



# Odontogenic-Like Pain in Partial Edentulism: An Unusual Presentation of Diffuse Large B-Cell Lymphoma of the Mandible

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## Abstract

Primary bone lymphoma is a rare entity and it usually occurs in long bones. Primary mandibular involvement is very rare, and it usually shows unspecific features, mimicking odontogenic inflammatory lesions. We present the unusual case of a diffuse large B-cell lymphoma (DLBCL) of the right mandibular body in a 91-year-old woman, who presented with acute pain in the mandibular region initially suspicious for odontogenic abscess. No significant findings were seen on orthopantomography (OPG) and her almost complete edentulism made the diagnosis of abscess unlikely. Computed tomography and magnetic resonance images showed an expansive mass around the right mandibular body with erosion of cortical bone and involving the right mandibular canal and nerve. Final diagnosis of DLBCL was pathologically proven. The presence of odontogenic-like pain in nearly complete edentulism should be suspicious for malignancy, and it needs further diagnostic workup despite the absence of signs on OPG.

## Keywords

- ▶ computed tomography
- ▶ diffuse large B-cell lymphoma
- ▶ magnetic resonance
- ▶ mandible
- ▶ orthopantomography

## Introduction

Non-Hodgkin lymphomas (NHLs) are the most common lymphoid malignancies in adults. Diffuse large B-cell lymphoma (DLBCL) accounts for 31% out of all NHLs, and it is the most frequent histological type, usually occurring in adults between the sixth and seventh decades.<sup>1,2</sup> DLBCL is a very aggressive lymphoma, but highly responsive to the current therapies, even in advanced stages.

However, the increasing number of elderly patients often makes their management challenging, due to the presence of

comorbidities. Thus, an early and prompt diagnosis is crucial for patient outcome.

Between 25 and 40% of NHLs may have an extranodal spread<sup>3,4</sup> and the head and neck region represents the second most common onset site, after gastrointestinal tract. However, primary NHL rarely occurs in maxilla or mandible accounting only for 0.6% out of all NHLs.<sup>3</sup> These percentages reflect the general low incidence of primary bone lymphomas (~5% out of all extranodal NHL and 2% out of all lymphomas).<sup>5,6</sup>

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Lymphoma of the jaw does not have any specific clinical and radiological features; it often appears as an odontogenic process, thus delaying the diagnosis.

We present imaging findings of a rare case of DLBCL arising from mandibular body in an elderly woman, who was almost completely edentulous.

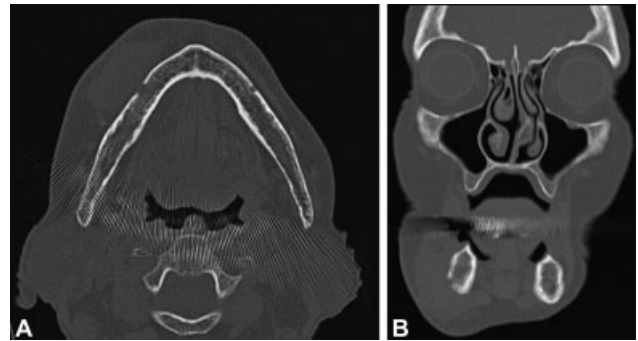
## Case Report

A 91-year-old woman was referred to our center to undergo orthopantomography (OPG) for possible odontogenic abscess. She complained of right mandibular pain for 2 months. Physical examination showed mild swelling in the right mandibular region, without any other signs or symptoms. Laboratory findings were normal, and there were no relevant events in her medical history. The OPG showed almost complete edentulism without other abnormalities, especially in the painful area (►Fig. 1), thus the hypothesis of odontogenic abscess was unlikely. As the pain worsened despite the anti-inflammatory therapy, the patient underwent a computed tomography (CT) without contrast agent injection. CT showed the presence of a 4.3-cm expansive mass centered in the right mandibular body, involving the adjacent submandibular and sublingual spaces. Erosion of the cortical bone and involvement of the right mandibular canal were evident (►Fig. 2A, B). Mandibular nerve, which runs through the mandibular canal, was also involved, thus explaining the pain. No locoregional lymphadenopathies were evident.

