Misdiagnosed Uterine Leiomyosarcoma with Metastasis to the Left Condylar Neck - A Case Report

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Abstract

Rationale: This is a case of metastatic leiomyosarcoma (LMS) lesion of the condyle arising from a primary LMS of the uterus in a 43-year-old woman. **Patient Concern:** The patient presented an uncommon clinical manifestation not justified by the presence of a leiomyoma. **Diagnosis:** The diagnostic process revealed that the patient was affected by uterine LMS, previously misdiagnosed as a leiomyoma, with a secondary metastasis of the left temporomandibular joint condyle. **Treatment:** Surgery was proposed with total excision of the lesion and parotidectomy, together with radiotherapy and chemotherapy as adjuvant therapy. **Outcome:** One month after the start of the treatment, the systemic condition of the patient started to quickly worsen, and she passed away due to complications associated with the disease. **Take-away Lessons:** Given the uncommon clinical scenario that the patient presented, guidelines for temporomandibular disorder should also include better indications on how to screen and intercept such possible life-threatening conditions.

Keywords: Condyle, leiomyosarcoma, malignancy, metastasis, temporomandibular joint

Introduction

Leiomyosarcoma (LMS) is a rare malignant mesenchymal neoplasm of smooth muscle origin.[1,2] In the case of temporomandibular joint (TMJ) involvement, the diagnosis can be particularly challenging, considering that the potential symptoms could overlap the manifestation of common temporomandibular disorders (TMDs), such as myofascial pain and arthralgia.

This paper aims to report the case of metastatic LMS lesion of the condyle arising from a primary LMS of the uterus in a 43-year-old woman. To the authors' knowledge, this is the first reported case of metastatic LMS lesion of the condyle.

Case Report

A 43-year-old woman, non-smoker, non-drinker with a family history of breast cancer and a diagnosis of uterine leiomyoma received the year before came to the attention of her family doctor and dentist. The patient's chief complaint was the sudden-onset of excruciating pain in the left pre-auricular area while eating. The intraoral assessment revealed the absence of any dental pathology, and the dentist prescribed an orthopantomography X-ray, from which a fracture of the left condylar neck emerged [Figure 1]. The patient was referred to the Department of Maxillofacial Surgery for further investigations. During the examination, the patient did not report any past or recent trauma to the left side of the face. The clinical assessment revealed the presence of swelling at the level of the left TMJ [Figure 2a and b].

The clinical examination also revealed hypoesthesia of the third branch of the trigeminal nerve, limitation of mouth movement of protrusion and laterality and reduced mouth opening <40 mm [Figure 3a]. However, the facial nerve function was not compromised. No palpable lymphadenopathy was detected at the level of the maxillofacial region. The intraoral exam revealed the presence of premature contact on the left side [Figure 3b].

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No clear diagnosis could have been made from the clinical inspection, and due to the patient's unspecific symptoms, further instrumental examinations were requested: an ultrasonography of the left TMJ and magnetic resonance imaging (MRI) with contrast medium. The MRI revealed the presence of a solid lesion of 33 mm × 34 mm × 36 mm at the level of the ramus and the mandibular condyle, with cortical erosion on both the internal and external sides and involvement of associated soft tissues [Figure 4a and b].

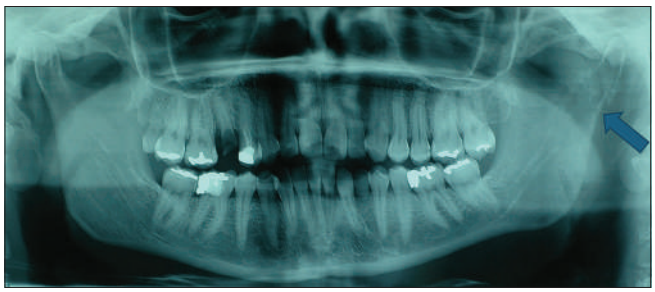

Figure 1: Lytic alteration of the left condylar process, longitudinally extended for 2.5 cm with separation of the cortical bone, compatible with pathological fracture

