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A case of systemic precursor T-cell lymphoblastic lymphoma presenting with single tooth mobility

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Abstract

Lymphoblastic lymphoma, seen primarily in children or young adults, is a type of non-Hodgkin lymphoma that originates from B or T lymphocyte precursors and rarely occurs in the oral cavity. A case of systemic precursor T-cell lymphoblastic lymphoma mimicking periodontitis of a lower second molar in a 20-year-old adult is currently presented. The case was initially misdiagnosed as periodontal disease and treated with tooth extraction by a dentist. Re-evaluation of the patient due to worsening of symptoms lead to cone beam computed tomography scanning that thoroughly revealed an extended osteolytic lesion of the right mandible. Afterward, a biopsy was performed, thus reaching the diagnosis of precursor T-cell lymphoblastic lymphoma. This report discusses differences in epidemiology of T-cell and B-cell lymphoblastic lymphomas, as well as their various intraoral manifestations that are mimicking a large family of oral pathology. It also focuses on conventional imaging findings that imply malignancy, which are often neglected during routine radiology interpretation.

Keywords

T-cell lymphoma, non-Hodgkin lymphoma, tooth mobility, panoramic radiograph interpretation

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Introduction

Although lymphomas constitute only 3.5% of all intraoral malignancies, they still are the second most common neoplasms of head and neck, second to squamous cell carcinoma.[1,2] In fact, almost half of all lymphomas occur in the head and neck region, involving mainly the tonsil and the parotid gland.[1]

Lymphoblastic lymphomas (LBLs) are an uncommon and aggressive type of non-Hodgkin lymphoma (NHL), associated with immature lymphocytes of B-cell (B-LBL) or T-cell lineage (T-LBL).[3] Traditionally, the term lymphoblastic lymphoma has been used to describe predominantly lymph-node based disease compared to acute lymphoblastic leukemia (ALL) that is associated with more bone marrow involvement (at least 20% lymphoblasts in marrow).[4] However, since 2008 World Health Organization (WHO) unified LBL and ALL under the term precursor B-cell and T-cell lymphoblastic leukemia/lymphoma based on new immunogenetic, morphological, and molecular characteristics.[4,5]

Precursor T-cell LBL is most common in teens or young adults compared to precursor B-cell LBL which is more common in children or young adults.[6] In general, LBLs predominantly affect males with an overall male-to-female incidence ratio of 2.5:1.[4,6] In 50%–65% of the cases, T-cell LBL presents with a mediastinal mass which may cause dyspnea and chest pain.[6] In comparison, 75% of precursor B-cell LBL cases develop lesions in extranodal organs including skin, breasts, liver, and bone, as well as the lymph nodes.[5,6] Apart from bone marrow, LBLs have also a predilection for central nervous system.[5] Precursor T-cell LBL has a poorer prognosis compared to precursor B-cell LBL, with an overall 5-year survival rate as low as 26%.[6]

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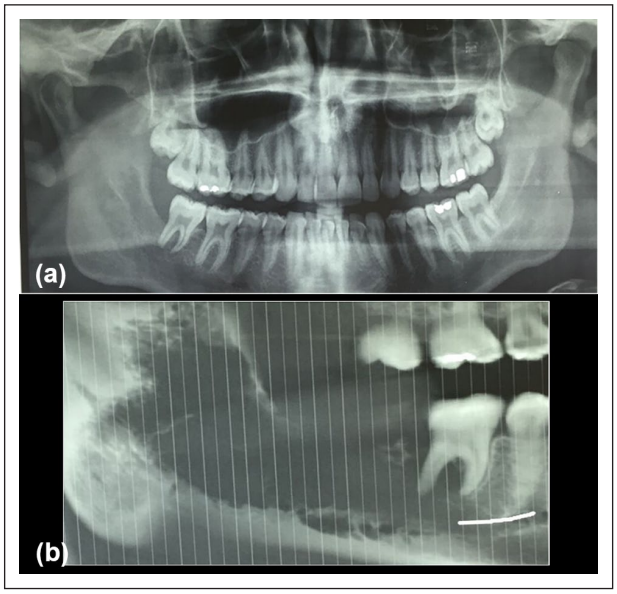

Figure 1. (a) Patient's panoramic radiograph: this radiograph was initially considered non-suggestive of bone destruction in the right mandibular body and ramus. However, a more careful observation would have revealed ill-defined radiolucency of the ascending ramus of the mandible (right side), loss of lamina dura of the second right molar (#47) and loss of the right mandibular canal wall, implying a possible malignancy. (b) CBCT revealed an osteolytic lesion of the right mandibular retromolar area, extending to the inferior alveolar canal and the tooth #46.

