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CASE REPORT

Leiomyosarcoma in sternocleidomastoid muscle: a case report

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Abstract

Introduction: Leiomyosarcoma (LMS) is rare, although commonly described in oral cavity. It can appear anywhere smooth muscle is present and there are reports of this type of neoplasm in many subsites in head and neck. **Case Report:** 44-year-old male patient with 2-year-history of cervical nodule, progressively growing, causing pain and neck movement limitation. Patient underwent *en bloc* resection of the tumor along with right ECM muscle, cervical levels II, III, IV and V. **Discussion:** Primary resection with wide margins is the treatment of choice for localized lesions. The present case originating in ECM may be unique until now, but it was handled based on the available literature and presented a satisfactory outcome.

Keywords: sarcoma; head and neck neoplasms; neck muscles; neck dissection

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Introduction

Leiomyosarcoma (LMS) is rare in head and neck region[1], although commonly described in the oral cavity. It can appear anywhere smooth muscle is present and there are reports of this type of neoplasm in the hypopharynx, oropharynx, external auditory canal, nasal cavity and maxillary sinus[2].

Generally, they are slow-growing asymptomatic lesions, with well-defined borders and non-ulcerated[3], which can be confused with benign lesions. Most symptoms result from impaired organ function, such as impaired speech and swallowing, in oral cavity cases, and nose breathing obstruction, in nasal cavity cases.

The recommended treatment is usually surgical resection with free margins[1,3].

Case report

A 44-year-old male patient with a history of type I neurofibromatosis, previous resection of a nasal cutaneous neurofibroma 10 years ago and severe scoliosis, complaining of a cervical nodule, progressively growing for 2 years, causing pain and neck movement limitation in the last 6 months, symptons that motivated the search for medical care and head and neck

surgeon referral. Physical examination showed a semi-fixed hard mass of about 8 cm, painless on palpation, associated with a hypervascularization area in the skin covering lesion´s most protruding portion (Figure 1). A cervical computed tomography with venous contrast showed a soft tissue density mass with heterogeneous contrast uptake, measuring 10.55 x 9.44 x 8.8 cm. The right sternecleidomastoid muscle (ECM) was diagnosed as disease´s epicenter (Figure 2). There were no palpable atypical lymph nodes, nor lesions in the oral cavity, oropharynx or other body regions on physical examination, neither on imaging exams.

The patient did not present any complaints other than those described, nor did he present changes in laboratory, electrocardiogram, endoscopic and chest tomography exams. After a multidisciplinary discussion, considering the malignant lesion hypothesis and the resectability of the lesion with free margins without greater morbidity to the patient, surgical resection was chosen as primary treatment.

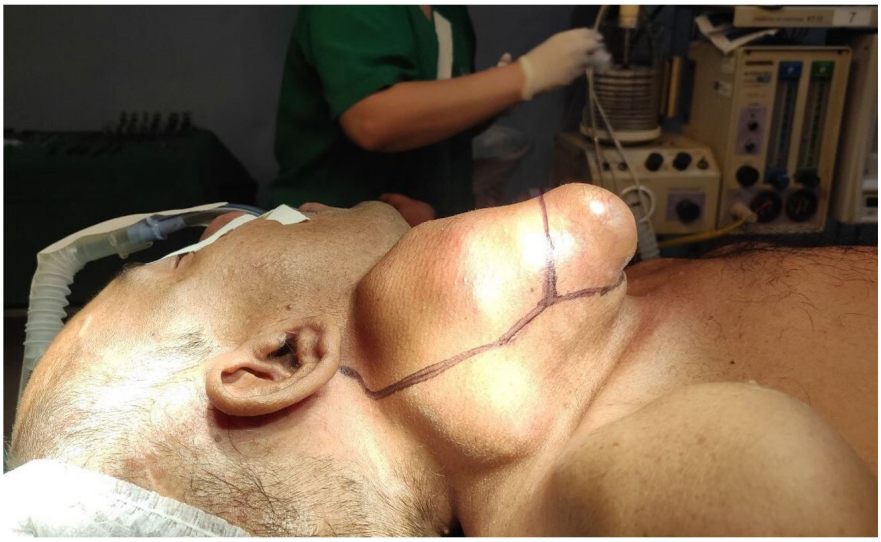

Figure 1. Cervical mass with hypervascular skin island (ressected en bloc with surgical specimen).

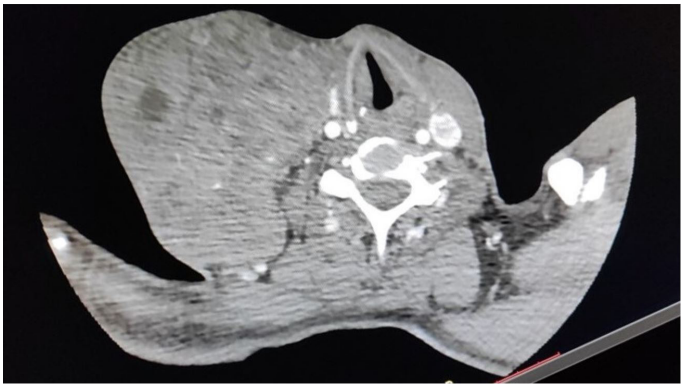

Figure 2. Tomography scan image showing normal left sternomastoyd muscle and leyomissarcoma comprising right sternomastoyd muscle.

