The surgical management of a leiomyosarcoma of the submandibular gland in a 95-year-old patient

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Primary sarcomas of the major salivary glands are rare and appear to originate from undifferentiated pluripotential mesenchymal cells. They must be distinguished from malignant supporting tissue neoplasms that secondarily involve the glands by direct extension or metastasis. Multidisciplinary management of head and neck soft tissue sarcomas is still controversial. We report a case of leiomyosarcoma of the submandibular gland in a 95-year-old man who was treated with excision of the right submandibular gland, extended to the surrounding tissues, without neck dissection. The patient tolerated the treatment well. Twenty-four months after surgery, the patient was doing well without any evidence of locoregional or distant disease. Surgery is the cornerstone of the management of leiomyosarcomas of the salivary glands. Wide surgical excision with histologically proven tumor-free margins was an appropriate treatment that may guarantee prolonged survival. **(Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;112:e34-e38)**

Leiomyosarcoma (LMS) is a relatively rare mesenchymal tumor that represents 5%-7% of all soft tissue sarcomas.[1,2] They usually occur in the uterus, gastrointestinal tract, and retroperitoneum, whereas only 3%-10% of all LMSs arise in head and neck.[1,3,4]

Primary sarcomas rarely arise in major salivary glands.[5] These tumors appear to originate from undifferentiated pluripotential mesenchymal cells and must be distinguished from malignant supporting tissue neoplasms that secondarily involve the glands by direct extension or metastasis.[6] Similar to their soft tissue counterparts, prognosis is based on the size, location, histologic grade, and complete resection of the tumor.[6] Surgery is the primary therapeutic approach for the management of head and neck sarcomas in all ages with some exceptions in the pediatric population.[7]

In this article, we present and discuss the management of a case of a primary LMS of the submandibular gland in a 95-year-old patient.

CASE REPORT

A 95-year-old white man was referred to the Division of the Maxillofacial Surgery, San Giovanni Battista Hospital, University of Turin, Turin, Italy, for evaluation of a right submandibular swelling that had appeared 6 months before. Clinical history revealed the presence of hypertension, an acute myocardial infarction that occurred 5 years before, and the presence of an atrioventricular block treated by pacemaker implantation.

Clinical examination revealed a right submandibular, firm, painless mass that was nontender to palpation (Fig. 1). There was no cervical lymphadenopathy.

Ultrasonography showed an expansile, solid, hypoechoic, heterogeneous mass measuring ~4 × 3 cm. It appeared to involve the right submandibular gland. No pathologic lymph nodes were found (Fig. 2).

A contrast-enhanced computerized tomography (CT) scan confirmed the presence of a heterogeneous lesion measuring about 4 × 3 cm involving the right submandibular gland. Enhancement of the mass was slow but progressive. No pathologic lymph nodes were diagnosed (Fig. 3). An ultrasound-guided fine needle aspiration biopsy was performed, which allowed to recognize fascicles of spindle-shaped cells, suggestive for a sarcomatous lesion. The distant metastasis workup (including positron-emission tomography–CT) was negative.

A resection of the right submandibular gland, extended to the surrounding tissues, was performed via submandibular approach under general anesthesia.

Microscopically, the tumor consisted of intersecting bundles and fascicles of spindle cells with ample amount of eosinophilic cytoplasm (Fig. 4). Elongated nuclei with dispersed chromatin and occasional small nucleoli were found. Mitosis averaged 5 per 10 high-power fields, and the stroma was characterized by necrotic foci. Immunohistochemistry demonstrated positive staining of the tumor cells for desmin and smooth muscle actin (SMA; Fig. 5) and negative staining for S-100 protein antigen and CD34.

Therefore, a final diagnosis of low-grade leiomyosarcoma of the submandibular gland was established. The tumor was

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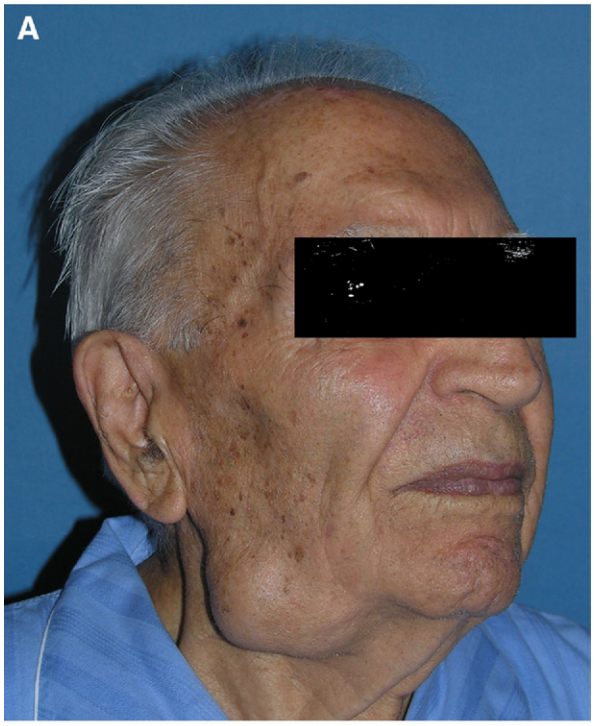

