CASE REPORT

A Report of a Rare Case of Anaplastic Large Cell Lymphoma of the Oral Cavity

Prasad G Kango^{1*}, Pushpak U Shah² and Revati S Deshmukh³

¹Department of Oral Pathology and Microbiology, Al- Ameen Dental College and Hospital, Bijapur-586101 Karnataka, India, ²Department of Oral Pathology and Microbiology, KLEVK Institute of Dental Sciences and Hospital, Belgaum-590010 Karnataka, India and ³Department of Oral Pathology and Microbiology, Bharati Vidyapeeth Dental College and Hospital, Pune, Maharashtra, India

Abstract: *Case history:* Malignant lymphoma is a neoplastic proliferative process of the lymphopoietic portion of the reticuloendothelial system that involves cells of either the lymphocytic or histiocytic series in varying degrees of differentiation and occurs in an essentially homogenous population of a single cell type. The character of histologic involvement is either diffuse (uniform) or nodular and the distribution of involvement may be regional or systemic (generalized), the process basically being multicentric. A case of oral cavity lymphoma was detected and after various panel of imunnohistochemical (IHC) markers it was diagnosed as Anaplastic large cell lymphomas (ALCL) of oral cavity. *Conclusion:* Lymphoma of oral regions are very rare and ALCL is rarest. It is of utmost importance to do the IHC, so that the prognosis of the lesion is known at the earliest. In this case since it was detected at a later stage the outcome was fatal.

Introduction

ALCL are very rare and the site of the lesion can be from auxiliary lymphnodes to any organ involvement. Described a case report of ALCL in mandible, which turned out to be fatal.

Case History

A 32 year old female patient visited a local dentist with a swelling on the lower right posterior mandibular molar region. After taking antibiotics and analgesics for few days the patient reported to the same dentist with an increase in swelling and was tender on palpation. The patient was then referred to the Department of Oral Pathology and Microbiology, Bharati Vidyapeeth University Dental College and Hospital, Pune. Patient gave a history of extraction of lower right first molar. All the vital signs were within normal limits except the blood pressure which was slightly elevated. The skin over the swelling was stretched, reddened and temperature over the swelling was elevated. Intra oral examination revealed an exophytic mass, round, reddish pink in colour and was measuring approximately 3x4x2 cms. The swelling was firm and bony hard in consistency. An ulceration was seen at one area which showed bleeding on probing and pus discharge. Right submandibular lymph nodes were palpable, fixed, non – tender and matted. Radiographic investigations showed an ill-defined radiolucency in right posterior border of the mandible.

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Pathologic Findings: Microscopically, the stained sections showed H&E no epithelium, with only a massive proliferation of lymphoid tumor cells (Fig.2). The tumor cells showed uniform proliferation with large sized atypical cells with cellular and nuclear pleomorphism. These cells showed multiple prominent nucleoli and dispersed chromatin (Fig.2).

Figure-3: Photomicrograph showing positivity of tumor cells for CD45, CD45RO, CD30 and EMA. (Immunohistochemistry)



Computerized tomography (CT) revealed osteolytic an lesion perforating the buccal cortical plate (Fig 1). Under a clinical diagnosis of malignancy of the mandible an incisional biopsy on the lesion was performed. The incised tissue was fixed in 10% neutral buffered formalin and embedded in paraffin, then cut into 4-um sections, and subjected to hematoxylin and eosin (H&E) staining.

Figure-2: Photomicrograph showing cells with abnormal mitosis (H & E original magnification X1000).



The tumor cells showed frequent atypical mitosis. A histologic differential diagnosis of Large 'B' or 'T' cell Lymphoma was made. confirm the То diagnosis immunohistochemical staining was carried out on formalin-fixed paraffin embedded tissues. Panel of antibodies used were CD 45, CD 79a, CD 45RO, CD 15, CD 30, EMA. The result showed that the tumor cells were positive for CD45, CD 45RO, CD 30 and EMA. Longo G et al did a study which included 36 patients of ALCL to

know the clinical characteristics, treatment outcome and survival of these patients. All their cases were CD30+, which was similar to our case [1].

