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Extranodal Non-Hodgkin's Lymphoma of the Oral Cavity Presenting as Gingival Swelling: A Case Report

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Abstract

Non-Hodgkin's lymphoma is a heterogeneous malignancy originating from proliferation of lymphoid cells or their precursors. , we report a case of a 6-year-old Iranian male child admitted at the Department of Oral Medicine, School of Dentistry, Mashhad, Iran, in March 2019. The case was complaining about gingival swellings worsening for one month. Clinical and pathologic examination, complemented by biopsy and immunohistochemistry, confirmed the diagnosis of high-grade B-cell non-Hodgkin's lymphoma. Treatment was based on chemotherapy. Intraoral involvement of lymphomas might be confused with inflammatory diseases; therefore the dentist has a crucial role in early diagnosis for appropriate management of the disease.

Key Words: Non-Hodgkin's lymphoma, Oral cavity, Gingival swelling.

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1- INTRODUCTION

Lymphoma is a heterogeneous malignancy which is resulted from proliferation of lymphoid cells or their precursors. In general, lymphoma has two major types: Hodgkin's lymphoma (HL) and non-Hodgkin's lymphoma (NHL) (1, 2). NHL is categorized into B-cell, T-cell and natural killer T (NKT) cell types. Almost 86 % of all lymphomas are NHL, with B-cell lymphomas being more common than T-cells types. Although the most NHLs are present in lymph nodes, 23–30% of patients with NHL show extra-nodal diseases (3). Extranodal NHL commonly involves skin, bones, gastrointestinal tract, Waldeyer's ring, spleen etc. Although relatively rare, lymphomas can occur inside the oral cavity in about 2% of cases, and can represent either the initial presentation or secondary involvement in the setting of a systemic disease (3, 4). Mostly, oral lesions appear as asymptomatic, soft-elastic lesions which might become ulcerated and hemorrhagic as a result of traumatic injuries. Bone involvement may also occur in some rare cases (1, 5). The aim of this report is to present a study of a case with extra-nodal, B-cell, non-Hodgkin's lymphoma arising on the gingiva as a secondary involvement of NHL disease, and to emphasize the difficulty of the diagnosis of gingival diseases as a manifestation of NHLs.

2- CASE REPORTS

A 6-year-old Iranian male child visited the Department of Oral Medicine, School of Dentistry, Mashhad, Iran, in March 2019 with the history of relatively rapid swelling of gingiva on both sides of the mandible in the previous month. His medical history further showed the swelling of the right testis without pain since 2 months ago. The patient also reported weight loss, nausea, vomiting and abdominal pain for the past two weeks. On extraoral examination, bilateral diffuse swellings were observed on both sides of the face and facial asymmetry was noticed due to the larger size of the swelling on the right side of the face. The right swelling was approximately 5×4×3 cm in dimension, extending superiorly from the zygomatic arch and inferiorly to the border of the mandible (**Figure.1**).

Fig.1: Photograph the patient showing the facial asymmetry.

The consistency of involved areas was firm, without any pain, with tenderness and localized rise in temperature. Regional lymph nodes were not palpable. Intraoral examination showed bilateral swellings of mandibular gingiva, extending from the deciduous canine to the retromolar pad on the both sides (**Figure.2**). The consistency of swellings on palpation was firm. A grade II mobility was detected in the associated teeth. The overlying surface had normal mucosa and tenderness was observed. Also, there were no symptoms of numbness. Based on the medical history and clinical examination (tooth mobility, rapid growth, bilateral swellings, testis involvement, and systemic symptoms) observed in the patient, a provisional diagnosis of non-Hodgkin's lymphoma of testis with the secondary involvement of gingiva was given. The patient was subjected to radiographic examination including panoramic (**Figure.3**).

Fig.2: Gingival appearance of the patient.

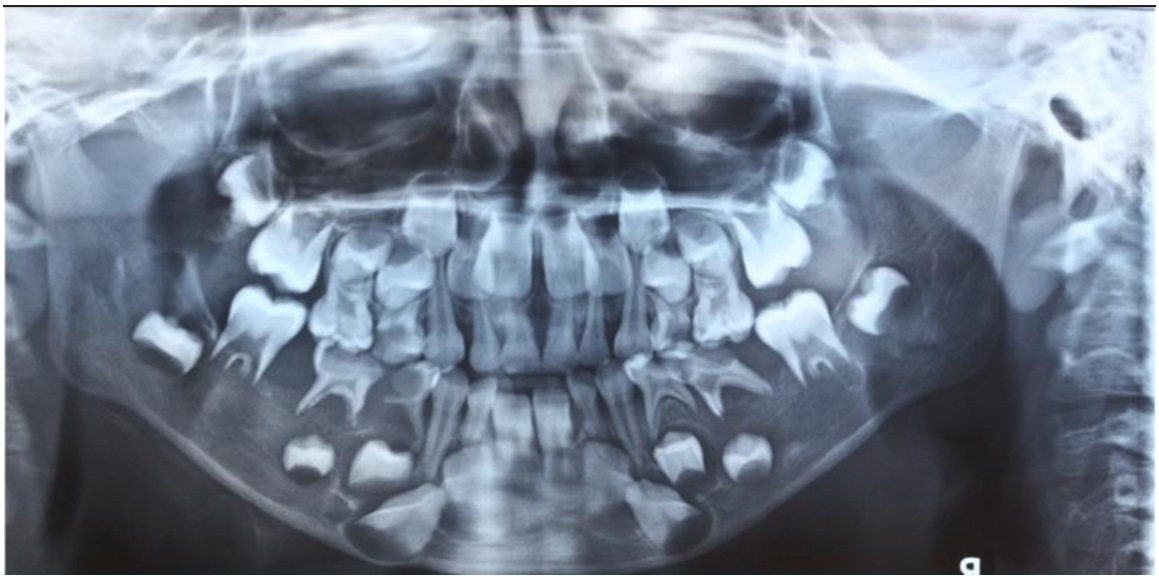

Fig.3: The radiograph shows horizontal alveolar bone loss of the mandible bilaterally.

