Case report

Lipoma of the larynx: a case report

Lipoma della laringe: un caso clinico

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SUMMARY

Lipoma is a benign tumour of mesenchymal origin with a very rare occurrence in the upper aero-digestive tract. To date, approximately 100 cases have been described in the literature. This lesion has a slow growth and, therefore, can present with various symptoms due to the mass effect with obstruction and compression on neighbouring structures, including dysphagia for liquid and solid food, dyspnoea and hoarseness. For a precise pre-operative diagnosis, indirect or direct laryngoscopy (flexible fibre-optic laryngoscopy) can be employed or, if necessary, also imaging techniques such as computed tomography scan and magnetic resonance imaging scan. These offer more useful information for better treatment planning. Surgery is the treatment of choice and includes endoscopic techniques and an external surgical approach (cervicotomy). It is very important to completely remove these benign neoplasms in order to avoid local recurrence. The present report refering to a case of laryngeal lipoma removed through an external surgical approach, aims to demonstrate that the choice of an external surgical approach is required for complete surgical removal of a large lipoma in order to prevent any possible recurrence. Furthermore, it is useful to keep in mind the possibility of recurrence of lipomas after long free intervals; therefore, it is mandatory to observe these patients at long-term follow-up.

KEY WORDS: Larynx • Lipoma • Imaging • Surgical techniques

RIASSUNTO

Il lipoma è un tumore benigno di origine mesenchimale la cui localizzazione a livello del tratto superiore delle vie aereo-digestive è molto rara. In letteratura finora ne sono stati descritti circa un centinaio di casi. Tale lesione presenta una crescita lenta e pertanto si può manifestare tardivamente con varia sintomatologia, dovuta soprattutto all'ingombro fisico e a fenomeni di compressione che essa può esercitare sulle strutture circostanti, che comprende disfagia per i liquidi e per i solidi, dispnea e raucedine. Per una esatta diagnosi pre-operatoria noi possiamo ricorrere alla laringoscopia indiretta e diretta (fibrolaringoscopia) e a tecniche di Imaging quali la Tomografia Computerizzata e la Risonanza Magnetica. Queste ci permettono di acquisire informazioni utili a pianificare un adeguato trattamento terapeutico. Il trattamento di scelta è di tipo chirurgico e comprende tecniche di tipo endoscopico o per approccio esterno (cervicotomia). L'importante è rimuovere completamente la neoformazione in modo da evitare eventuali recidive. Questo elaborato è un report di un caso di lipoma laringeo asportato attraverso un approccio chirurgico per via esterna. Il nostro obiettivo è di dimostrare che la scelta di un approccio esterno è necessaria per una completa asportazione di un lipoma di grosse dimensioni in modo da prevenire possibili recidive. Inoltre, è utile ricordare la possibilità di una ricorrenza dopo un lungo periodo libero da malattia; pertanto, è preferibile sottoporre il paziente ad un follow-up post-operatorio a lungo termine.

PAROLE CHIAVE: *Laringe • Lipoma • Imaging • Tecniche chirurgiche*

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Introduction

Lipomas, which represent 4-5% of all benign tumours of the body, occur most commonly in the trunk and lower and upper limbs where subcutaneous fat tissue is more abundant [1,2].

This kind of mesenchymal tumour, on the contrary, rarely occurs in ENT districts (head, neck); they very rarely occur in the upper aero-digestive tract (larynx, hypopharynx) where they represent 0.6% of all benign neoplasms [3,4]. They may appear as a pseudo-cystic or pedunculated mass; they cause few and aspecific symptoms and should be taken into consideration in the differential diagnosis of all benign head and neck masses.

To date, approximately 100 cases have been described in the Literature [5-8].

The present report refers to a patient in whom a large pseudo-cystic mass, presenting in the right ary-epiglottic fold, was revealed and, following surgical removal, was found, upon histological examination, to be a lipoma composed of mature adipocytes.

