AMERICAN JOURNAL OF DIAGNOSTIC IMAGING, 2023
VOL 9, NO. 2, PAGE 16–21
10.5455/ajdi.20221213075555

CASE REPORT

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Primary diffuse large B-cell lymphoma of maxillary sinus: Case report and literature

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ABSTRACT

Background: Diffuse large B-cell lymphoma (DLBCL) is the most common type of malignant lymphoma, accounting for 30%–40% of non-Hodgkin lymphoma (NHL) cases in adults. A specific type of NHL, primary paranasal sinus lymphoma, is a rare neoplasm of extranodal NHL and can often be difficult to diagnose. It generally has a poor prognosis because of the complex anatomy of the region and it can usually be noticed at an advanced stage.

Case Presentation: In this case report, we present DLBCL of the maxillary sinus in a patient who attended to our clinic with a complaint of swelling in the maxillary anterior region for 3 months. Before coming to our clinic, the patient was examined by several physicians who misdiagnosed her condition as a lack of additional imaging methods and incomplete examination, assuming dental origin of lesion. After radiological evaluations and histopathological examination the correct diagnosis was made and the patient was referred to the hematology clinic where chemotherapy was planned.

Conclusion: Despite the advancement of modern diagnostic methods, timely recognition of disease symptoms still forms the basis of early diagnosis. This case report demonstrates the need for systematic examination of anatomical structures during clinical and radiographic evaluation in terms of early diagnosis and to prevent delays in treatment.

ARTICLE HISTORY

Received 13 December 2022
Accepted 11 February 2023
Published 26 March 2023

KEYWORDS

Diffuse large B-cell lymphoma; maxillary sinus neoplasms; cone beam computed tomography; lymphoma; case report

Background

Non-Hodgkin lymphoma (NHL) is a neoplasm originating from lymphoreticular system cells [1]. Although it is highly variable, it usually tends to affect organs and tissues that do not contain lymphoid cells [2,3]. It is a heterogeneous group of diseases characterized by clinical features, cell of origin, molecular features, and frequently recurrent mutations and differentially variable outcomes [4–9]. Primary paranasal sinus lymphoma (PPSL) is a rare neoplasm of extranodal NHL that accounts for only 2% of all lymphomas [4,10]. It is most commonly seen in male patients aged 50–70 years [11]. Initially, tumors in this area cause few symptoms and are usually in an advanced stage at diagnosis. Ophthalmological signs and symptoms occur in the early stages of the disease due to the anatomical proximity of the orbit to the paranasal sinuses [12]. The most common symptoms for diffuse large B-cell lymphoma (DLBCL) originating from the maxillary sinus are unilateral, painful swelling of the maxilla, nasal congestion, pain, mucopurulent rhinorrhea, recurrent epistaxis, diplopia, and rapidly expanding masses. It usually presents with both local symptoms and systemic symptoms such as fever, recurrent night sweats, and weight loss [2,13–16]. This article is aimed at presenting a rare case of DLBCL of the maxillary sinus and sharing the clinical and radiographic findings of this tumor in light of the literature for dentists.

Case Presentation

A 68-year-old female patient was admitted to our clinic with complaints of swelling in the anterior

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maxillary region for 3 months and narrowing of the upper total prosthesis. From the patient's anamnesis, it was learned that she was non-smoker, that she was not using alcohol and that she had a history of hypertension and diabetes, which she kept under control with regular medical treatment. As she has no professional occupation, there were no exposure or co-exposure to chemicals and another risk factors related to occupation. Except for cardiovascular conditions there were no diseases in her family history, including the malignancy. Before noticing the swelling in the anterior maxilla, the patient attended to an ophthalmologist with complaints of left eye watering and numbness in the left anterior zygomatic region and was prescribed eye drops whose name she did not know. Furthermore, it was discovered that the patient had visited the family doctor several times in the previous 3 months with complaints of swelling and pain and had been prescribed mouthwash as a treatment. The patient applied to a dental clinic because her complaints did not improve, and from there she was referred to our clinic. In the extraoral examination of the patient, significant upper lip protrusion, subnasal swelling, and pain on palpation in the left maxillary sinus region were detected (Fig. 1).

