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METASTATIC LEIOMYOSARCOMA OF THE MANDIBLE: A CASE REPORT AND REVIEW OF THE LITERATURE.

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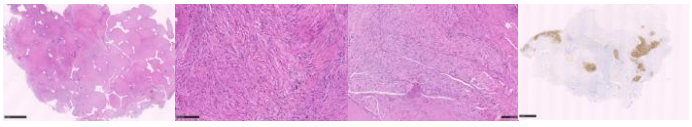
OBJECTIVES

The aim of this case report is to present an unexpected metastatic leiomyosarcoma (LMS) localized to the right mandibular body of a patient treated at the San Paolo Hospital of Milan- Department of Maxillofacial Surgery.

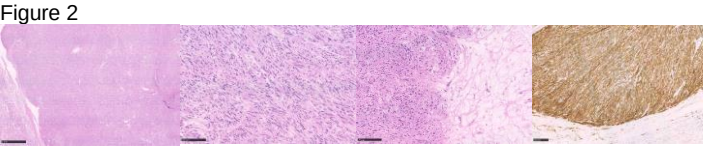
FIGURES

Figure 1



A B C D

Figure 2



CASE REPORT

A 78-year-old man presented to the Maxillofacial Surgery Unit, Head and Neck Department, with persistent odontalgia for 10 months. The orthopantomography showed an osteolytic and radiolucent neoformation with well-defined borders in the right mandibular region, contiguous to the inferior alveolar canal. Additionally, tooth mobility was observed in teeth 46 and 47. No mucosal lesion was evident. The clinical diagnosis was either mandibular cyst or inferior alveolar nerve lesion. The patient reported right adrenalectomy in 2016, without documentation.

The patient underwent extraction of the dental element 46 and simultaneous removal of the endosseous lesion which macroscopically appeared solid. Histologically, the lesion consisted mainly of hypertrophic nervous tissue as a "traumatic neuroma" (figures 1A,C) around which there was a solid proliferation of atypical spindle cells, without necrosis with minimal mitotic activity (figures 1 A,B) (semiquantitative determination of Ki67 less than 5 %). The immunohistochemical profile of the proliferation revealed positivity for smooth muscle actin, caldesmon and desmin (Figure 1D). The lesion was diagnosed as a leiomyomatous proliferation with cytological atypia suggestive of "degenerative" alterations.

Once the histological diagnosis was known, the patient specified that the adrenalectomy he underwent was due to the removal of a retroperitoneal leiomyosarcoma of medium degree of differentiation (G2) (Figure 2). Together with the comparison with the aforementioned lesion, the degenerative aspects previously detected in the mandibular leiomyomatous lesion were interpreted as a possible metastatic repetition. Magnetic resonance imaging had showed multiple lymphadenopathies and CT-PET-total body pulmonary and hepatic nodulations suspicious for metastases.

DISCUSSION

The occurrence of leiomyosarcoma (LMS) in the oral cavity is very rare, accounting for approximately 1% of all cases of LMS and 6% of cases of head and neck LMS [1,2,3,4,5]. The search on PubMed in the period from 2000 to 2023 using the keywords "jaw leiomyosarcoma" and "metastatic jaw leiomyosarcoma" extracted 24 cases of jaw LMS of which only 2 were well-documented cases metastatic LMS[1,2].
The onset of metastases from leiomyosarcoma worsens the overall survival which drops below 50% at 5 years [1,3,5].
In our case the metastatic nature of the lesion was strongly supported by anamnestic report of former surgical remove of retroperitoneal leiomyosarcoma.

CONCLUSION

The detection of leiomyosarcoma of the oral cavity represents a rare and its metastatic origin is an exceptional occurrence. Nevertheless, leiomyosarcoma is to be considered among the lesions of mesenchymal origin in the oral cavity and also in the intramandibular site. In front of a finding of this type of neoplasm, it is essential to investigate its possible metastatic nature.

REFERENCES

1 Azevedo RS, Pires FR, Gouvêa AF, Lopes MA, Jorge J.Leiomyosarcomas of the oral cavity: Report of a radiation-associated and a metastatic case. Oral Maxillofac Surg.2012; 16:227---32.5.

2 de Araújo, G.R.; Costa, S.F.D.S.; Mesquita, R.A.; Gomez, R.S.; Dos Santos, J.N.; Pontes, H.A.R.; de Andrade, B.A.B.; Romañach, M.J.; Agostini, M.; Vargas, P.A.; et al. Leiomyoma and Leiomyosarcoma (Primary and Metastatic) of the Oral and Maxillofacial Region: A Clinicopathological and Immunohistochemical Study of 27 Cases. *Head. Neck Pathol.* 2022, *16*, 294–303.

3 Lombardi N, Varoni EM, Rabbiosi D, Cucurullo M, Moneghini L, Lodi G. Primary oral leiomyosarcoma of the maxillary bone and sinus: case report and up-to-date review of literature. Braz J Otorhinolaryngol. 2022 Nov-Dec;88 Suppl 4(Suppl 4): S205-S211.

4 Patel K, French C, Khariwala SS, Rohrer M, Kademani D. Intraosseous leiomyosarcoma of the mandible: a case report. J Oral Maxillofac Surg. 2013 Jul;71(7):1209-16.

5 Bala M, Ray A, Saraf A. Leiomyosarcoma of Mandible: A Diagnostic Dilemma; Case Report and Review of Literature. Indian J Otolaryngol Head Neck Surg. 2019 Oct;71(Suppl 1):848-851