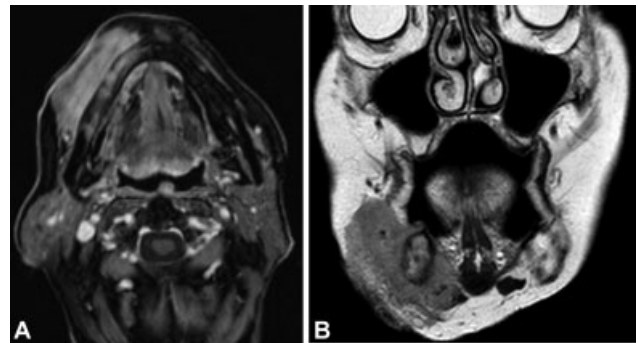
The patient also underwent contrast-enhanced magnetic resonance imaging (MRI) to precisely evaluate locoregional extension of the disease and the possible presence of perineural spread along the mandibular nerve. MRI confirmed the presence of an expansive lesion of the right body of the mandible, involving the mandibular canal and nerve. The lesion showed the same intermediate signal of muscles on T1-weighted images and high signal intensity on fat-suppressed T2-weighted images; mild homogeneous enhancement was evident after administration of contrast agent (►Fig. 3). High signal intensity on high *b*-value was also demonstrated on diffusion-weighted imaging (DWI) with low signal intensity on corresponding apparent diffusion coefficient (ADC) map,



**Fig. 1** Orthopantomography shows the presence of teeth No. 31, 42, and 43 without any periapical lesion at the apex of the teeth suggestive for odontogenic process, particularly in the right body of the mandible. No bone lesion is evident, despite the presence of a light osteopenia.



**Fig. 2** (A) Axial and (B) coronal unenhanced CT images show the presence of a 4.3-cm expansive mass around the right body of the mandible with only a small area of erosion of cortical bone (A and B). CT, computed tomography.



**Fig. 3** (A) Enhanced axial T1-weighted MRI sequence with fat suppression and (B) unenhanced coronal T2-weighted MRI sequence demonstrate the presence of an expansive lesion in the right body of the mandible with mild homogeneous enhancement (A). A mild intramedullary enhancement is visible in the right side of mandible body without a diffuse erosion of cortical bone, due to a permeative diffusion pattern (A). MRI, magnetic resonance imaging.

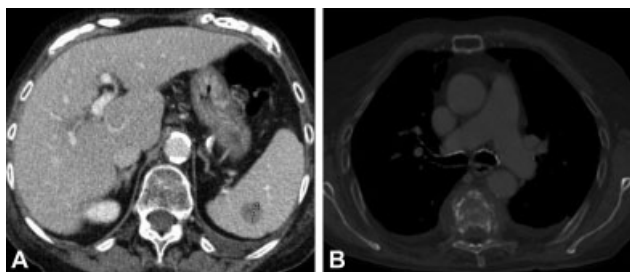
as usually occurs in hypercellular lesions. No signs of perineural spread were evident along the course of inferior alveolar nerve. The lesion also involved mylohyoid, anterior belly of digastric, buccinator, and platysma muscles.

A biopsy of the mass was performed and histological diagnosis of DLBCL with high proliferation rate (>90% Ki-67+) was done.

Contrast-enhanced total body CT was also requested for staging, and it showed the presence of two osteolytic lesions in the seventh and eighth dorsal vertebrae and a 1.8-cm hypodense lesion of the spleen (►Fig. 4). The patient was referred to the hematology department for treatment. After a multidisciplinary counseling, locoregional radiotherapy was started because of the size of the lesion, the presence of pain not responsive to the medical therapy, and the age of the patient.

## Discussion

DLBCL accounts for ~30% out of all NHLs, and it is the most common lymphoma in adults, occurring usually between the sixth and seventh decades.<sup>1,7</sup> DLBCL is classified in different types and subgroups according to morphologic variants, molecular and immunophenotypic features.<sup>1</sup>



**Fig. 4** (A, B) Enhanced axial CT images. (A) CT of the abdomen shows the presence of a 1.8-cm hypodense lesion of the spleen. (B) CT of the thorax with bone window shows the presence of an osteolytic lesion in the seventh dorsal vertebra (A). CT, computed tomography.

About 40% out of all DLBCLs arise from extranodal sites and among these cases, a localized presentation is more frequent than disseminated disease.<sup>1,4</sup> The most common site of extranodal NHL is the gastrointestinal tract and the second is the head and neck region,<sup>3</sup> where NHL occurs more frequently within soft tissues.