During the intraoral examination of the patient, it was observed that she was completely edentulous. A highly vascularized, soft, and slightly painful swelling on palpation was detected in the maxillary anterior region, the vestibular sulcus, the inner surface of the upper lip and the anterior of the hard palate (Fig. 2).

Intraoral ultrasonography revealed a hypoechoic lesion of uniform shape with low vascularity causing resorption in the maxillary corpus starting from the maxillary midline and extending to the left premolar region. In the submandibular region, a reactive lymph node was observed (Figs. 3 and 4).

In cone beam computer tomography (CBCT) imaging taken for further examination, it was observed radiolusency in the anterior of the maxilla, in the bilateral incisor region and anterior to the hard palate. The radiolusency was associated with the nasal cavity and maxillary sinus, perforating the buccal cortex. Perforation was also observed in the floor and left lateral wall of the nasal cavity, anterior, medial, lateral, inferior, and superior walls of the left maxillary sinus, and the floor of the left orbit (Figs. 5–8).

Except for the high neutrophile concentration, the patients' blood results were within the normal range (Table 1).

Maxillary sinus malignancy was suspected based on anamnesis, clinical, and radiographic findings, and the patient was referred to the ear, nose, and throat clinic. An incisional biopsy was performed from the patient under general anesthesia (Table 2).

In the histopathological examination of the biopsy material taken after the fine needle aspiration biopsy the lesion was reported as a malignant tumor. Discohesive atypical cells with coarse chromatin, prominent basophilic nucleoli, and atypical mitoses were observed (Fig. 9a).



Figure 1. A prominent upper lip protrusion is seen in the photo taken laterally.

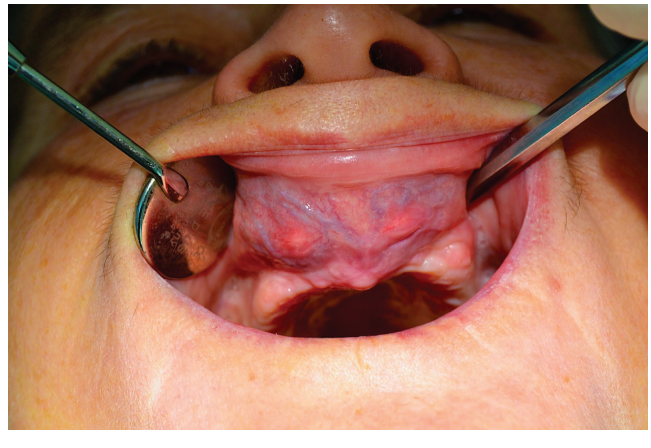


Figure 2. Intraoral tumoral lesion with hemorrhagic appearance, soft and slightly painful on palpation.

In the immunohistochemical examination applied to determine the nature of tumor cells; CD20, Mum-1, Bcl-2 were found to be widely positive (Fig. 9b–d). The Ki-67 index was above 80% (Fig. 9e). The case was reported as "Diffuse large B-cell Lymphoma – Non-germinal center cell." On positron emission tomography (PET) images, a hyperactive soft tissue mass filling the left maxillary sinus was observed, and another focus was observed with the related soft tissue mass filling the nasal cavity anteroinferior, which was continuous with the lesion involving the alveolar arch anteriorly and the nasal septum in the middle. No suspicious lesion or involvement of pathological activity was detected in the thorax, abdomen, pelvis, or skeletal system.

