for neoplasms in the head and neck. Eighty percent of parapharyngeal tumours are benign, while 20% are malignant<sup>5</sup>.

There are three main groups of parapharyngeal space tumours, in particular salivary tumours from the deep lobe of the parotid gland, neurogenic tumours most commonly schwannomas and paragangliomas like as chemodectomas. Benign mixed salivary cell tumours and schwannomas are the most commonly noted benign tumours<sup>6</sup>.

Retropharyngeal lipoma (RL) causing OSA are very rare, 9 of them has been reviewed by Tuncyurek et al<sup>7</sup>, other two are those in the Senchenkov et al<sup>8</sup> and Srinivasan et al<sup>9</sup> reports. Among the 12 published cases, 8 were men (67%) and 4 women (33%), only 1 patient was an 11 year old child and all others 11 patients were older than 30 year-old. RL could be a potential misdiagnosed condition; subtle clinical symptoms such as snoring, dysphagia and globus sensation, might be attributed to other causes; indeed, the case of RL causing sleep apnea reported by Tuncyurek et al<sup>7</sup> was a 1-year complaint, and its diagnosis was delayed because no imaging examination was performed.

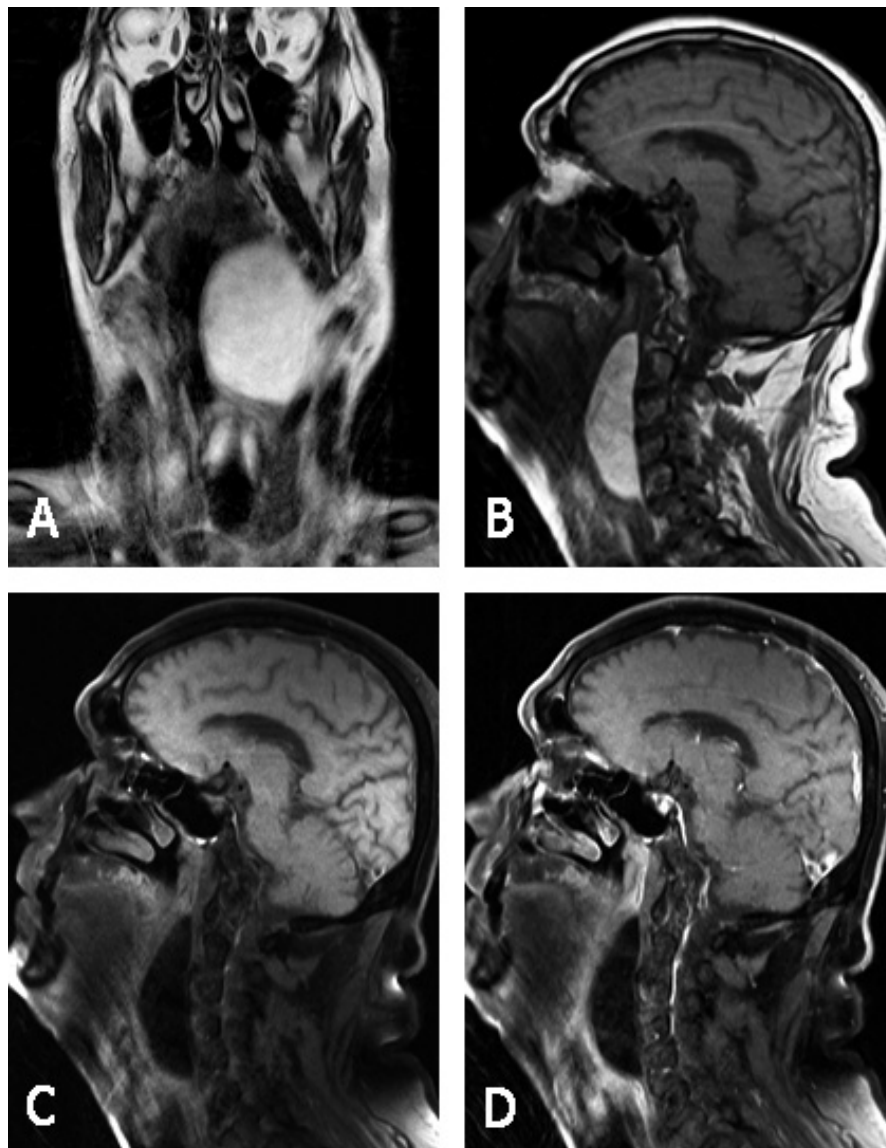
Hockstein et al<sup>10</sup> reported incidentally a diagnosis of PPS lipoma when a large retropharyngeal mass was noted on the scout film of computed tomography (CT) scan, performed to evaluate paranasal sinuses because sinusitis was suspected.

MRI findings of lipomas are typical and diagnosis isn't usually difficult. Lipoma tends to demonstrate high signal intensity on T1 weighted sequences and T2 weighted fast spin echo MRI sequences. Lipomas, unlike fatty deposits (lipomatosis), are generally homogeneous mass (Figure 1) with a good plane of cleavage from muscular-vascular structures and internal septae in most cases. However, the possibility of liposarcoma cannot be excluded and Gadolinium has a role in differential diagnosis of lipoma; irregular interface between lipoma and muscle has been considered predictive for malignant neoplasia (12).

