Pseudoepitheliomatous Hyperplasia of the Head and Neck and Its Diagnostic Challenge

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Abstract

Pseudoepitheliomatous Hyperplasia (PEH) is a reactive epithelial proliferation seen in response to wide variety of conditions including infections, neoplasia, inflammation and trauma. It is characterized by hyperplasia of epidermis and adnexal epithelium and it closely mimics Squamous Cell Carcinoma (SCC). In this article we mentioned the diagnostic challenges for reaching the diagnosis of PEH with several biopsies and radiological examination.

Keywords: Pseudoepitheliomatous Hyperplasia (PEH) • Pseudocarcinomatous hyperplasia • Squamous Cell Carcinoma (SCC)

Introduction

Pseudoepitheliomatous Hyperplasia (PEH) is a reactive epithelial proliferation seen in response to wide variety of conditions including infections, neoplasia, inflammation and trauma. It is characterized by hyperplasia of epidermis and adnexal epithelium and it closely mimics Squamous Cell Carcinoma (SCC) [1]. This aspect of the condition poses a clinical and histopathological dilemma in the final diagnosis which has implications in treatment and prognosis. The pathophysiology of PEH is unclear and is hypothesized to be physiological response to several forms of skin damage. It is thought to act as defensive mechanism for transepithelial elimination of foreign body material [2].

Case Presentation

A 36 year old male with no past medical history presented with a 6 month history of progressive left side cheek swelling associated with pain, trismus and pus discharge. He used to chew tobacco for several years and stopped the habit 2 months before the presentation. No other constitutional symptoms or a family history of malignancy was reported.

Clinically there was a 8 cm × 5 cm indurated, convoluted and fluctuant area on the left cheek. In the center of the mass a sinus discharging pus was noted. Patient had trismus with 1 cm mouth opening. Intraorally an exophytic lesion in the buccal mucosa extending from the commissure to retromolar area with surrounding leukoplakic changes was visible (Figure 1).

Overall gross features were consistent with oral submucous fibrosis, with a verruco-proliferative growth suspicious of malignancy. Neck exam revealed palpable lymph node in the left submental triangle.

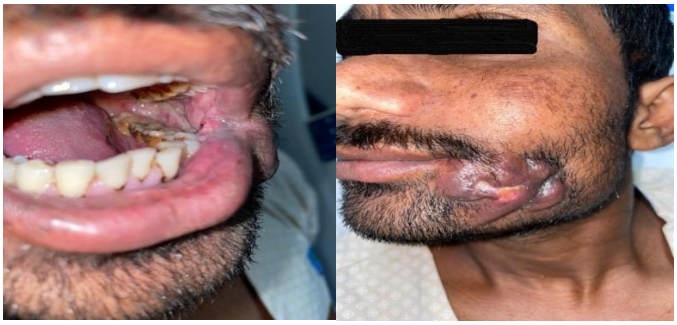
CT face and neck showed left buccal ulcerated mass lesion measuring approximately 4.8 cm × 2.4 cm, seen extending inferiorly along the left gingivobuccal sulcus. The lesion showed irregular outline with significant heterogenous enhancement and involvement of the opening of the parotid duct with dilatation of almost the entire duct was noted. The underlying bones appeared grossly normal with no evidence of cortical erosions. Multiple enhancing lymph nodes were seen in the left submental and submandibular regions (Figures 2 and 3).

MRI face and neck confirmed the CT findings (Figures 4 and 5). CT thorax and abdomen were unremarkable. Initial biopsies of left buccal mucosa showed lichenoid inflammation with scar formation and focal ulceration. The possibility of lichen planus was considered histologically, but the overall features fell short of a definite diagnosis. There was no dysplasia or malignancy.

However since the histopathological findings did not correlate with clinical findings, it was decided to perform further deeper biopsies after examination under general anesthesia. Pathological examination of the deeper tissue samples showed pseudoepitheliomatous hyperplasia with widespread background of mixed inflammation including multinucleate giant cells and focal ulceration (Figures 6 and 7).

Excision of enlarged submental lymph node was additionally performed and revealed reactive follicular hyperplasia, with no evidence of malignancy.

Patient was thereafter planned for excision of the lesion and reconstruction with a locoregional flap. However the patient refused further treatment.