Clinical, diagnostic and therapeutic features are described.
The aim of this report is to demonstrate that the choice of an external surgical approach, carried out on several of the tumour characteristics, is required for complete surgical removal of a large lipoma in order to prevent any possible recurrence.
Furthermore, due to the possibility of recurrence of lipomas after long free intervals, it is mandatory to observe these patients at long-term follow-up.

Case report

A 62-year-old male came to our attention complaining of increasing symptomatology, which had commenced several months earlier, including dysphagia, throat discomfort (like a lump in the throat) and dyspnoea (on exertion and at rest).
The ENT examination revealed a large, smooth, pseudocystic mass, 2-3 cm in diameter, arising from the right ary-epiglottic fold. This lesion was of a yellowish appearance and covered by normal, non-haemorrhagic mucosa.
Upon admission to our Department, the patient was submitted to an angio-computed tomography (CT) scan of the head and neck regions that showed a bulky, oval mass measuring 4 cm in maximum diameter, with well-defined and regular margins. This lesion, which was encapsulated, was of lipomatous density and internal thin septal structures were depicted.
The mass arose from the right para-laryngeal space and presented an intra-luminal projecting portion that extended upwards with involvement of the hypopharynx, to the level of the hyoid bone, narrowing the pyriform sinus (Figs. 1a-d, 2a-b, 3).
This lesion exerted compression on the larynx, that showed deviation to the left and a narrowed lumen, and on right infra-hyoid muscles; this mass was well-circumscribed with respect to the neck vessels.
Radiological findings indicated a diagnosis of a bulky lipomatous mass, originating from the right para-laryngeal space.
The patient was then submitted to microlaryngoscopy; during this procedure several biopsy specimens of the lesion were collected. Histological examination demonstrated a fibrous adipose tissue.
During endoscopic evaluation, considerable narrowing of the upper airway, caused by this mass, was detected, above all at the level of the supra-glottic larynx and, therefore, tracheotomy was deemed necessary.
Due to the large size (> 2 cm in maximum diameter) and site of this lesion, and the compression exerted on the surrounding anatomical structures, it was decided to proceed with surgical management, via an external (trans-cervical) approach, in order to ensure complete removal of the tumour.
Thus, after having performed a cervical skin incision corresponding to the level of the hyoid bone, a myo-cutaneous flap was prepared and raised to expose the pre-laryngeal muscles resected at the level of insertion at the inferior border of the hyoid bone.
A further surgical step was performed exposing the hyothyro-epiglottic space followed by a right lateral pharyngotomy.
By means of this surgical approach, it was possible to remove the mass, located at the level of the right ary-epiglottic fold and involving the homolateral pyriform sinus, with preservation of the epiglottis that had previously been stretched and folded over.
The final histological diagnosis demonstrated a lipoma composed of mature fat cells.
No complications occurred during the post-operative period; the patient received nutrition by means of a nasogastric feeding tube positioned during the surgical procedure.