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Figure-4: Photomicrograph showing negativity of tumor cells for CD79a, CD15 (Immunohistochemistry).



The tumor cells were negative for CD79a and CD15. ALCL of the oral cavity was the final diagnosis as it showed positive immunohistochemical staining for CD45, CD45RO, CD30 EMA (fig 3). The tumor cells were negative for CD79a and CD15 (Fig 4).

Discussion

ALCL was first described by Stein et al. in 1985. The disease is characterized by proliferation of anaplastic large lymphoid cells with abundant cytoplasm which strongly express CD30 antigen. ALCL is subdivided into 3 categories depending upon the clinical criteria and immunohistochemistry:

- 1) Primary systemic anaplastic lymphoma kinase (ALK+) ALCL;
- 2) Primary systemic ALK(+) ALCL; and
- 3) Primary cutaneous ALCL. [2]

Histologically, ALCL is characterized by neoplastic cells that have abundant basophilic cytoplasm and have a grey-blue appearance in H & E stained sections. The nucleus is often lobulated (horseshoe or kidney shaped). The neoplastic cell also has a prominent Golgi zone. The nuclear features of these neoplastic cells may vary from lobulated to round to oval shape. The nuclei may be placed in the centre or at one end with a paranuclear halo [3].

The neoplastic cells with these cytologic features are called as "hallmark" cells and are a consistent finding in ALCL [4-5].

Other histopathologic variants of ALCL

1.	Small cell variant	3.	Hodgkin's-like ALCl
2.	Lymphohistiocytic variant	4.	Sarcomatoid variant

The histologic appearance of ALCL can sometimes be confused with that of a poorly differentiated carcinoma, amelanotic melanoma or Hodgkin's lymphoma. Thus an immunohistochemical examination is helpful in distinguishing it from those other disease entities. Here it has to be noted that ALCL must be distinguished from malignant melanoma (MM) as this one also shows CD30 positive cells, but MM shows positivity for HMB45 and S-100 protein, whereas ALCL does not have such reactivity [1].

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The first report in the literature of an ALCL involving the oral cavity was in 1991, in an HIV-positive patient. Three cases were later reported in 1993. Again in 1996 a 12 year old boy was affected with ALCL, the lesion extending from hard palate to nasal cartilage and floor of the orbit. Recently ALCL of the lip was reported and successfully treated with combination therapy [4].

In our case the patient had a lesion on the gingiva which is a common site for ALCL in the oral cavity, as 44.4% of the cases studied by Matsumoto et al [2], occurred on the gingival. Other sites involving the oral cavity were lip (22.2%), palate (11.1%) and tongue (11.1%). In this case the patient's initial complaint was that of pain and swelling which was in concordance with the finding of Matsumoto et al. [2] Submandibular lymph nodes were palpable, fixed, non-tender and matted. Because of the initial signs and symptoms it was difficult to distinguish from other inflammatory lesions. Only histologic and immunohistochemical analysis made it possible to give the correct diagnosis. In order to categorize the cell size, the neoplastic cells can be compared with the reactive histiocytes interspersed among the lymphoma cells. Small, medium-sized and large cells refer to cells with nuclei smaller than, approximately the same size as, and larger than those of the reactive histiocytes respectively.

If histiocytes are not found, the endothelial cells nuclei can be used instead as the "ruler" although they are certainly less satisfactory because of their ovoid or elongated shape.

The general leukocyte marker is CD45RO

- → Stains → cell membrane
- Cells all leucocytes except plasma cells {which are negative or variably positive}
- → For diagnosis of all lymphomas and leukemias
- → Negative for R.S cells in Classical Hodgkin's Lymphoma.

Malignant oral lymphomas are rare, contributory 2-5% of all oral malignancies.

Conclusion

In conclusion this review is consistent with the previous reports of ALCL with reference to clinical presentation, histopathology and immunohistochemistry of ALCL.

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*All correspondences to: Dr. Prasad G. Kango, Assistant Professor, Department of Oral Pathology and Microbiology, Al-Ameen Dental College and Hospital, Bijapur-586101 Karnataka, India E-mail: kangopg@yahoo.co.in