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CASE REPORT

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LYMPHOEPITHELIOMA-LIKE CARCINOMA OF THE ORAL CAVITY - A DIAGNOSTIC PERPLEXION

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ABSTRACT

Lymphoepithelioma-like carcinoma (LELC), a rare tumor in oral cavity is characterized by non-keratinizing, undifferentiated squamous cell carcinoma with lymphocytic infiltration bearing microscopic resemblance to (nasopharyngeal carcinoma). A 45 year old female presented clinically with an unusual maxillary swelling and pain in right upper posterior jaw with intermittent serous like discharge from the nose. A diagnosis of rare case of intraoral LELC was made.

Keywords: Immunohistochemistry, Lymphoepithelioma-like carcinoma, Maxillary swelling, Nasopharyngeal carcinoma, Undifferentiated Squamous Cell Carcinoma

INTRODUCTION

Lymphoepithelioma-like carcinoma (LELC), a rare neoplasm in the oral cavity histologically shows non-keratinizing, undifferentiated squamous cell carcinoma with lymphocytic infiltration. LELC, a medical term refers to a histological type of malignant tumor which arises from the uncontrolled mitosis of transformed cells of epithelial tissue origin, that resemble lymphoepithelioma (nasopharyngeal carcinoma) microscopically.[1] The past data reveals a similar biological behavior to that of nasopharyngeal lymphoepithelioma. Identical neoplasm has been described in the skin, stomach, salivary glands, larynx, thymus and uterine cervix, both histologically and immunophenotypically.[2] Histologically, LELC reveals neoplastic epithelial component in association with a reactive lymphoid infiltrate. Tumor cells are present in the form of nodules, trabeculae, narrow cords isolated or anastomosing islands, round to oval nests. Atypical polygonal cells with vesicular nuclei and prominent nucleoli represent the epithelial component of the tumor. Therefore, the histologic diagnosis of LELC may be complicated due to the variable architectural and cytological appearance of the epithelial cells and the dense lymphoplasmacytic infiltrate which might obscure the epithelial component. Immunohistochemical evaluation is helpful and serves to exclude other histologically similar lesions. Patients with suspected LELCs should have a thorough otolaryngologic examination, due to the close histologic similarity to nasopharyngeal lymphoepithelioma, including indirect laryngoscopy to rule out a metastatic nasopharyngeal lymphoepithelioma.1 Our case report sheds light on a rare case

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of intraoral LELC occurring at an unusual site and is amongst the very few reported in the Indian sub-continent.

CASE REPORT

A 45 yr old female reported in the department with complaint of pain and swelling in the upper right posterior region of jaw and intermittent serous like discharge from the nose since past four months. The swelling gradually increased in size to attain the present size. Extra oral examination revealed single diffuse oval swelling on the upper right side of face; (Figure 1A). Intraorally, on palpation the swelling was diffuse, tender and hard in consistency with indistinct margins that merged with the surrounding structures. Teeth associated were vital and the overlying mucosa and gingiva were normal. Cervical lymph nodes were non-palpable. An OPG revealed large well defined unilocular lesion on the right maxilla (Figure 1B). Based on the intraoral and radiographic findings , a provisional diagnosis of oral squamous cell carcinoma, and differential diagnosis of fibro-osseous lesion and unicystic ameloblastoma was made. The patient was taken up for surgery under local anaesthesia and complete surgical excision of the lesion was done. The excised mass was then sent for histopathological examination. The gross specimen were sent in the form of multiple pieces which were firm in consistency with irregular margins and creamish white to brown in color. (Figure 1C). Microscopically, the sections revealed bony cortex with adjacent tissue showing a tumor disposed in sheets closely intermingled with dense inflammatory cells. No keratinization was seen in the tumor. A diffuse pattern of round to polygonal cells displaying smooth outlined vesicular nuclei was present. A mixed inflammatory infiltrate of plasma cells, lymphocytes, polymorphonuclear cells and eosinophils with prominent nucleoli were present. Cells undergoing abnormal mitosis was also noted. Further immunohistochemical examination revealed strong immunoreactivity for PAN cytokeratins and epithelial membrane antigen (EMA) (Figure 2A,B,C,D). Morphological and immunohistochemical

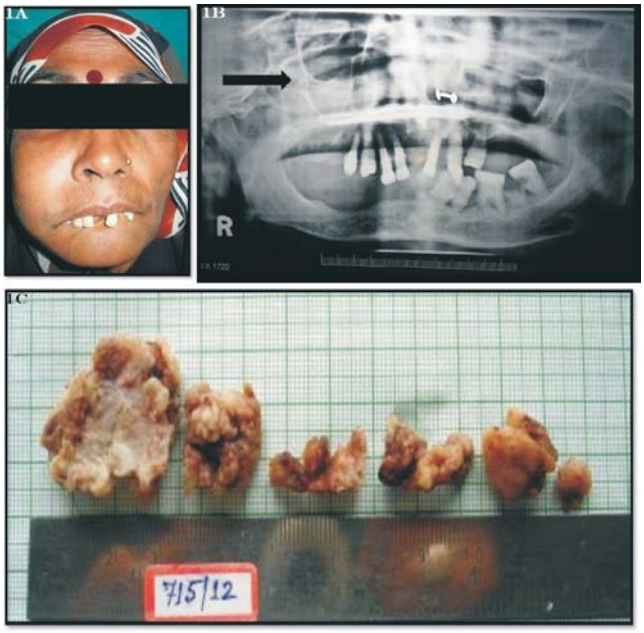

Figure 1A: Single diffuse swelling on upper right side of face, 1B: Large well defined unilocular radioluscency on right side of maxilla, 1 C: Gross- firm tissue with irregular margins.