Primary involvement of the bone is less common, accounting for ~5% out of all extranodal NHLs.<sup>5</sup> Primary lymphoma of the bone usually involves long bones, as femur, while secondary lymphoma usually affects the axial bones.<sup>8</sup> Maxilla or mandible involvement is even more unusual. The maxilla is more frequently affected and mandibular lesions account for only 0.6% out of all NHLs.<sup>3,9</sup>

Lymphoma of the mandible is very easily misdiagnosed with consequent therapeutic delay due to its rarity and absence of specific signs and symptoms.

On clinical evaluation, it usually presents symptoms more commonly seen in odontogenic diseases rather than malignancies, such as swelling and pain, sometimes with ulceration of the mucosa and teeth mobility. In this scenario, the presence of neurologic signs should be suspicious for malignancies.

The World Health Organization classifies solid mandibular lesions as benign or malignant, according to the predominant odontogenic tissue involved by disease.<sup>10</sup> Both benign and malignant odontogenic mandibular lesions often occur in young people, without acute symptoms (e.g., pain and rapid-onset swelling).<sup>11</sup> In elderly, infectious etiologies (e.g., abscess or osteomyelitis) and primary nonodontogenic lesions (e.g., sarcomas, lymphoma, and metastasis) should be considered.<sup>11</sup>

Radiographic findings on OPG images may be absent in up to 20% of lymphomas,<sup>3,12</sup> and usually they are unspecific. The most common appearance of primary bone lymphomas is a permeative pattern, characterized by spread of tumor cells from the bone marrow through small vascular channels that go through the cortex into the surrounding soft tissue. The result is an extensive marrow disease centered in a bone segment with a surrounding soft tissues mass without an extensive cortical destruction, causing lack of significant radiological signs – **Fig. 1**.<sup>8,13</sup>

When signs are present, an area of radiolucency with ill-defined borders is the most common finding. Low mineral

content in patients with osteoporosis can reduce sensitivity of radiographic bone evaluation. Sometimes indirect signs may be present, such as displacement of teeth and loss of the mandibular canal wall, often due to the presence of malignancies.<sup>14</sup>

CT is sensitive in detecting the presence of a lesion and defining its extension. The most common finding is the bone cortex destruction with bone marrow replacement by the tumor, often extended to the adjacent soft tissues.<sup>14</sup> It is rare to see periosteal reaction, which is usually more suggestive for the presence of osteomyelitis or osteosarcoma.<sup>3</sup> However, all these signs are unspecific and, even if they suggest the presence of malignancies, tissue biopsy is necessary to clarify the real nature of the lesion.<sup>15</sup>

Even MRI findings are variable. Usually, lymphomas show intermediate signal intensity, similar to the adjacent muscles on T1-weighted images, higher signal intensity on T2-weighted images, and variable degrees of enhancement, usually homogeneous, after injection of contrast agent.<sup>3</sup>

MRI is useful to define the extension of the disease, the involvement of the adjacent structures, and the presence of perineural spread. However, all these findings are unspecific. DWI may be helpful for a more specific diagnosis, highly suggestive of lymphoma. In fact, low signal intensity on ADC map is suspicious for lymphoma, due to the typical higher cellularity.<sup>14</sup>

Fluorodeoxyglucose positron emission tomography/CT or total body CT is necessary for staging and evaluating the presence of disseminated disease after histological diagnosis of lymphoma, even if extranodal lymphomas are most frequently localized than disseminated.

In summary, in this case, the presence of acute pain in the right mandibular body without any other symptoms was initially suspicious for inflammatory disease. However, the absence of any significant radiographic finding and persistence of symptoms despite the anti-inflammatory therapy needed further evaluation with CT and MRI, which showed findings highly suspicious for malignancy. In particular, the epicenter of the lesion was the mandible and replacement of bone marrow suggested bone primitivity rather than a tumor rising from soft tissues. Osteosarcoma is the most frequent diagnosis among bone malignant lesions, but the possibility of other diagnoses should always be considered.

## Conclusion

This case report underlines that, even if rare, lymphoma should still be considered in the differential diagnosis of mandibular lesions presenting with signs and symptoms typical of inflammatory disease.

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### Conflicts of Interest

None declared.

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