

HEAD AND NECK SECTION

Treatment of relapses of benign latero-cervical pathology: a narrative literature review

Trattamento delle recidive della patologia benigna latero-cervicale: revisione narrativa della letteratura

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SUMMARY

Benign laterocervical pathologies are not without pitfalls. Many may relapse after many years and, sometimes, they cannot be predicted. The purpose of this review is to describe the surgical measures necessary for the treatment of relapses of the most common benign laterocervical masses. We searched PubMed, Embase and Cochrane Central Register of Controlled Trials databases for articles describing the treatment of the most common benign cervical disease recurrences, and summarised available evidence in this narrative review. We overviewed observations about recurrent benign mixed tumour (pleomorphic adenoma), parapharyngeal space tumours and carotid body paragangliomas, thyroglossal duct anomalies and branchial cleft anomalies. Proper surgical technique is crucial for safely and effectively managing the relapses of benign latero-cervical diseases. Radiotherapy is indicated in several cases of recurrence such as pleomorphic adenoma and unresectable paraganglioma. Long-term follow-up is of utmost importance to promptly recognise and treat recurrences.

KEY WORDS: benign pathologies, laterocervical relapse, parotid gland tumor, paragangliomas, thyroglossal duct, branchial cleft

RIASSUNTO

Le patologie benigne laterocervicali non sono prive di insidie. Molte di esse possono andare incontro a recidiva a distanza di molti anni e, talvolta, non è possibile prevedere se questo accadrà. Lo scopo di questa review è di descrivere gli accorgimenti chirurgici necessari al trattamento delle recidive delle più comuni masse benigne laterocervicali. Sono stati analizzati gli articoli scientifici inerenti al trattamento delle recidive delle più comuni patologie laterocervicali nei database internazionali PubMed, Embase e Cochrane Central Register of Controlled Trial, e sono state sintetizzate le evidenze nel presente manoscritto. In questa revisione narrativa sono state descritte le osservazioni riguardanti il trattamento delle recidive delle patologie benigne laterocervicali. In particolare, sono stati inclusi il tumore misto benigno (adenoma pleomorfo), i tumori dello spazio para-faringeo e i paragangliomi carotidei, le anomalie del dotto tireoglossale e quelle degli archi branchiali. Una tecnica chirurgica appropriata è essenziale per gestire in sicurezza e con efficacia le recidive delle patologie benigne laterocervicali. La radioterapia è indicata in alcuni casi di recidive come l'adenoma pleomorfo e il paraganglioma non resecabile. Il follow-up a lungo termine è fondamentale per una precoce diagnosi e il trattamento delle recidive.

PAROLE CHIAVE: patologie benigne, recidive laterocervicali, tumori della ghiandola parotide, paragangliomi, dotto tireoglossale, archi branchiali

Introduction

Patients presenting with laterocervical masses are usually relieved when diag-

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nosed with a benign disease. However, although they rarely metastasise, these conditions are not without pitfalls such as recurrences. Communication with the patient is essential for a successful diagnostic-therapeutic process, since compliance during follow-up is essential to guarantee the correct treatment and to identify relapses of the disease.

The most common benign laterocervical pathologies include pleomorphic adenoma (PA), also called benign mixed tumour, tumours of the parapharyngeal space including paragangliomas of the carotid body, thyroglossal duct cysts and brachial cleft anomalies. Parotid gland tumours are 3% of all head and neck tumors and PA is the most common¹. Parapharyngeal space tumours account for only 0.5% of head and neck cancers and 80% are benign^{2,3}. Salivary gland neoplasms are the most common followed by neurogenic ones⁴. The most frequent congenital anomaly of the neck is the thyroglossal duct cyst, which affects 7% of the population⁴. Branchial cleft anomalies are 30% of congenital neck masses and can present as cysts, sinuses or fistulas, and usually become symptomatic in the first few years of life^{4,5}.

The purpose of this review is to analyse the surgical techniques for treatment of relapses of the most common benign laterocervical conditions.

Materials and methods

Search strategy

In this narrative review of the literature, we searched PubMed, Embase and Cochrane Central Register of Controlled Trials for articles about the treatment of the benign cervical disease recurrence. The diseases we included in our research were: pleomorphic adenoma, thyroglossal duct cyst, branchial cleft anomalies and carotid body paraganglioma. We also hand searched through the bibliographies of articles to find relevant papers.