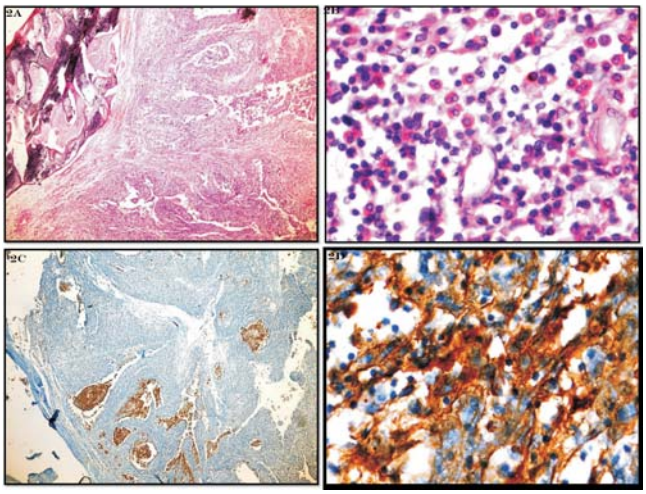

Figure 2A: Microscopically - bony cortex with adjacent tissue showing a tumor disposed in sheets closely intermingled with dense inflammatory cells. (H/E, 10X). 2B: Diffuse pattern composed of round to polygonal cells displaying smooth outlined vesicular nuclei and inflammatory cells comprised of lymphocytes, plasma cells, polymorphonuclear cells and eosinophils with prominent nucleoli (HE, 40 X). 2C: Strong immunoreactivity for PAN cytokeratins and epithelial membrane antigen (EMA) (20 X). 2D: Strong immunoreactivity for PAN cytokeratins and epithelial membrane antigen (EMA) (40 X).

findings fulfilled the criteria for the diagnosis of lymphoepithelioma-like carcinoma (LELC). The patient has been under regular follow up for the past 2 years is currently disease free.

DISCUSSION

An undifferentiated nasopharyngeal carcinoma with lymphoid stroma and non-keratinizing squamous cells with distinctive etiological, clinical and epidemiological features is called by various names like Lymphoepithelial carcinoma/ lymphoepithelioma/ nasopharyngeal carcinoma type III and Schmincke –Regaud tumor.[3]

LELC is a carcinoma which arises outside of the nasopharynx, resembling lymphoepithelioma both immunophenotypically and histologically. It is a malignant epithelial tumor which reveals large uniform cells with indistinct cytoplasmic borders, leading to a syncytium, round to vesicular and ovoid nuclei, with large central nucleoli.[4] Most of the times, an inflammatory infiltrate rich in mature lymphocytes (predominantly CD8+ and T-cells) and occasionally eosinophils are also present that partially conceal the neoplastic epithelial component.[5]

Immunophenotypically and histologically identical tumors which arise outside the nasopharynx are designated LELCs and they have been described in various body parts like gastrointestinal tract, lung, salivary glands, thymus, the skin, larynx, thyroid, uterine cervix, prostate, and urinary tract. Indeed, LELCs are characterized by either cohesive nests or by a diffuse growth of malignant epithelial cells in a background of inflammatory cells that are predominantly lymphocytes.[2,6]

The cases of LELCs have traditionally been classified as 'high grade' due to their poor histological differentiation. The main line of treatment is surgical resection. Despite their high histological grade and poor differentiation, LELCs are very radiosensitive and show better prognosis than the other high-grade salivary gland tumors such that radiotherapy has been used instead of surgery as the primary treatment of LELC in some cases.[7]

Prognosis of LELCs depends on various factors like stage and grade of the cancer, the completeness of surgical resection, incomplete resection and postoperative radiotherapy.[8-10] More so, a clear distinction between non-keratinizing carcinoma and large-cell lymphoma could be difficult. Immunohistochemical stains that use epithelial markers and common leukocyte antigens help in its accurate diagnosis. In the present report, features which favored the diagnosis of carcinoma include the presence of cohesive cell groups having poorly defined cell borders and an immunohistochemical profile of the neoplastic cells which showed strong positivity for cytokeratin and EMA.

Our case of LELC was finally diagnosed after immunohistochemistry & correlating it morphologically. Keeping in mind the unilocular radioluscency in the OPG, squamous cell carcinoma(SCC) was thought to be the most probable diagnosis followed by fibroosseous lesion or unicystic ameloblastoma. The latter two were ruled out after the histopathological findings and IHC further ruled out SCC.

In conclusion, this report described an unusual and rare case of LELC of the oral cavity. Difficult cases pose a challenge and usually bring to light interesting results. Patients with LELC must be kept under a regular follow-up to rule out any metastasis thereby leading to a good prognosis for the patient.

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