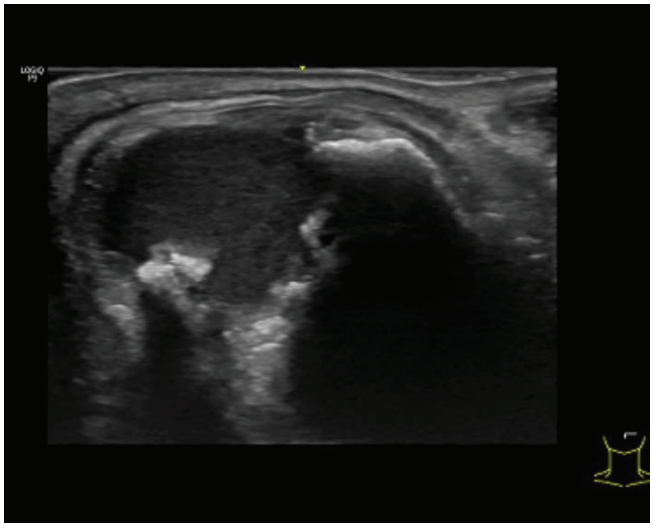


Figure 3. Intraoral image of the tumor obtained by placing the probe in the transverse plane from the anterior.

The patient was referred to the hematology clinic, and chemotherapy was planned. The patient's follow-up and treatment continue.

Discussion

NHL usually originates in lymphoid organs and tissues. However, it has been reported that up to 35% of cases originate primarily from extranodal regions. PPSL constitutes 2% of all primary extranodal lymphomas [17]. It has been reported that the maxillary sinus is most affected by NHL among the paranasal sinuses and DLBCL is the subtype with the highest histological incidence [18–20].

Sinonasal lymphomas can progress with nonspecific complaints and findings that do not make an early

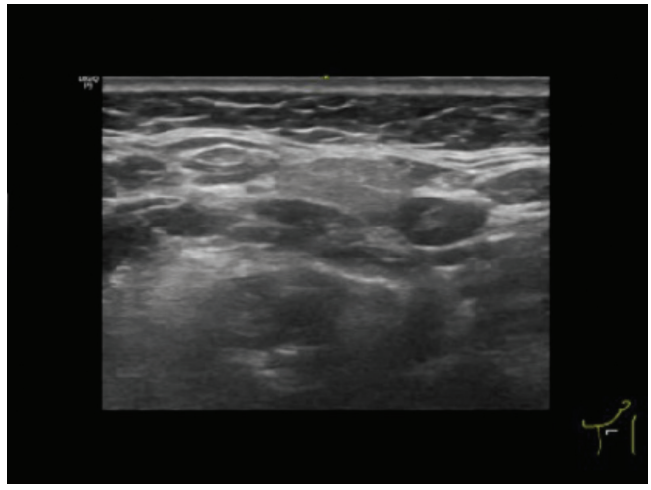


Figure 4. 1.16 × 0.44 cm left submandibular lymph node.