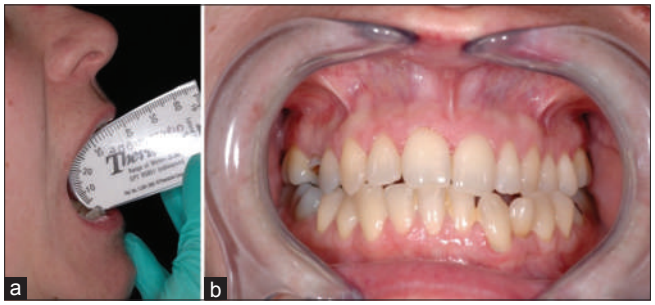

Figure 3: (a) Reduced mouth opening <40 mm. (b) Intraoral view, presence of a precontact on the left side

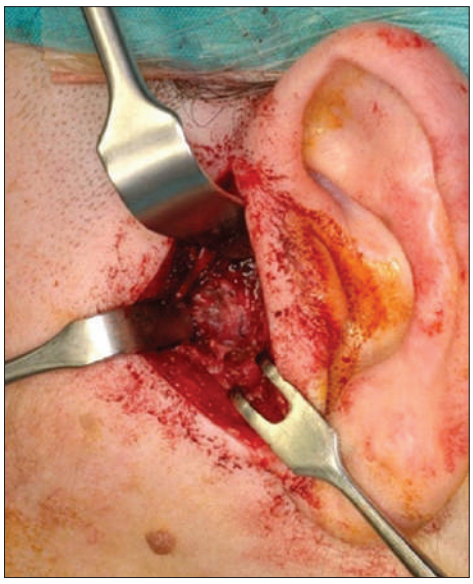

Figure 5: Preauricular access for open biopsy

The post-contrast impregnation unveiled a magnetic resonance image compatible with a primary heteroplastic lesion. However, no effusion was present at the level of the TMJ; the glenoid cavity and temporal eminence of the TMJ were preserved. The diagnostic hypothesis of osteosarcoma was initially formulated. Other conditions considered for the differential diagnosis were chondroma, chondrosarcoma, myoma, myosarcoma, hemangiolymphocytoma and osteoma. An open biopsy with left preauricular access was performed to confirm the diagnosis [Figure 5]. The anatomopathological examination revealed the presence of a LMS.

Due to the low prevalence of primary LMS of the mandible, the hypothesis that the lesion could be a secondary metastasis arising from another anatomical site was made. A second anatomopathologist reviewed the previous histological exams of the uterus leiomyoma, revealing a wrong initial diagnosis and that the patient was affected by a LMS of the uterus. It was subsequently decided to perform a total computerized tomography (CT) of the body to rule out the presence of other metastatic lesions. The investigation did not show the presence of any other secondary lymph node or visceral lesions. At the 1-month follow-up after the biopsy, the lesion further

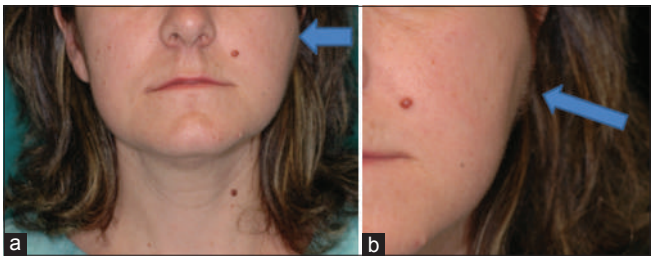

Figure 2: (a and b) Swelling of the left temporomandibular joint

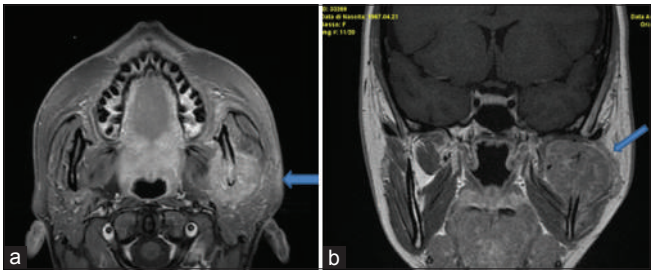

Figure 4: (a and b) Magnetic resonance imaging (MRI) of the patient. MRI compatible with the presence of neoplastic lesion. The expansive lesion is involving the left side of mandibular ramus and condyle