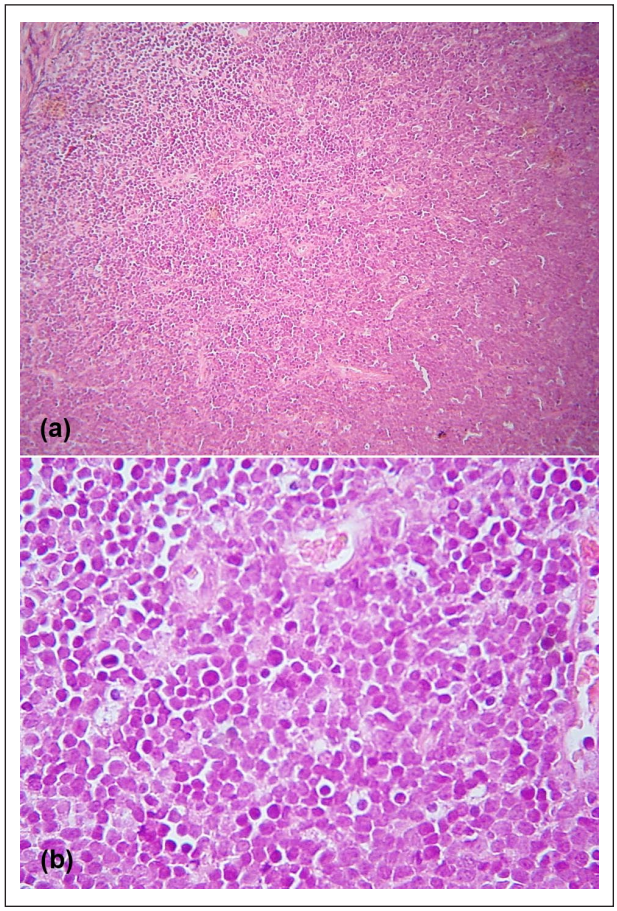

Figure 2. (a) Histopathological photomicrograph of the oral lesion illustrating lymphocytic infiltration of the sub mucosal connective tissue. H and E stained section 250×. (b) Higher power showing a monotonous population of numerous atypical lymphocytes with hyperchromatic irregular nuclei with inconspicuous nucleoli. Isolated mitotic figures of the tumor cells are also seen. H and E stained section 400×.

Since 1970, when the first case report on this topic was published, the most frequently reported lymphoma type in head and neck is diffuse large B-cell lymphoma, associated mainly with older males (in their seventh decade of life).[1,2,7,8] The most common site for NHL in the head and neck region is Waldeyer's ring.[8,9] Other sites are the orbit, nasal cavity, paranasal sinuses, oral cavity (palate, gingiva, and tongue), salivary glands, mandible, and thyroid.[5,8,9] There have also been reports of "double-hit" NHLs that arise in more than one intraoral site and are associated with poorer prognosis.[10]

In this article, we describe an interesting case of systemic precursor T-cell lymphoma that was initially presented with single tooth mobility and a panoramic X-ray that failed to reveal the extent of mandibular bone destruction.

Case report

A 20-year old male presented with chief complaint of mobile right lower second molar to his private dentist. The patient had no family or medical history of hematological malignancies, weight loss, or malaise.

Upon clinical examination, no swelling of soft tissues was noticed and no clinical signs of lymphadenopathy were apparent. Panoramic radiograph was taken, and the diagnosis of periodontal disease was made (Figure 1(a)). The tooth was subsequently extracted with careful curettage of the extraction socket, and post-extraction antibiotic treatment was prescribed.

During re-evaluation of the case after 2 weeks, swelling of soft tissues at the extraction site was apparent with unilateral cervical lymph nodes positive in palpation. The patient was immediately referred to an oral and maxillofacial surgeon and underwent cone beam computed tomography (CBCT) that revealed an extensive osteolytic lesion of the right mandibular retromolar region (Figure 1(b)). The right alveolar nerve was included in the lesion, although there were no signs of neurological defect.

Subsequently, a biopsy of the intraoral lesion was performed. Histopathological examination showed a diffuse proliferation of small-to-intermediate-sized lymphoid cells that infiltrated the sub-mucosal connective tissue stroma (Figure 2(a)). The tumor cells exhibited hyperchromatic nuclei, a high nuclear cytoplasmic ratio, and irregular nuclear contours, while mitotic figures were also seen (Figure 2(b)).

The histopathological findings were consistent with the diagnosis of malignant lymphoid neoplasm. On immunohistochemical analysis, tumor cells expressed the LCA, CD2, CD3, CD4, CD8, CD43, CD99, and terminal deoxynucleotidyl transferase (TdT). Tumor cells were negative for CD20, CD79a, CD30, CD15, CD56, ALK-1, TCL-1, CD1a, and TIA-1. The Ki67 proliferative index was approximately 70%. Based on histopathologic findings and immunohistochemical staining results, the final diagnosis of precursor T-cell lymphoblastic lymphoma was rendered.