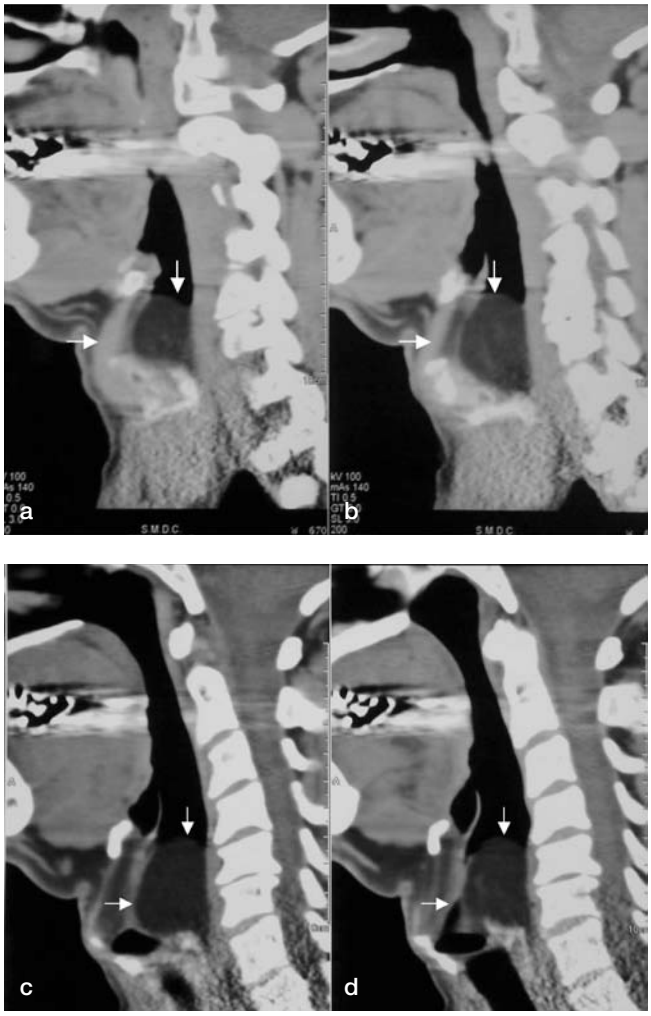

Fig. 1a, b, c, d. Sagittal reconstruction of axial CT scan without contrast-enhancement shows a bulky, expansive, oval mass (white arrows) with maximum dimension 4 cm, well-defined and regular margins, encapsulated, with frankly lipomatous density involving hypopharynx, at level of hyoid bone, and exerting compression on larynx with a markedly narrowed lumen.

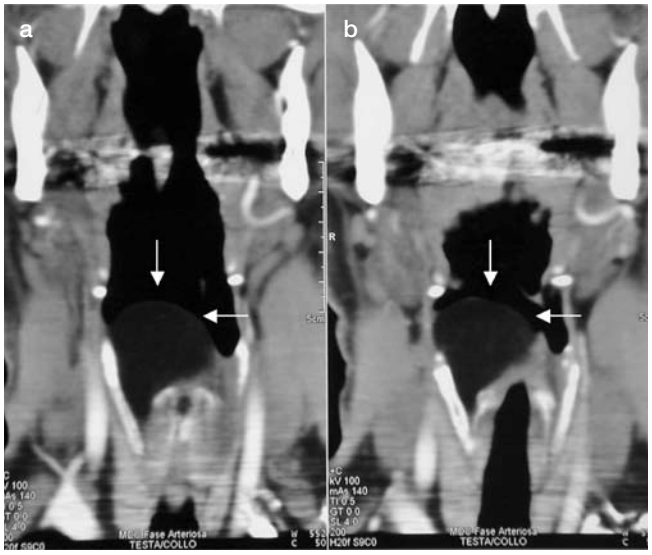

Fig. 2a, b. Coronal reconstruction of axial contrast-enhanced CT scan (arterial phase) shows that lesion (white arrows) arises from right para-laryngeal space and with an intra-luminal projecting portion extending superiorly with involvement of hypopharynx, covering the ipsilateral pyriform sinus. Lesion exerts compression on larynx that is deviated to left with a markedly narrowed lumen.

Once swallowing rehabilitation was successfully achieved and once adequate nutrition via oral intake had been restored, the naso-gastric feeding tube was removed on the 8th post-operative day while the tracheostomy tube was removed, and tracheostomy closed, on the 17th post-operative day.

On account of the possibility of recurrence of lipomas after long free-intervals, the patient is still periodically observed with physical ENT examination as well as imaging procedures (CT scan, Magnetic Resonance Imaging [MRI]) as part of the extended period of long-term follow-up.

So far, no evidence of recurrence has been detected.