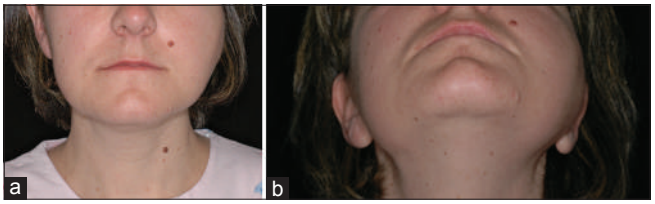

Figure 6: (a and b) Last follow-up pictures of the patient. The swelling increased compared to the picture taken 1 month before

increased in dimension, revealing the aggressive nature of the tumour [Figure 6a and b].

As a therapeutic option, surgery was proposed with total excision of the lesion and parotidectomy, together with radiotherapy and chemotherapy as adjuvant therapy. Eventually, the patient accepted the treatment and received a surgical intervention to remove the neoformation and perform condyle remodelling. The patient subsequently started the oncologic therapy for primary and secondary LMS lesions. Unfortunately, one month after the start of the treatment, the systemic condition of the patient started to get quickly worsen, and she passed away after some further weeks due to complications associated with the disease. Figure 7 shows the timeline of the case report.

Discussion

The present paper reports the case of a patient who, despite reporting common symptoms potentially related to common types of TMDs such as myofascial pain and arthralgia, had a LMS metastasis of the condyle, a very rare and, so far, uniquely described clinical scenario. A recent search in the Surveillance, Epidemiology and End Results database regarding malignant TMJ tumours found a total of 734 cases, of which only 2 were LMS.[3] Moreover, given the widespread distribution of the trigeminal nerve, orofacial neoplasia almost always causes pain.[4] Therefore, when such symptoms not specifically related to TMD are present,[5] life-threatening conditions should always be ruled out since a delay in the diagnosis can have a negative impact on the patient's prognosis. The best way to