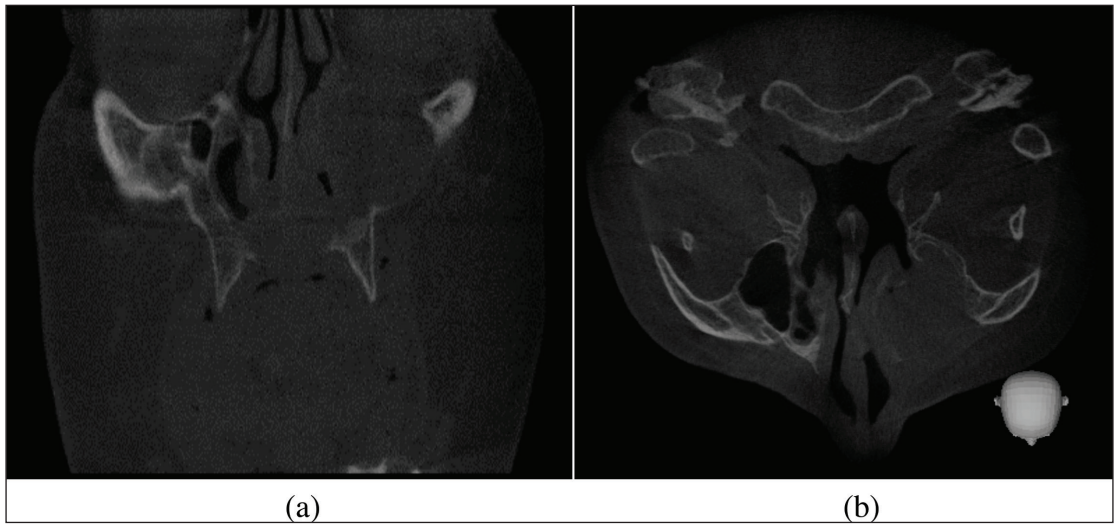


Figure 5. (a and b) Large perforation areas at the base of the orbit, the anterior hard palate of the maxilla, the base of the nasal cavity, the medial wall of the left maxillary sinus, and an expansive mass at the soft tissue density, respectively, in the coronal and axial section of the CBCT.

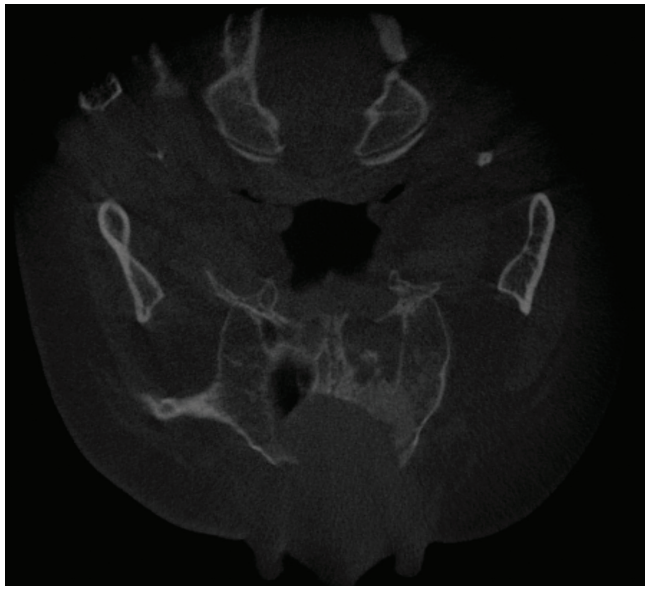


Figure 6. An irregular limited large mass containing a bilateral incisor and a canine region in the maxilla anterior to the axial section of the CBCT.

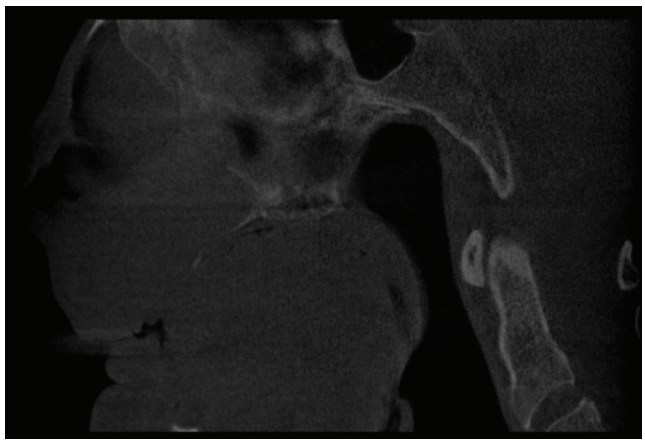


Figure 7. The mass that perforates the base of the maxillary sinus in sagittal cross-section of the CBCT.

diagnosis possible. Rhinosinusitis, pain, swelling of the face and mucous membranes, nasal congestion, fever, epistaxis and weight loss are the main findings of paranasal DLBCL [16,18–23]. Although DLBCL of the maxillary sinus is rare, its differential diagnosis with some aggressive inflammatory or infectious diseases is required [21,22].