Eligibility and inclusion criteria

Prospective and retrospective studies, both randomised and non-randomised, as well as literature reviews and case series, were included. Case reports and rare diseases were excluded. We analysed articles with no time limit and excluded non-English papers. At least two authors for each topic independently searched the evidence in the literature for each disease.

Data extraction

We screened the selected papers by title and abstract, and then considered the entire text of the selected articles to search for evidence and descriptions of the surgical tech-

niques to be adopted in the treatment of relapses of laterocervical benign diseases.

All authors discussed evidence and the conflicts were resolved by our senior author MdV. We summarise our conclusions in the present manuscript.

Results

From the initial search, after removal of duplicates, 977 articles were identified for recurrent benign mixed tumour, 317 for recurrent parapharyngeal space tumour, 281 for carotid body paraganglioma, 302 for thyroglossal duct anomalies and 238 for branchial cleft anomalies. We screened the articles by title and abstract, and then read the full text and identified 48 papers as relevant to our review.

Recurrent benign mixed tumour (Pleomorphic Adenoma) of the parotid gland

PA accounts for 65% of benign parotid tumours⁶. Ultrasound is the first-level examination in the diagnosis of parotid tumours. It allows to determine the characteristics, anatomical relationships, dimensions and acts as a guide for the execution of a needle aspiration⁷. One of the best indicators of tumour proximity to the facial nerve can be detected with MRI or ultrasound using the retromandibular vein as a reference⁷. MRI is the technique of choice in recurrent forms of PA and can sometimes be useful in primary forms⁸.

Psychogios et al. indicated radiological/ultrasound criteria for choosing the type of intervention. They defined the indication to perform extracapsular resection of the tumour for masses ≤ 2 cm in size as “ideal”, especially if they are palpable, mobile and not in contact with the facial nerve, except for tumours near the Stenos duct. In all other cases, superficial or partial parotidectomy is recommended. Total parotidectomy is reserved for malignant disease and some benign tumours of the deep lobe⁷.

Local recurrence of benign mixed tumour of the parotid gland may occur in an unifocal or multifocal fashion. The latter is more common in patients who have undergone superficial parotidectomy; multifocal recurrence is the most common and has usually nodules involving the overlying skin. The histopathological features associated with an increased risk of recurrence are a predominant myxoid composition and extracapsular extension^{9,10}. Rupture of the capsule during surgical excision, the presence of satellite lesions or pseudopodia, and positive resection margins are risk factors for the development of recurrence.¹¹ The relapse rate with a non-enucleation procedure is between 1% and 4%, and are typically observed after 7–10 years from surgery^{12,13}.

The multinodular recurrent PA has usually microscopical seedings even if the number of nodules is limited. They

usually involve the facial nerve, especially if it was dissected during primary surgery. Given the complexity of execution, the increased risk of facial nerve damage, the need for extensive resections and, sometimes, reconstructive surgery, some authors suggest observation instead of surgery in elderly patients ^{1,8}.

TREATMENT

Preservation of the facial nerve is much more complex in surgery of recurrence of PA. This is due to the formation of scar tissue which makes it more difficult to isolate the nerve. In addition, the loss of normal planes further complicates the ability to locate the nerve. For these reasons, some authors suggest identifying the nerve in the mastoid segment or with retrograde technique when performing revision surgery. It is also recommended to remove the scar of the previous surgery “en bloc” with the neoformation ¹.

Rates of facial nerve dysfunction increase with each revision procedure due to previous dissection making it difficult to distinguish the facial nerve from scar tissue.

Some techniques are reported in literature to avoid facial nerve lesions during retrograde facial nerve dissection ⁸. The dissection starts from the temporal facial nerve branch, which is generally the more undisturbed branch in the first surgery. In parotid surgery, intra-operative facial nerve monitoring (IFNM) is of great value in the treatment of recurrences due to the greater difficulty encountered in locating the nerve in scar tissue. Thanks to the use of neuromonitoring, it is possible not to expose the branches of the nerve, decreasing the incidence rate of permanent facial paralysis and shortening the recovery time of post-operative facial nerve function. Removal of a recurrent PA can be accomplished by reducing mechanical manipulation and surgical trauma to the nerve ⁸. Some authors have observed

Table I. Characteristics of the main laterocephal benign pathologies.