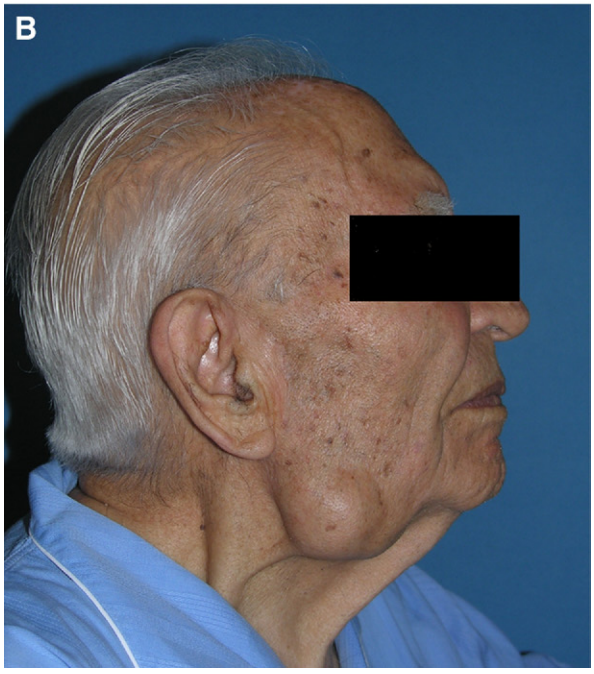

Fig. 1. Preoperative images of the patient: a right submandibular mass can be appreciated.

rated as grade I according to the Federation Nationale des Centres de Lutte Contre le Cancer (FNCLCC) classification.[8]

The patient is currently followed; 24 months after surgery, the patient was doing well without any evidence of locoregional or distant disease.

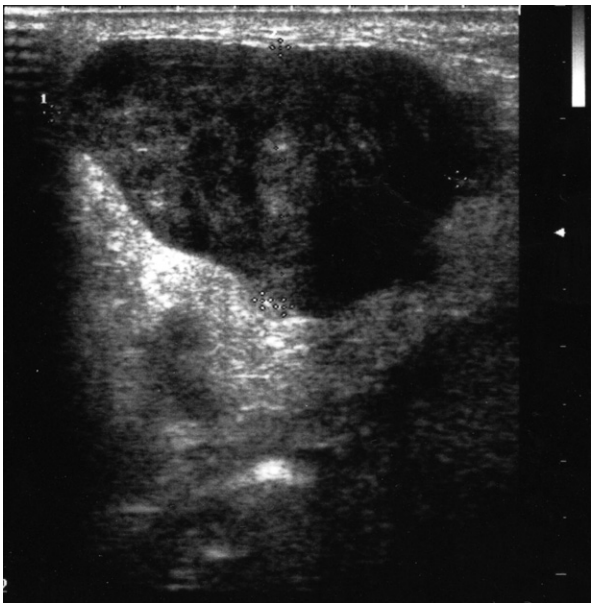

Fig. 2. Ultrasonography finding showing an expansile, solid, hypoechoic, heterogeneous mass involving the right submandibular gland.

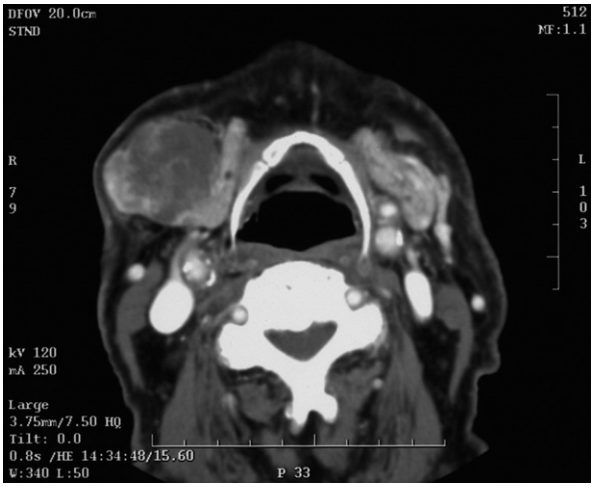

Fig. 3. Contrast-enhanced computerized tomographic scans confirming the presence of a heterogeneous lesion measuring ~4 × 3 cm and involving the right submandibular gland.

This article was exempted from review by our Institutional Review Board human studies committee; we followed the guidelines of the Helsinki Declaration.

DISCUSSION

LMS is a malignant mesenchimal tumor that is considered to be one of the most uncommon types of soft tissue sarcoma in the head and neck.[7]