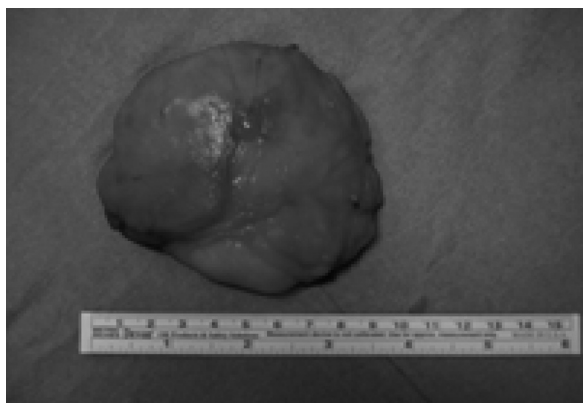
When possible the treatment of choice of PPS lipoma is surgical excision, after which symptoms usually subside; when surgery is not possible or indicated, biopsy and imaging follow-up are required. The PPS tumours can usually lead to no symptomatic cases, but the combination of snoring and a visible intra-oral mass or pharyngeal mass should raise the suspicion. In patients presenting with symptoms suggestive of OSA, it is essential to exclude any physical cause and perform a comprehensive ear, nose and throat examination with fiberoendoscopic upper airway examination before referral for sleep study and management; in rare cases, this simple step could enable early detection of causes, estimating that in about 80% of OSA patients an anatomic abnormalities was found<sup>13</sup>. The causal diagnosis of OSA in obese patients could be insidious, because for the strong association between OSA and obesity it is easier to not consider the hypothesis of an underlying obstructive anatomical cause of OSA<sup>14</sup>. The ideal management for snoring and OSA involves treating the underlying cause. If no definitive cause is identified, management should begin with conservative measures such as lifestyle changes and a weight loss program; if these are unsuccessful, CPAP, oral appliances or upper airway surgery could be considered.

In a OSA patient with oral or pharyngeal mass the role of imaging and surgery is obvious, but in the most case the obstruction site it is very difficult to identify in the awake, nonsupine patient, although all surgical procedures must be directed toward the anatomic site of obstruction<sup>15</sup>.

Better diagnostic methods should be developed to help direct treatment options and improve therapeutic outcomes.



**Figure 1.** Cervical MRI shows a solid mass lesion located at parapharyngeal space and obliterating the air column on left side.



**Figure 2.** Solid huge (90 × 60 mm) neoplasm, localized in the retropharyngeal space after excision.

## References

- 1) ROGERS J, PATIL Y, STRICKLAND-MARMOL L, PADHYA T. Lipomatous tumors of the parapharyngeal space: case series and literature review. *Arch Otolaryngol Head Neck Surg.* 2010; 136: 621-624.
- 2) SMITH JC, SNYDERMAN CH, KASSAM AB, FUKUI MB. Giant parapharyngeal space lipoma: case report and surgical approach. *Skull Base* 2002; 12: 215-220.
- 3) PRESUTTI L, MOLteni G, MALVÈ L, MARCHIONI D, GHIDINI A, TASSI S, CHIARINI L, ALICANDRI-CIUFFELLI M. Parapharyngeal space tumors without mandibulotomy: our experience. *Eur Arch Otorhinolaryngol* 2002; 269: 265-273.
- 4) ALAND JW. Retropharyngeal lipoma causing symptoms of obstructive sleep apnea. *Otolaryngol Head Neck Surg* 1996; 114: 628-630.

- 5) PANG KP, GOH CHK, TAN HM. Parapharyngeal space tumours: an 18 year review. *J Laryngol Otol* 2002; 3:170-175.
- 6) MORAITIS D, PAPAOKOSTA K, KARKANEVATOS A, COAST GJ, JACKSON SR. Pleomorphic adenoma causing acute airway obstruction. *J Laryngol Otol* 2000; 114: 634-636.
- 7) TUNCYUREK O, EYIGOR H, OZKUL A. What is the importance of radiology in obstructive sleep apnea? *J Craniofac Surg* 2011; 22: 1971-197.3
- 8) SENCHENKOV A, WERNING JW, STAREN ED. Radiographic assessment of the infiltrating retropharyngeal lipoma. *Otolaryngol Head Neck Surg* 2001; 125: 658-660.
- 9) SRINIVASAN V, DAVIES JE. Snoring with sleep apnoea:deceptive appearances. *J R Soc Med* 2000; 93: 81-82.
- 10) HOCKSTEIN NG, ANDERSON TA, MOONIS G, GUSTAFSON KS, MIRZA N. Retropharyngeal lipoma causing obstructive sleep apnea: case report including five-year follow-up. *Laryngoscope* 2002; 112: 1603-1605.
- 11) ROSELL A, GARCIA-ARRANZ G, LLAVERO MT, MARTINEZ-SAN-MILLAN J. Imaging case study of the month. Lipoma of the retropharyngeal space. *Ann Otol Rhinol Laryngol* 1998; 107: 726-728.
- 12) DOOMS GC, HRICAK H, SOLLITTO RA, HIGGINS CB. Lipomatous tumors and tumors with fatty component: MR imaging potential and comparison of MR and CT results. *Radiology* 1985; 157: 479-483.
- 13) MULLA O, AGADA F, DAWSON D, SOOD S. Obstructive sleep apnoea and snoring—is examination necessary? *Aust Fam Physician* 2011; 40: 886-888.
- 14) BABADEMEZ MA, GÜNBEY E, ACAR B, GÜNBEY HP. A rare cause of obstructive sleep apnea syndrome: lingual thyroid. *Sleep Breath* 2012; 16: 305-308.
- 15) SITTITAVORNWONG S, WAITE PD. Imaging the upper airway in patients with sleep disordered breathing. *Oral Maxillofac Surg Clin North Am* 2009; 21: 389-402.