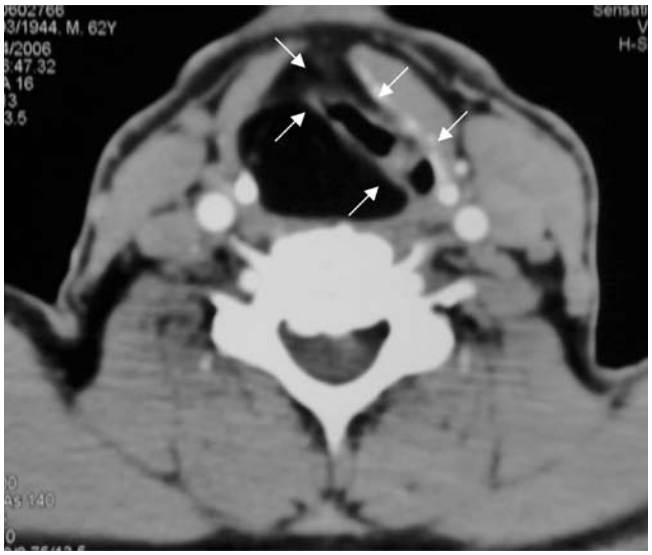

Fig. 3. Axial contrast-enhanced CT scan (arterial phase) shows an encapsulated lesion, originating in right para-laryngeal space with narrowing of homo-lateral pyriform sinus, that exerts compression on right infra-hyoid muscles and is well-circumscribed with respect to neck vessels. No evidence of involvement of the cervical or jugo-digastric lymph nodes.

Discussion

Lipomas are benign, slow-growing primary mesenchymal tumours, which represent 4-5% of all benign tumours in the body [2].

They are more frequent in the trunk and limbs where subcutaneous fatty tissue is abundant; in the case of a large tumour, the clinical implications may give rise to a cosmetic problem or interfere with function as a result of the anatomical position.

It has been estimated that 13-15% of lipomas occur in the head and neck region. The upper aero-digestive tract is a very rare localization [9 10].

Lipoma represents fatty tissue dysplasia with an increase in the histologically typical fat tissue, with not unlimited, slow growth.

Lipoma often presents a thin fibrous capsule and the mature adipose tissue is subdivided into lobules by a meshwork of fibrous strands.

Lipomas can occur as a single or as multiple lesions; in the latter situation, they can represent a clinical manifestation in association with neurofibromatosis, Gardner's syndrome, Launois-Bensaude's syndrome, adiposalgia (Madelung's disease) and Dercum's disease [11-15].

In the two latter diseases, neurological and endocrine symptoms can be found. The aetiology of lipoma is unknown. Several Authors have suggested that lipomas could arise from embryogenetic lipoblast cells or metaplastic muscle cells, while others have suggested a possible aetiopathogenetic role of familial and endocrine factors, trauma, infections or chronic irritating conditions. To date, no definite aetiological factors, such as excessive alcohol use, tobacco smoking, occupational exposure to several toxic chemicals, have been identified [1 8].

Lipomas rarely occur in paediatric age [16]; they are more frequent between the 4th and 5th decade of life (50%) [4], while 28% occur after the 5th decade and 27% between the 3rd and 4th decade.

Lipoma is more frequent in males (62.5%) [17].

Histologically, lipomas, partly or totally encapsulated, may be composed of mature fat cells, similar to normal cells, varying slightly in size and shape, with no evidence of pleomorphism, lipoblasts or atypical adipocytes with hyperchromatic nuclei**,** or of an infiltrative growth into the surrounding tissues. They are richly vascularized but the vascular framework may be difficult to appreciate due to compression by over-distended adipocytes resulting from lipid vacuole storage.

Histological examinations reveal secondary changes such as haemorrhage, fat necrosis, calcification, formation of

cysts as well as metaplastic components, such as cartilage and bone tissue [8].

This type of benign lipogenic neoplasm arises from white fat cells and must be distinguished from benign tumours that arise from brown fat (hibernoma).