A B

Figure 1. Clinical pictures showing the lesion (A) intraorally and (B) extraorally.

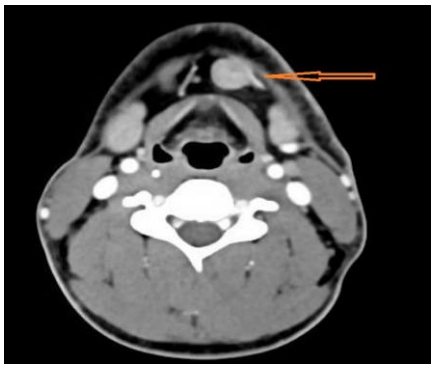


Figure 2. CT neck with contrast demonstrating enlarged left submental lymph node

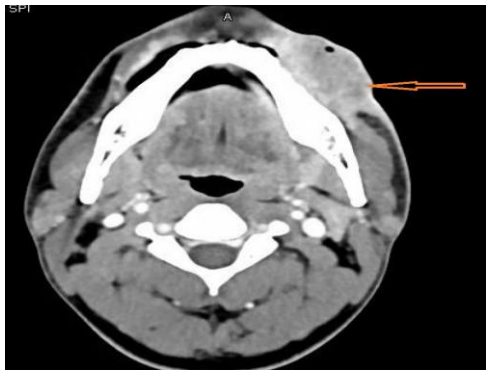


Figure 3. CT face with contrast showing Left buccal ulcerated mass lesion measuring approximately 4.8 cm x 2.4 cm is seen extending inferiorly along the left gingivobuccal sulcus. Involvement of the insertion of the parotid duct with dilatation of almost the entire duct. The underlying bones appear grossly normal with no evidence of cortical erosions.

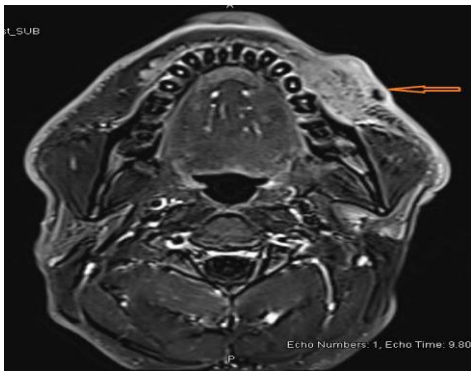


Figure 4. T1 MRI is showing left cheek ill-defined approximately 5 cm lesion. Significant contrast enhancement and enlarged vessels within the lesion noted. with no bony involvement.

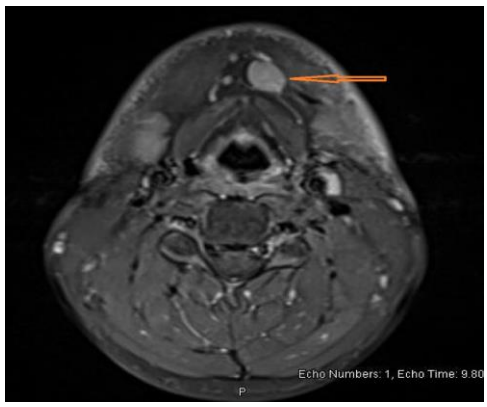


Figure 5. Post contrast T1 MRI showing the same enlarged submental lymph node.

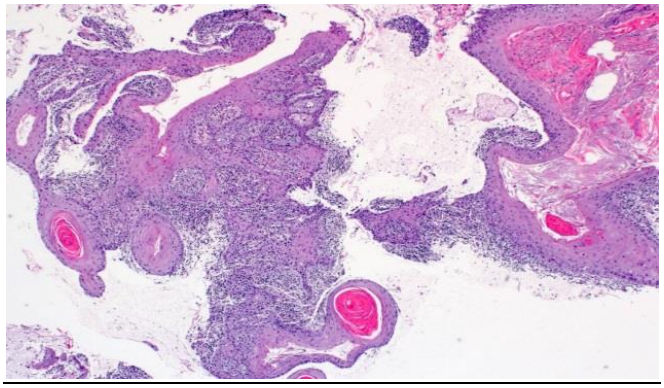


Figure 6. Low power view showing fragments of squamous mucosa exhibiting pseudoepitheliomatous hyperplasia, with dense background inflammation (H and E×2)