As a result, the patient was immediately referred to the hospital. According to the chest X-ray, there was large left pleural effusion, and the trachea was deviated to the normal hemithorax. To this end, a chest tube was placed, and 3 L of pleural fluid was drained from the pleural cavity. Pleural effusion was exudative and a concentration of 5% of pathological cells was reported. The parameters of the full blood count were as follows: Ht = 40.9%, red blood cell (RBC) = $5.36 \times 10^3/\mu L$, white blood cell (WBC) = $6.37 \times 10^3/\mu L$ (neutrophils 69%, lymphocytes 29%, monocytes 2%), platelet (PLT) = $307 \times 10^3/\mu L$, and lactate dehydrogenase (LDH) = 419 IU/L. The patient was also tested HIV seronegative.

After performing neck, chest, upper, and lower abdomen computed tomography scans, a mediastinal lymph node enlargement was revealed. The largest lymph node was 9 cm in diameter. The mediastinal mass constituted another systemic manifestation of the disease, apart from the osteolytic lesion in the right mandibular retromolar region.

Bone marrow aspirate and trephine biopsy from right posterior superior iliac spine, as well as flow cytometry, were negative for bone marrow infiltration (if there was infiltration of the bone marrow with >20% leukemic cells, it would have been considered T-lymphoblastic leukemia). As such, this was characterized as a case of systemic stage IV T-cell lymphoblastic lymphoma (mediastinal lymph node involvement with extranodal osseous disease and no evidence of bone marrow infiltration).

The patient received chemotherapy treatment according to a German Multicenter ALL (GMALL) protocol and he also underwent intrathecal infusion of methotrexate. During the chemotherapy-induced leukopenia phase, he acquired a nosocomial bacterial infection that progressed to septic shock. He was intubated and died the next day.

Discussion

Oral manifestations of lymphomas account for only 3% of all lymphoma cases and 4% of lymphomas in patients with AIDS, mimicking a large family of oral pathology like periodontal disease (tooth mobility or displacement), pericoronitis, osteomyelitis, lock jaw, paresthesia, swelling, facial asymmetry, and various malignancies of the oral cavity.[7,11] Almost half (40.52%) of all lymphoma cases with oral manifestations are initially misdiagnosed and are treated with tooth extractions, or with periodontal and antibiotic treatments.[7]

The panoramic X-ray of the present case was initially considered non-suggestive for bone destruction. However, a more careful observation would have revealed the following findings: ill-defined radiolucency of the ascending ramus of the mandible (right side), loss of lamina dura of the second right molar (#47), and loss of the right mandibular canal wall. These findings are all suggestive of malignancy, but they may not be perceptible to the untrained eye. Other findings that have also been reported which may not be taken into consideration when interpreting a conventional image are widening of periodontal ligament, teeth displacement, and widening of the mandibular canal and mental foramen.[9]

Concerning conventional imaging, a recent report on panoramic X-ray findings among four cases of diffuse large B-cell lymphoma (NHL) involving the mandible concluded that an apparent radiolucent lesion was present in only 50% of the cases.[9] An ill-defined bone destruction may not be a common panoramic finding for NHL involving the mandible, especially at an early stage and prior to any tooth extraction that may accelerate bone destruction.[9] NHL involving the mandible tends to show slight or mild cortical bone destruction relative to the extent of the tumor involvement, thus making apparent radiolucency often imperceptible on panoramic images.[9]

T-LBL and T-cell ALL are considered to be the same disease with different clinical presentations.[4] T-LBL frequently presents with a large mediastinal mass, as in the present case. T-LBL is arbitrarily defined by the presence of enlarged lymph nodes and <20% lymphoblasts in the bone marrow, contrasting with >20% blasts in ALL.[4] Our patient had no evidence of bone marrow infiltration. Treatment with conventional regimens for aggressive lymphomas results in short disease-free survival and is not recommended.[12] Therefore, T-LBL is treated with intensified ALL-like regimens.[12] In the case under discussion, the GMALL 07/2003 protocol was initiated, a protocol of 12 month duration with alternating blocks of intensified chemotherapy, used for the treatment of young patients with ALL.[12] Unfortunately, patient did not survive due to complications.

Conclusion

Despite the rarity of this case, two learning points can be deducted. First, tooth mobility in a young person, without history of juvenile periodontitis and with excellent oral hygiene, should raise high suspicion of an underlying systemic disease, thus avoiding dental treatment in favor of obtaining a detailed medical record. Under this notion, hematological evaluation along with an HIV test (when indicated) seems to be a critical step in patient management and should not be avoided. Second, imaging has a key role for patient management. For example, it is obvious that an intraosseous lesion appearing with unilocular radiolucency with or without tooth resorption in a periapical or panoramic X-ray should raise high concerns for CBCT and biopsy. However, even in cases like the one presented here, where initial clinical examination

and radiological findings were not considered suggestive of an underlying pathology, a CBCT should have been performed to definitely exclude bone implication in tooth mobility. The clinician should always keep in mind that a more careful interpretation of conventional imaging may reveal findings that imply malignancy which are often neglected during routine radiology interpretation.

Declaration of conflicting interests

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Ethics approval

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Informed consent

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