There are several histological forms, occurring most frequently in other ENT districts (oral cavity), but also reported, by several Authors, in the region of the larynx and pharynx [8], characterised by an admixture of various mesenchymal components amongst which: myxolipoma (mucoid substance storage) [16 18 19], fibrolipoma (fibrous-connective component) [20-23], spindle cell lipoma [24-26], angiomyolipoma, pleomorphic lipoma.

Possible malignant types are represented, above all, by well-differentiated liposarcoma (well-differentiated lipoma-like liposarcoma) [27-31]. The histological features, allowing a differential diagnosis with respect to their benign counterparts, include: evidence of pleomorphism and atypical cells, of an infiltrative growth, of lipoblasts.

Malignancy should be suspected in the event of one or more recurrences after surgical excision [8].

Possible malignant transformation is rare in cases of a single lipoma whereas it has been described in association with multiple laryngo-pharyngeal lipomatosis and histological features of malignancy are more evident in recurrent lipoma [32 33].

It is well known that laryngeal lipomas are very rare; they involve, above all, the supraglottic larynx and arise from fat tissue of the laryngeal vestibule, ary-epiglottic fold and epiglottis [34]. A subglottic lipoma has also been reported [8].

Pharyngeal lipomas, on account of the reduced amount of fatty tissue in this region, are rare and occur in the hypopharyngeal region and include the pyriform sinus that, in a purely anatomical sense, may be referred to the laryngeal portion of the pharynx. The clinical, pathological and biological features of these lipogenic lesions are identical to those arising from the larynx [8].

As in our case, lipomas often resemble a retention cyst. On account of the slow and insidious growth, lipomas are clinically asymptomatic for a long time; but once a large mass has been grown, these may give rise to symptoms due to physical discomfort resulting from the size and compression on the surrounding anatomical structures.

Lipomas arising from the upper aero-digestive tract present with continuous or intermittent symptoms, occurring over a period ranging from a few months to several years, which include dysphagia, hoarseness, throat discomfort, like the feeling of a lump in the throat, cough and, in the case of airway obstruction, stridor and dyspnoea, upon exertion or at rest, in most severe cases [33 35].

In the latter case, it is necessary to urgently perform tracheotomy, as in our case, since asphyxia and fatal respiratory arrest episodes, due to a pedunculated hypo-pharyngeal lipoma flopping into the laryngeal lumen, are possible [9 18 36].

Pain is unusual and may depend upon the amount of pressure exerted, by a large lipomatous neoplasm, on the surrounding tissues.

Upon physical examination, these tumours may appear as a sessile or pedunculated mass, retention-like cyst, with a smooth or lobulated surface, encapsulated, covered by a pinkish-yellowish normal mucosa [7 8].

Size may vary from a few millimetres to 5-6 cm, in the maximum diameter, in a very large mass.

It is very difficult to differentiate this condition from other benign lesions such as retention cysts or laryngoceles [37].

Upon evaluation with endoscopic techniques (flexible fiberoptic laryngoscopy, oesophagoscopy), the lipoma may have the appearance of a submucosal or polypoid mass, sometimes pedunculated. In the case of deeply situated tumours, it is necessary, for diagnostic purposes, to collect several biopsies, specimens of which are submitted to histological examination.

Pre-operative diagnosis is possible with the use of imaging techniques such as CT scan and MRI.

On CT scans, lipoma has the appearance of a homogeneous lesion, with low ionizing radiation attenuation values (0 or negative values of Hounsfield Units) and a density lower than that of water, and the extension of this tumour is accurately depicted [17 38 39]. The accuracy rate of CT is 75-90% [40].

The MRI is to be preferred with respect to CT since it allows better examination of the soft tissues, on account of the better definition achieved, the patient is not exposed to ionizing radiation and iodine contrast agents are not needed. MRI allows a more accurate and specific diagnosis, also a clearer indication of the origin of the peduncle (high resolution rate in coronal and sagittal scans), of its cranial-caudal extension in para-laryngeal and para-pharyngeal spaces and of its relationship with surrounding cervical structures.