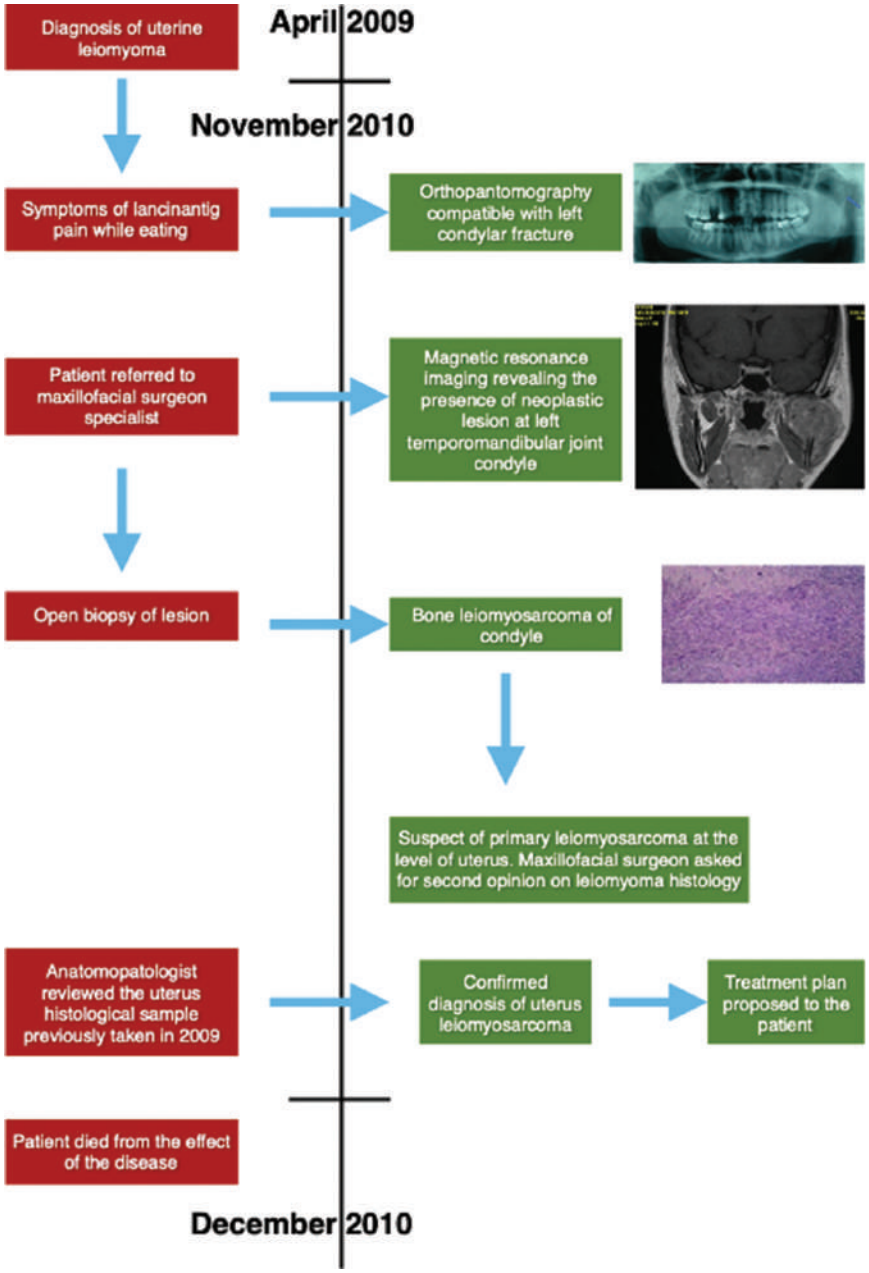

Figure 7: Diagnostic pattern of the case

screen for the presence of such life-threatening conditions, when suspected, is to make use of second-level diagnostic tests such as CT and MRI.

What makes the present case history unique is also the absence of metastases in other parts of the body. It is well-known that jawbone metastases occur only in the last stages of cancer when primary cancer has disseminated to different parts of the body.[6] In the literature, there is no proposal for a possible mechanism of metastasis diffusion from the primary lesion to the TMJ condyle. Considering the presence of bone marrow at the level of the condyle, it could be hypothesized that the possible diffusion happened through a hematologic dissemination.[7] At present, no guidelines could have helped clinicians to reach a final diagnosis in such a clinical scenario since the primary diagnostic system (i.e., diagnostic criteria for temporomandibular disorders, [DC/TMD]) does not contain a specific diagnostic algorithm or scheme to differentiate neoplastic conditions mimicking TMD from other conditions, but just categorise neoplasia as a disorder under the big umbrella of 'joint diseases'. Such a lack could contribute to a diagnostic delay. In this view, the expansion of the DC/TMD taxonomy provides a better classification of the different conditions that can involve the TMJ, suggesting, however, very generic diagnostic tips.[8] Given the previous diagnosis of leiomyoma, the histological results indicating the presence of a LMS of the condyle, and the rarity of primary LMS of the jaw, it was suspected that the origin of the lesion could, in reality, be a secondary metastasis of an LMS. Indeed, further review of the anatomopathological investigations confirmed the real nature of the disease.

Due to the few cases of jawbone LMS reported in the literature, data on the effectiveness of treatment options and prognosis are scarce. Wide surgical excision remains the main treatment option for jawbone LMS, while adjuvant therapies can have a role as a palliative. Chemotherapy alone can increase the survival rate of women affected by metastatic uterine LMS by 8 months. However, the prognosis remains poor. Only 10%–15% of women with metastatic LMS survive at 5 years.[9] Thus, it is possible to hypothesise that an earlier diagnosis would have had a positive impact on survival, with still poor long-term outcomes. In the present case, what contributed to the negative outcome was most likely a combination of the aggressive nature of the LMS together with the delayed diagnosis, which led to inappropriate initial treatment. Recent advances in the field led to the development of promising approaches, including targeting DNA damage repair. However, despite representing a promising solution, such strategies have been tested only at the molecular level, while clinical trials on humans have yet to be performed.[10]

Conclusions

The paper reported a case of a patient affected by uterine LMS, previously misdiagnosed as a leiomyoma, with a secondary metastasis at the level of the left TMJ condyle. Given the uncommon clinical scenario that the patient presented, guidelines for TMD should also include better indications on how to screen and intercept possible life-threatening conditions such as the present case.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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