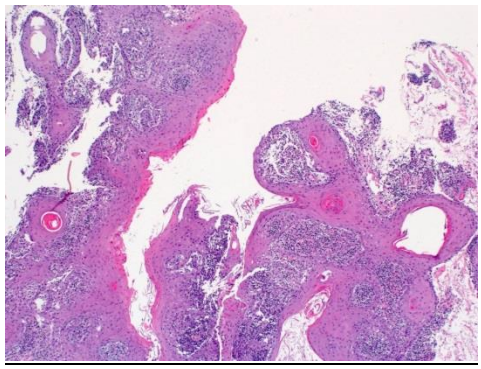


Figure 7. Higher power view of the lesion. Note the marked background inflammation and absence of cytological atypia (H and E ×10)

Discussion

PEH is a benign condition characterized by reactive irregular hyperplasia of the surface epithelium. PEH may be primary (e.g., primary gingival PEH) or secondary (e.g., granular cell tumor or chronic irritation) [3]. It can be a result of various conditions such as infections, inflammation, trauma, and malignancy and is sometimes also referred to as pseudocarcinomatous hyperplasia. Incidence of PEH in head and neck is unknown and current knowledge stems from sporadic case reports. It is fair to assume that they are very rare. Though the exact pathogenesis of this condition is unknown, most of them are believed to be due to the effect of the cytokines released from the inflammatory process or from an underlying tumor cell mass.

Frequently associated findings which favour or assist in the development of PEH are chronic persistent inflammation in the adjacent areas, chronic non-healing wound, infection (mycobacterial, fungal and parasitic), malignancy and retained foreign bodies. Oral mucocutaneous PEH has been reported to occur in association with various conditions like nontuberculous atypical mycobacterial infection, tuberculosis, actinomycosis, fungal and viral infections, granular cell tumor, pleomorphic adenoma, intraoral keratoacanthoma, malignant melanoma, oral submucous fibrosis, and epulis fissuratum [4].

These entities may mimic or give rise to PEH. The differentials are relatively easier to diagnose histologically. The challenge remains to avoid misdiagnosing PEH as SCC, as the implications for such a diagnosis can be life changing. A case of intraosseous mandibular PEH in post surgical and adjuvant CTRT setting has been reported [5].

Both clinical and histological appearances can be alarming and are fraught with pitfalls for the diagnostician. It is seen as tongue like epithelial proliferation invading the connective tissue and should not be mistaken for squamous cell carcinoma. In contrast to squamous cell carcinoma, these reactive lesions do not exhibit atypical mitotic figures, atypical nuclei, individual dyskeratotic keratinocyte; and show no evidence of vascular, lymphatic or perineural invasion.

Difference between squamous cell carcinoma and PEH

Histologically, it can be quite difficult to distinguish PEH from SCC. Some studies have reported increased staining for p53 and MMP-1 and less intense staining for E-cadherin in SCCs, as compared to PEHs [6]. But is important to note that the key universal histological criterias for SCC such as nuclear enlargement, nuclear hyperchromasia, irregular nuclear outline, coarse nuclear chromatin, and prominent nucleoli, are not observed or only focally present in PEH [7,8].

Other findings, which can help differentiate squamous cell carcinoma from PEH were studied. Langerhans cells in squamous cell carcinoma were found in a very low density compared to that of PEH. This finding was correlated with decreased expression of E-Cadherin in squamous cell carcinoma [9]. Also expression of p53 is increased in case of squamous cell carcinoma compared to that of PEH [10], and the expression of p53 is mostly restricted to the basal layer in case of PEH, which is in contrast to the squamous cell carcinoma, where it involves more superficial dysplastic cells [11].

A review of literature has left some crucial gaps in our understanding of this condition such as the natural history of the pathology, whether it is self limiting, potential for transformation into malignancy, predisposing factors and other non surgical modalities of management.

Complete excision with appropriate reconstruction is the management of choice for this condition cited in literature [12], which though a benign process poses significant quality of life issues due to disfiguring growth.

Conclusion

As PEH clinically and pathologically closely mimics SCC, it is crucial to rule out the later as the treatment is completely different. In this case multiple biopsies and lymph node biopsy ruled out malignancy, therefore the appropriate treatment is complete excision with reconstruction.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of Interest

The authors declare not to have any conflict of interest.

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