Patient underwent *en bloc* resection of the tumor along with right ECM muscle, cervical levels II, III, IV and V on the same side. Right spinal accessory nerve and an apparently compromised skin island were included in ressection. The mass had a hard and friable consistency, it had well defined borders located in the upper two thirds of right ECM. Due to the tumor driven skin's expansion effect, it was possible to do a primary synthesis of the resulting skin defect.

It was reported intermediate grade LMS diagnosis after histology showing eosinophilic spindle cells fascicles arrangements intersecting each other, differentiation grade 2, 12 mitosis/10 High-power fields (HPF) and tumoral necrosis up to 50%, meeting French Federation of Cancer Centers Sarcoma Group (FNCLCC) system criteria. No metastatic lymph nodes where found among 5 in level II, 8 in level III, 3 in level IV and 12 in level V. The tumor was 11 x 9.5 x 9 cm and it has comprised cervical level III on right side as well as adjacent ECM. Skin was not microscopically involved, there no angiolymphatic nor perineural invasion and resssection margins were free. Immunohistochemistry pannel has demonstrated positivity for desmin and smooth muscle actin. Positive Ki-67 around 15% showed accelerated cellular growth, whereas negative S100 was useful to exclude neural sheath origin.

There were no adverse events in postoperative period and the patient did not undergo adjuvant therapy. By now, he has been in clinical follow-up for 08 months, with no evidence of disease.

Discussion

There are few LMS reports in head and neck, with only 189 between 2000 and 2018[1], with no case originary in cervical muscles. This report presents a case in ECM, most likely originated from smooth muscle cells of a intramuscular vessel´s wall, which can be seen but is not mandatory for the diagnosis[4]. The authors considered ECM as the origin, due to the findings in physical examination, imaging exam and intraoperative observation, which showed an entirely continuous lesion into the muscle and a totally disease-free right sternal jugular vein (Figure 3). Other possible origins reported in the literature are: skin's erector pilli musculature, circumvallate papillae of the tongue and salivary glands' myoepithelial cells.

There is no consensus on wich age and/or sex are more likely to have LMS. With regard to age, there are two peaks of incidence: under 50 years-old and over 80 years-old[5]. However, there is no significant difference in incidence between male and female[1,5].

Due to the rarity of this type of tumor, usually, systematic reviews, large centers-based retrospective studies and case reports guide treatment definition. Primary resection with wide margins is the treatment of choice for localized lesions[2,3], whereas neck dissection is restricted to cases in which there is lymph node involvement in clinical staging[4,5]. It was decided to perform neck dissection in the present case because there was macroscopic suspicion of metastatic lymph nodes at right level Vb during resection, which was excluded by the pathologists.

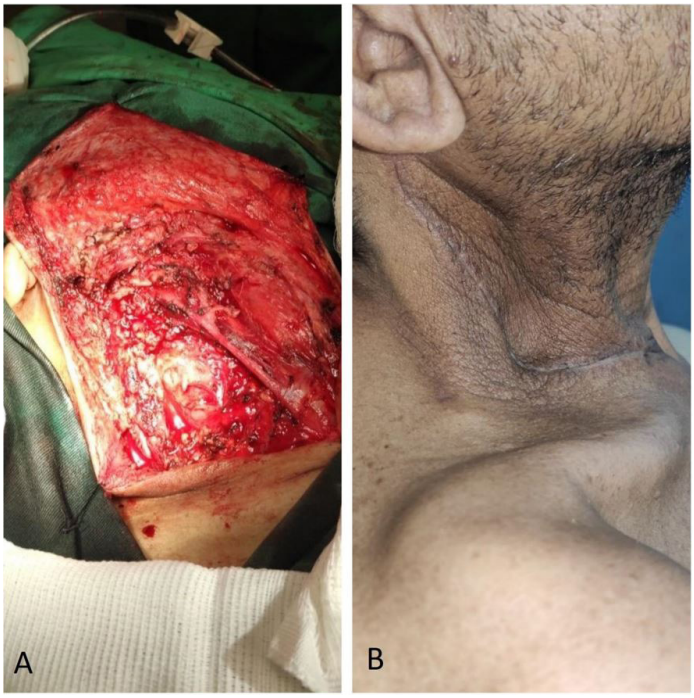

Figure 3. A - Intraoperative aspect after mass excision (right internal jugular vein preserved). **B -**: 6 months after surgery during clinical follow-up consultation.

According to most authors, prognosis of patients treated primarily through surgery with wide margins is better than those who have undegone non-surgical treatment. However, one must interpret these conclusions carefully, as there is a paucity of prospective clinical studies of LMS and studies available are mostly retrospective, though, they are likely to have indication bias. For a rare condition, prospective studies are difficult to be performed. In addition, different head and neck subsites affected by LMS makes casuistry accessment an even more arduous mission. The present case originating in ECM may be unique until now, but it was handled based on the available literature and presented a satisfactory outcome.

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