Disease	Symptoms	Relapse risk factors	Treatment	Surgical pitfalls	Ref
Pa	Mass in the parotid region	Surgical risk factors: histopathological risk factors: predominant myxoid composition and extracapsular extension	Radiation therapy. Superficial or total parotidectomy with retrograde nerve identification, removal of the overlying skin and previous scar. Flap reconstruction may be required	Complex preservation of facial nerve	6, 9, 10, 15, 16
MPA	Mass in cervical lymph nodes, metastasis	Not known	Surgical removal of masses, total parotidectomy, neck dissection. Radiation therapy	Poor prognosis	19-22
Parapharyngeal space tumours	Cervical or intraoral mass	Incomplete excision	Surgical excision of the mass and eventual lymph nodal metastasis. TORS. Radiotherapy in case of non-resectable lesion, involvement of the internal carotid artery and negative balloon occlusion test, patients with contralateral vagus or hypoglossal nerve deficiency		3, 25, 26, 34, 36-39, 42
Carotid body paraganglioma	Cervical mass	Incomplete excision	Surgical excision with pre-operative embolisation and eventual adrenergic blockade		33
Thyroglossal duct cyst	Cervical mass in the midline, infection of the mass	Incomplete excision usually with plan removal	Sistrunk procedure	Extensive resection of the duct	4, 40-42
First branchial cleft anomalies	Swelling in pre-, infra- or post-auricular region	Incomplete excision	Surgical excision.	Excise skin and cartilage of the external auditory canal. Dissection from the facial nerve	43, 46, 47
Second branchial cleft anomalies	Swelling between the SCM and pharynx	Increasing their size after upper respiratory infections that leads torticollis or dysphagia	Surgical excision	Cannulation of the tract with a 2-0 ore 3-0 monofilament suture probe or the injection with methylene blue	4, 5, 42, 43
Third and fourth cleft anomalies	Hypoglossal nerve palsy in case of infection or lower neck swallowing	Incomplete excision	Surgical excision	Fourth arch anomalies often require hemithyroidectomy to completely excise the mass and, sometimes, resection of thyroid cartilage to expose the pyriform sinus	4, 42, 43, 50

PA: pleomorphic adenoma or benign mixed tumour; MPA: malignant pleomorphic adenoma; SCM: sternocleidomastoid muscle; TORS: transoral robotic surgery.

that its use reduces the operating time, the recovery time in case of temporary dysfunction of the cranial nerve septum and the incidence of permanent dysfunction¹⁴.

In case of cutaneous invasion, it is necessary to remove, together with the nodules and any parotid tissue still present, the involved preauricular and cervical skin. In such cases, flap reconstruction is required¹⁵ (Tab. I).

Surgical treatment for PA recurrence can be performed with conventional excision or parotidectomy, depending on the specific case. Usually, this surgery requires a high degree of expertise and more extended surgical approach; superficial or total parotidectomy are the techniques of choice in most cases^{8,16}. Reoperation complicates the preservation of the facial nerve, which must always be sought by the surgeon. In case of nerve damage during the excision of the recurrent PA it is advisable to perform an immediate repair using a nerve graft¹⁷. Adjuvant radiotherapy may reduce the recurrence rate in recurrent PA¹⁸. Witt et al. observed that radiation therapy has better outcomes in controlling recurrent pleomorphic adenoma compared to surgical excision; however, treatment must be individualised depending on the patient's conditions and disease⁸.

MALIGNANT PLEOMORPHIC ADENOMA

PA can rarely metastasise even without malignant histological degeneration; it is called malignant pleomorphic adenoma (MPA) and parotid gland is the most affected site. According to the 2005 classification of the WHO, there are three distinct clinicopathologic types for malignant pleomorphic adenoma: carcinoma ex pleomorphic adenoma (CXPA), carcinosarcoma, and metastasising mixed tumour. MPA is a condition with histological characteristics of PA with the capacity to generate local recurrences and distant metastases (both to local lymph nodes and other organs)¹⁹. Most occur after one or more surgical procedures and histological examination is not predictive regarding the ability to metastasise. The hypothesised aetiology is the cell spreading during surgery^{19,20}.

Nearly 30% of MPA cases are localised in the lymph nodes, and for this reason it is recommended to also perform neck dissection. MPA relapse treatment should involve total parotidectomy with conservation of facial nerve when it is not infiltrated^{19,22}.

Radiotherapy is considered as a complementary treatment in recurrent disease^{19,22,23} (Tab. I).

Despite the histological features of benignity, it has poor prognosis (50% 5-year survival rate); therefore, it is classified as a malignant tumour by the WHO²⁴.