The signal intensities, in the T_1- and T_2-weighted sequences, are like those of subcutaneous fat tissue, thus suggesting that a fatty lesion has been revealed. No enhancement, after the application of Gadolinium-DTPA, suggests a benign lesion [10 41].

Angiography may also have a diagnostic role since it offers the possibility of detecting tumour hyper-vascularization that represents a probable sign of malignant transformation.

In the case described here, an Angio-CT was performed which allowed more accurate information to be acquired regarding localization of the lesion, its extension and its anatomical relationship with surrounding structures; moreover, it revealed the fatty origin of the tumour, with a high rate of probability.

It is tempting to suggest that this lesion, which was fairly large (4-5 cm in maximum dimension), arose from the right para-laryngeal space, had an intra-luminal projecting portion that extended superiorly with involvement of the hypopharynx and obliterating the right pyriform sinus.

Moreover, the mass exerted compression on the laryngeal airway, that was deviated towards the left and markedly narrowed, and on the right infra-hyoid muscles.
Some Authors have performed, for diagnostic purposes, a standard barium swallow examination [10]. In the case of hypopharyngeal lipoma, a delay in barium progression and alterations in the contrast column (lateral or posterior deviation) can be detected. However, this procedure, with respect to the imaging techniques previously considered, does not add any important diagnostic information and, moreover, exposure to ionizing radiation of the thyroid gland is somewhat high.
The findings, obtained with CT or MRI scans, allow better planning of the management strategy and, in turn, better treatment [17].
Surgery is the treatment of choice for laryngeal lipomas.
A conservative endoscopic procedure is preferred in the case of a tumour of small dimensions [34]; while an external surgical approach (via lateral pharyngotomy, laryngofissure, subhyoid pharyngotomy) may be suitable above all for a large tumour (> 2 cm in maximum diameter) [15 22 33 37].
In this latter case, an external approach offers adequate and good exposure of the tumour allowing its complete *en-bloc* surgical removal in order to prevent any possible recurrence.
Therefore, the choice of an external surgical approach is based on several characteristics of the tumour such as site, submucosal growth, size, vascularity, and potential malignancy.
In the case described in this report, removal by means of an external surgical approach, via lateral pharyngotomy, was preferred because of the site and size of the tumour**.**
Surgical removal of lipomas must be complete in order to avoid recurrence; this possibility is fairly frequent in the case of racemose tumours.
Post-operative management includes, in our experience, nutrition by means of a naso-gastric feeding tube, for a few days, then once swallowing rehabilitation has been successfully achieved, the patient may return to an oral diet.
Since lipomas can relapse, even after several years, as proven in a review of the Literature, long-term follow-up, for an extended period of time, is mandatory [15 20 21].
Therefore, when our patient was discharged, indications were given to periodically perform ENT examinations as well as other types of assessment (CT scan, MRI).
Several months after surgery, the patient shows no evidence of recurrence.

Conclusions

Lipomas are benign mesenchymal tumours with rare localization in the upper aero-digestive tract.
On account of slow growth, these tumours may remain clinically undetected for several years; if large in size, they may lead to compression on the surrounding cervical structures and, sometimes, to life-threatening symptoms (dyspnoea, asphyxia).
For a more accurate diagnosis, imaging techniques (CT scan, MRI), are helpful and are useful, in particular, in planning the best therapeutic strategy for the individual patient.
Surgery is the treatment of choice and includes either endoscopic or external surgical procedures.
The choice of these surgical approaches is based on several characteristics of lipoma (site, size, submucosal growth).
Total removal of these lesions is important to avoid recurrences, a possibility that has been described in the Literature.
The possibility of recurrence after long free-intervals should be borne in mind; thus an extended period of long-term follow-up is mandatory.

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