The panoramic image showed horizontal alveolar bone loss of the mandible bilaterally. The patient was referred to the department of pediatric oncology for further assessment. Right testis biopsy was carried out. Histopathological examination of hematoxylin and eosin stained section showed testicular tissue structure composed of seminiferous tubules and intervening stroma which infiltrated with neoplastic small round cells. It was finally diagnosed as small round cell tumor suggestive of lymphoma.

On immunohistochemical analysis, tumor cells were positive for leukocyte common antigen (LCA), and cluster of differentiation 20 (CD20), but negative for CD3, CD99, terminal deoxynucleotidyl transferase (Tdt), methylation-inhibited binding protein 1 (MIB-1), Desmin and Synaptophysin. Hereby, the diagnosis of high-grade B-cell lymphoma was made. Ultrasound imaging showed a small lymph node of 8.8 x 3.6 mm dimensions in the right inguinal region, and a lymph node of 6.8x3.5 mm in the left inguinal area. Abdominal and pelvic ultrasound examination revealed a hypoechoic mass with a smooth, distinct dimension of 22.6x17 mm in the right iliac corresponding to a muscle and a similar but lobule-shaped mass of 21x13.6 mm dimension in the mid-trunk of the pancreas.

Routine blood investigations were carried out as well. White blood cell count, red blood cell count and platelet count were within the normal range. Combined Lymphome malin B (LMB) 96 /Advanced diffuse large cell lymphoma (DLCL) chemotherapy was started and the patient responded well to the treatment. Follow-up data shows that the patient has been in complete remission of B-cell lymphoma for approximately 1 year from the initial diagnosis and treatment. No evidence for local recurrence was observed.

3- DISCUSSION

Lymphomas are rare malignancies that affect the lymphoid tissues. Lymphomas are divided into two groups based on their morphology: Hodgkin's (HL), and non-Hodgkin's lymphomas (NHL). The risk of extra nodal involvement varies greatly between the two groups, from a minor 1% in Hodgkin's lymphoma to about 23-%30 in non-Hodgkin's Lymphoma. The most affected extra nodal sites in NHL are the gastrointestinal tract followed by the head and neck region (6). In the present case report, a 6-year-old male was referred after a 1-month rapid growth in the mandibular gingiva. In a study by Roh et al. 77.9 % of NHL patients were younger than 10 years old which is consistent with our case (7). Furthermore this case involved a male patient which contributes to the fact that pediatric lymphomas show predominance in males (8). The diagnosis of oral lymphomas is often difficult as clinical features of oral lymphoma such as local swelling, pain or discomfort might mimic an inflammatory and reactive process.

Only a limited number of reports indicate the locations of intraoral NHLs. Sirsath et al. reported seven patients with NHL on oral cavity. The sites involved were tongue, alveolus, gingivobuccal sulcus and hard palate (9). Kemp et al. studied forty patients with oral cavity involvement, with the most common locations being the upper jaw, mandible, palatal soft tissue and gingiva (10). Epstein et al. indicated NHL in 30 patients on the tongue, 17 on the palate, 7 on the gums and 3 on the lips (11). Bagan et al. reported 30 intraoral BNHLs, and the most frequent location was the gingiva (12). Donaduzzi et al. reported a case with NHL located on the buccal and lingual gingiva of the mandibular incisors (6). In a study by Urun et al. presented one case of NHL arising on the gingiva (13). Parihar et al. have reported NHL of gingiva in a 50-year-old female on the anterior mandible (14).

Basavaraj et al. and Patil et al. also indicated NHL of gingiva in HIV-positive patients (15, 16). NHL can be cured with different combinations of chemotherapy, radiotherapy and surgery. In the present case, the patient's chemotherapy regimen was LMB 96 /Advanced DLCL. The 5-year survival rate for children with NHL, after multi-agent chemotherapy, is about 70% (7). Bagan et al. and Roh et al. reported a 5-year survival rate of 81%, and 78.3% for patients with B-cell lymphomas, respectively (7, 12). In general, intraoral involvement of lymphomas remain nonspecific and can be mistaken for many different lesions of reactive or inflammatory origins. Therefore, it is of the utmost importance not to misdiagnose malignant lesions.

4- CONCLUSION

In conclusion, oral lymphomas presenting as gingival swelling mimic common oral and dental pathologic conditions and can be misdiagnosed as other lesions of reactive or inflammatory origins. This can delay the correct diagnosis of the oral lymphoma. It is important to avoid misdiagnosis of such malignant lesions. Early diagnosis of oral lymphoma is one of the responsibilities of dentists to improve the prognosis of the disease and decide on the appropriate treatment.

5- ETHICAL CONSIDERATIONS

The study protocol was approved by the Ethics Committee of Mashhad University of Medical Sciences, Iran.

6- AUTHORS' CONTRIBUTION

All authors contributed to the writing of the manuscript.

7- CONFLICT OF INTEREST: None.

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