Parapharyngeal space tumours

The parapharyngeal space shape resembles as invert-

ed pyramid, the tip is located at the greater horn of the hyoid bone and the floor at the skull base (petrous portion of the temporal bone and sphenoid bone). Medially there is the buccopharyngeal fascia, laterally the medial pterygoid muscle, the mandible, the posterior belly of the digastric muscle and the retromandibular portion of the parotid gland; anteriorly the pterygomandibular raphe; posteriorly the carotid sheath²⁵. Parapharyngeal space tumours constitute the 0.5% of all head and neck neoplasms². Eighty percent are benign and surgical excision is the standard treatment in most cases^{3,25,26}. The most common are salivary gland tumours, accounting for 40-50% of cases²⁶.

Neurogenic tumours represent about 40% of parapharyngeal space neoplasms and 95% are benign. There are three main subtypes, paragangliomas, schwannomas and neurofibromas. The most common presenting symptom is the growth of a cervical or intraoral mass (50% and 47%, respectively)³. During treatment of parapharyngeal space tumours, it must always be considered that 40% of sporadic and 80% of familial cases have multiple tumours, and that 10% of sporadic and 30% of familial neurogenic tumours are bilateral. Therefore, a careful analysis of MRI and CT images is also necessary in the contralateral area to that in which the symptoms occur to identify small, unrecognised neoformations^{3,25}.

TREATMENT

The main treatment of the primary tumour or relapses is surgical excision. Surgical management should also consider the presence of lymph node metastasis. It is recommended to perform a neck dissection involving at least levels II to IV during resection of these tumours^{3,25,26}. Relapse risk is mostly related to incomplete excision or tumour dissemination during surgery, and accurate follow-up is of utmost importance for early diagnosis and treatment of relapses⁸.

Paragangliomas can also have malignant forms with lymph node metastases, and, as with pleomorphic adenoma, their malignant behaviour is not histologically predictable²⁷.

Along with traditional CT and MRI, it may be useful in cases suspect of malignancy, once a lesion of vascular origin has been excluded, to perform a fine needle aspiration. The approach can be both pre-oral and transcervical. The result is useful to characterise the lesion in the treatment of both primary neoplasms and relapses^{28,29}.

To avoid recurrence in case of incomplete resection, adjuvant therapy can be considered²⁵.

In case of uncomplete paraganglioma resection, radiation therapy is a valid option. Low radiation doses are suf-

ficient to stop tumour growth with a low incidence and severity of adverse events. However, in most cases low radiation doses do not eradicate the mass^{8,30-32}.

Less than 10% of paragangliomas are malignant; however, as in the case of pleomorphic adenomas, their malignancy cannot be predicted on the basis of histological features^{24,33}. It is necessary to carry out accurate and prolonged follow up.

In case of recurrent disease for which neck dissection was not performed in the previous surgical treatment, it is recommended that the patient undergo the procedure.

The criteria for ineligibility for surgery are the same as for primary forms such as unresectable lesion, involvement of the internal carotid artery and negative balloon occlusion test, or contralateral vague or hypoglossal nerve deficiency. In these cases, the therapeutic choice must lean towards radiotherapy³⁰.

The selection of the appropriate surgical approach is similar to the one used for primary lesions. The most widely used approach is the trans-cervical one. However, cervical-parotid, mandibular split, trans-cervical trans-mastoid and infratemporal approaches can also be used depending on tumour localisation and size³⁴. These techniques can also be combined. It is sometimes necessary to perform a mandibulotomy for better exposure³⁵. Intra-operative monitoring of the facial nerve can also be useful for preserving facial motility³⁶.

TORS can be used in patients with good oropharyngeal exposure and neoplasm with a defined cleavage plane from neurovascular bundle. This technique is associated with increase in capsule disruption percentage, but no increase in the recurrence rate³⁷⁻³⁹. TORS has an advantage in aesthetic terms. The blemish produced by a cervicotomy is often poorly accepted by patients, especially if they are young. Being able to propose a safe treatment, which does not increase the risk of relapse and does not undermine the patient's quality of life and aesthetics, represents a great advantage in terms of compliance and acceptance of the intervention.

Surgery is not the treatment of choice in case of patients that fail balloon occlusion, the elderly, or unresectable tumors that require sacrifice of cranial nerves. In such cases, the available options include watchful waiting and radiation therapy²⁵ (Tab. I).