Common findings in radiographic imaging of DLBCL include an isolated mass (59%), sinus opacification (53%), and/or bone erosion (35%) [14].

In the CBCT images taken, it was observed that there was destruction in the nasal cavity and maxillary sinus walls, anterior of the maxilla and hard palate.

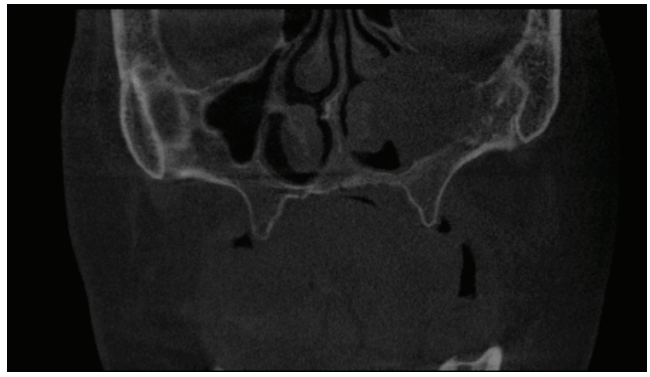


Figure 8. Large mass leading to perforation at the base of the left orbit. Explains the patient's orbital symptoms.

Table 1. Blood levels taken from the subject.

Tests applied	Result	Unit of	Reference
Hemoglobin	14.9	g/dl	12–16
Hematocrit	43.9	%	35–52
Leukocyte	9.57	Th/mm3	4.37–9.68
Neutrophile	74.6	%	43–65
Erythrocyte	5.23	million/mm3	4–6
Lymphocyte	18.7	%	20.5–45.5
Eosinophile	0.7	%	0.9–2.9
Monocyte	5.7	%	5.5–11.7
LDH	179	U/l	135–214
CRP	3.42	mg/l	<5

In line with these findings, the patient was referred to the ear nose and throat clinic with a pre-diagnosis of malignancy originating from the maxillary sinus. The preliminary diagnosis was confirmed by PET/CT and magnetic resonance imaging (MRI) examination while the final diagnosis was madehistopathologically by biopsy. Although there is a general consensus that lymph node involvement is common in oral lymphomas, it was determined that our patient did not have lymph node involvement [24].

Patients with sinonasal neoplasm may apply to the dentist with complaints about the oral cavity. In addition, the lesion can be detected incidentally on radiographs taken during a routine examination without any symptoms [25,26]. The diagnosis of the early stages of DLBCL is often delayed due to non-specific symptoms. The early stage of DLBCL in the maxillary sinus is frequently misdiagnosed as maxillary sinusitis or a dental abscess. Therefore, surgical biopsy is required for definitive diagnosis, together with the use of radiological examinations such as CT, MRI, and PET/CT [27].

Table 2. Patient's disease history.

1	**Patient (gender, age)**	68 years old, female
2	**Final diagnosis**	Primary DLBCL
3	**Symptoms**	Swelling in the anterior maxillary region, left eye watering and numbness in the left anterior zygomatic region
4	**Medications**	-
5	**Clinical procedure**	CBCT, ultrasonography, PET, biopsy
6	**Specialty**	Oral and maxillofacial radiology

Conclusion

This case report demonstrates the need for further investigation and radiological evaluations when patients presenting with unusual symptoms are encountered. In order to make an early diagnosis and avoid treatment delays, dentists must thoroughly examine anatomical structures other than teeth during clinical and radiographic evaluation. A multidisciplinary approach, including additional imaging methods and histopathological examination, is required when a neoplastic lesion is suspected by radiological examinations.

Conflict of interest

The authors declare that there is no conflict of interest regarding the publication of this article.

Financial disclosure

This study received no financial support.

Consent for publication

Informed consent to present this case was obtained from the patient.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

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