CAROTID BODY PARAGANGLIOMAS

Carotid body tumour paragangliomas are vascular lesions that splay apart internal and external carotid arteries without narrowing them. They usually absorb a large amounts of contrast medium and have a "salt and pepper" appearance on T2 weighted images. Since they are neuroendo-

crine neoplasms, they have receptors for somatostatin and have intense indium-111 octreotide uptake³³.

The treatment for carotid body tumour paragangliomas is surgical resection, and it is prudent to perform a pre-operative embolisation and consider adrenergic blockade. After complete surgical resection, recurrence and metastasis may occur years later. Surgery remains the treatment of choice in most cases since chemo- and radiotherapy do not have significant benefits³³ (Tab. I).

Thyroglossal duct anomalies

Thyroglossal duct cyst is the most common congenital anomaly of the neck, and occurs in 7% of the population⁴. Its origin is from epithelial remnants of the thyroglossal duct, a diverticulum located at the base of the tongue that displaces caudally to the lower neck. This duct usually obliterates at the eighth week of gestation, and its failure results in thyroglossal duct cyst⁴⁰. The most common clinical presentation is a neck mass in the midline which sometimes gets infected and treatment is surgical excision using the Sistrunk procedure^{41,42}.

Once clinical suspicion has been placed, in the primary diagnosis it is recommended, together with physical examination, to perform ultrasound and evaluation of the thyroid stimulating hormone (TSH). If median ectopic thyroid is suspected, usually in the case of a solid mass or hypothyroidism, it is advisable to perform a thyroid scan before removal⁴³. Although some authors assert that thyroid scintigraphy should be performed in all patients due to the risk of ectopic thyroid^{42,43}, MRI or CT are recommended only in complicated cases and relapsing forms.

A relapse can be suspected, especially in patients who underwent plain excision instead of a Sistrunk procedure, which has a significantly lower recurrence rate (5.3% vs 55.6%). Post-operative infection is also associated with recurrence⁴⁰. Relapses can be removed surgically with extensive application of the Sistrunk technique. On occasion, trans-oral removal of the cyst is also useful (e.g., tongue)⁴⁴. Thyroglossal duct cyst relapses can occur in the area between the blind foramen of the tongue and the thyroid gland. The most effective technique to avoid thyroglossal duct cyst recurrence is to choose the correct surgical technique, the Sistrunk procedure. Head and neck surgeons should avoid plain excision because of the high risk for relapses. Sistrunk technique, with hyoid bone body excision, is the key to avoid relapses and treat them⁴⁰. An accurate intra-operative exploration of the thyroglossal duct cyst tract reduces the risk of leaving residues of the duct.

OK-432 (Picibanil) injection after cyst fluid aspiration showed encouraging results in a paediatric population with recurrent thyroglossal duct cyst with no need of surgery or anaesthesia (either general or local)⁴⁵ (Tab. I).

Branchial cleft anomalies

Branchial cleft anomalies represent 30% of congenital neck masses⁴. By the end of the fourth gestation week there are four pairs of arches and two rudimental arches separated by clefts externally and pouches internally. These latter gradually obliterate, and their incomplete obliteration leads to branchial anomalies⁴³. They can present as cysts, sinuses, or fistulae. Cysts have no external opening, sinuses have an external opening, and fistulae involve a communication with the pharynx resulting in a pharyngocutaneous fistula⁴. Diagnosis of branchial arch anomalies includes history and physical examination accompanied by endoscopy to rule out pharyngeal communication. For the same reason, the piriform sinus and tonsillar cough should be carefully examined during this procedure. Once the lesion has been identified, fine needle aspiration is recommended in adults to exclude laterocervical metastases of cervix-cephalic neoplasms, while it is not recommended to perform this procedure in children.

The radiological investigation of choice is CT, but ultrasound and MRI can also be helpful in some cases⁴³.

FIRST ARCH ANOMALIES

First arch anomalies are only 1% of all branchial cleft anomalies⁵. The first brachial arch is also called the mandibular arch and forms the maxillary process of the upper jaw and part of the inner ear. The first cleft and pouch form the external auditory canal, eustachian tube and middle ear, and mastoid air cells. Consequently, first cleft anomalies involve these structures. Consequently, these structures must be explored both clinically and radiologically before proceeding to surgery. They can be located above, between or below the facial nerve and are classified as Type I or II. Type I lesions course lateral to the facial nerve, and manifest as swelling near the ear. Type II lesions pass medial to the facial nerve, and can present as a preauricular, infra- or postauricular swelling anterior to the sternocleidomastoid muscle^{4,5,43}. It has been observed that fistulas with respect to the sinuses run more frequently in depth than nevus and that young people have more frequent lesions deep to the seventh cranial nerve. Furthermore, open lesions in the external auditory meatus are usually superficial to the nerve. In any case, it is advisable to always locate the main trunk of the facial nerve early during surgery⁴⁶.

To avoid and treat a recurrence of first arch anomalies, it is necessary to excise the skin and cartilage of the external auditory canal involved by the mass. If the tract goes medial to the tympanic membrane, a second surgical procedure may be necessary to remove it. In the latter case, the tract lies deep to the facial nerve or can split around it^{43,46,47}. Disease relapse is common, and the average num-

ber of procedures necessary to achieve complete resection are 2.4 per patient⁴⁷. In case of recurrence, it is therefore necessary to carefully explore the patient's ear both clinically and radiologically.

In particular, it is important to subject the patient to MRI, CT, and otomicroscopy before surgery⁴⁸.

In case of recurrent disease, it is fundamental to perform a radicalisation surgery by extending the resection to the external and middle ear in case of extension to those areas (Tab. I). Each repeated surgery increases the risk of facial nerve injury due to fibrosis^{5,47}.

SECOND CLEFT ANOMALIES

Second cleft anomalies represent 95% of all branchial cleft anomalies⁴. The second arch evolves in hyoid bone and structures of the adjacent area. The anomalies are classified in four categories. Type I are anterior to the sternocleidomastoid muscle (SCM) and not contacting the carotid sheath. Type II lesions are deep to the SCM and anterior or posterior to the carotid sheath, and are the most common second cleft anomalies. Type III lesions are adjacent to the pharynx and pass between the external and internal carotid artery. Type IV lesions are between the carotid sheath and the pharynx, close to the tonsillar fossa.

Second cleft anomalies are diagnosed after increasing their size after upper respiratory infections that leads to torticollis or dysphagia^{4,42,43}.

In case of recurrence, it is essential to explore the fistula tract and excise it. Several techniques have been proposed to make this procedure easier, such as cannulation of the tract with a 2-0 or 3-0 monofilament suture probe or injection with methylene blue. Spinal accessory and the hypoglossal and vagus nerve are at risk of injury, in particular in surgery of relapses because of scar. It can also be useful to explore the tract from the tonsillar fossa using a finger to better expose it^{5,43} (Tab I).

THIRD AND FOURTH CLEFT ANOMALIES

Third and fourth branchial pouches form the pharynx below the hyoid bone. Sinuses and fistulae deriving from these structures are rare and usually enter the pyriform sinus. The fourth arch anomalies on the right-side loop around the subclavian artery and enter the pharynx at the pyriform apex of cervical oesophagus. On the left, they loop around the aortic arch and ascend into the neck to enter the pyriform apex of the cervical oesophagus^{4,42,43}. The clinical manifestation of the third arch cyst causes hypoglossal nerve palsy in case of infection or lower neck swallowing.

For the diagnosis of anomalies of the second and third branchial arch, in addition to CT and sometimes ultrasound and MRI, a barium oesophagogram can be useful with a sensitivity of up to 80%⁴⁹. For anomalies of the fourth arch,

together with CT, endoscopic examination must be performed with particular attention to the piriform sinus with which the fistula could communicate⁵⁰.

The surgical treatment is the excision of the mass, usually with a cervicotomy approach. Fourth arch anomalies often require hemithyroidectomy to completely excise the mass and, sometimes, resection of thyroid cartilage to expose the piriform sinus⁵⁰ (Tab. I).

Conclusions

There is increasing evidence in the scientific literature of recurrence of benign latero-cervical pathologies even more than 10 years from the first excision. Communication with the patient is essential to obtain the necessary compliance for long-term follow-up, which is of utmost importance to promptly recognise and treat recurrences.

Surgical excision of recurrences should be performed by experienced surgeons because they usually require high expertise, and need some precautions to avoid further re-interventions. Radiation therapy has shown to be effective in the treatment of recurrent pleomorphic adenomas and to reduce the growth of unresectable paraganglioma.

Conflict of interest statement

The authors declare no conflict of interest.

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Author contributions

PGM: writing original draft, supervision; AC: writing original draft and critical revision of the manuscript; AM: supervision; DA: literature review; FC: literature review; MR: critical revision of the manuscript; MdV: supervision.

Ethical consideration

Not applicable.

Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

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