Metastatic tumour of the oral cavity – Report of a case

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Abstract: The oral region is an uncommon site for metastatic tumour cell colonization and is usually evidence of a widespread disease. Based on the numerous isolated case reports and large case series published by van der Waal RIF et al (2003) & Hirshberg A et al (2008), oral metastasis was found to be the first sign of the metastatic spread in 25 % of cases and was the first indication of an undiscovered malignancy at a distant site in 23 % cases. Because of its rarity & deceiving clinical presentation, the diagnosis of a metastatic lesion in the oral region is challenging, both to the clinician and to the pathologist. Recognition of such lesions & determination of primary site is important for early treatment instillation. This article aims to present a case of oral metastases as afirst indication of undiscovered malignancy at a distant site.

Keywords: Oral metastasis, Primary tumour, Distant carcinoma.

I. Introduction

Metastasis is the process by which tumour cell leaves the primary tumour, travels to a distant site via the circulatory system and establishes a secondary tumour. This term was used by Joseph Claude Recamier, a French Physician (1862).¹Acquisition of tissue invasiveness and sustained angiogenesis are the prerequisite traits for metastatic process.²Oral metastatic lesions are rare and account for less than 1% of all oral malignant tumours.³Because of its rarity and deceiving clinical presentation, diagnosis of such lesion is challenging for both clinician and pathologist. Themajorities of cases go undetected due to micro metastasis or in view of the poor prognosis and terminal stage of disease, lose follow up or dead before presenting to a clinician.

II. Case Report

A 65 year female patient came with a complaint of pain and swelling in the lower left posterior region of the jaw since 3-4 months. Swelling increased in size rapidly after its appearance, accompanied by difficulty in speech. She had the habit of tobacco and betel nut chewing since 20-25 years. The patient suffered a chronic cough since 2-3 years with overall weak general health. Clinical examination revealed a roughly oval, firm soft tissue swelling in the left mandibular region, approximately 3.5×3 cm in size extending from parasymphysis to angle of mandible antero-posteriorly and crossing the inferior border of mandible. (Fig. 1) Left submandibular lymph node was enlarged, tender and fixed. Intraorally, the swelling was obliterating buccal vestibule extending posteriorly to retromolar area with ulcerated surface and indurated margin. (Fig. 2) Pantogram and periapical radiograph showed radiolucent lesion around related teeth due to periodontal bone loss.

Histopathological features of intraoral incisional biopsy specimen were showing islands of epithelial cells forming ductal patterns of varying sizes and shapes mixed with moderate degree of inflammatory infiltrate.(Fig. 3a and 3b)These ductal structures were lined by cuboidal or columnar cells with a pseudostratified arrangement in places. Lining cells were showing dysplastic features like nuclear hyperchromatism and pleomorphism. (Fig. 4) Ductal structures showed focal epithelial proliferations at places with pale eosinophilic, mucin like material in the lumen. Surface epithelium was ulcerated due to invasion from underlying lesional tissue. Overall features were suggestive of low grade adenocarcinoma.

CT neck revealed a soft tissue lesion of 3.1×2.7 cm in the left mandibular region with multiple cervical node involvement (Fig. 5) and CT chest revealed a lobulated soft tissue density lesion of 3.7×2.5 cm at the hilum of left upper lobe bronchus causing distal complete collapse of lung. Multiple enlarged aorto-pulmonary and pretracheal nodes were seen with largest being 1.3×1.3 cm. (Fig. 6) Patient was posted for CT guided biopsy for mass in lung but she refused to undergo any further investigative procedure. After 3 months of her first clinical visit, patient succumbed to disease. Therefore, the lesion in head and neck region was probably metastatic from a probable primary tumour in lung and diagnosed as low grade adenocarcinoma.

III. Discussion

Metastasis occurs when genetically unstable cancer cells adapt to a tissue microenvironment that is distant from the primary tumour. It is a Greek word meaning 'beyond place'. It results from the interplay of wandering tumour cells with permissive target tissues. Organ selectivity depends on pattern of blood flow as explained in mechanistic theory or by provision of a fertile environment in which compatible tumour cells could grow as in Seed and soil theory.⁴Oral region is an uncommon site for metastatic tumour cell colonization and is usually evidence of a widespread disease. In 25 % cases, oral metastases were found to be the first sign of metastatic spread and in 23 % cases; it was the first indication of an undiscovered malignancy at a distant site as in thepresent reportedcase.⁵Pathogenesis of metastasis in the oral cavity is not yet clear. Most prevalent routes are considered to be hematogenous route, althoughthe lymphatic route and direct spread are also among other routes. Batson (1942) had proposed that increased intrathoracic pressure directs the blood flow to valve less vertebral venous plexus from caval and azygous venous system, bypassing filtration from the lungs. This has been believed to increase distribution of metastatic cells to axial skeleton and head and neck region.²

The jaw bones, particularly the mandible, were more frequently affected than the oral soft tissues (2:1). The presence of hematopoietic marrow and sluggish circulation in the posterior mandible region makes it a preferred site for tumour cell lodgment. Newly formed, leaky vessels in a rich capillary plexus of inflamed gingival tissues and implantation of exfoliating cancer cells in a retrograde manner from the respiratory tract or by cough predisposes oral soft tissues to tumour cell colonization.³In present case, intrabony swelling was present around periodontally weakened teeth. The clinical presentation of the metastatic lesions differs between the various sites in the oral region. In the jaw bones, most patients complain of swelling, pain and paresthesia which develop in a relatively short period. In the oral soft tissues, the attached gingiva or alveolar mucosa is the most commonly affected site. Early manifestation of gingival metastases may resemble a hyperplastic or reactive lesion, such as pyogenic granuloma, peripheral giant cell granuloma, or fibrous epulis.⁵ In present case, patient complained of pain and rapidly enlarging swelling.

Localisation of the primary tumour is often difficult and sometimes is impossible.⁶Although no particular malignancy seems to favor spread to oral cavity, some primary tumours are found more often than others. The several retrospective studies by Hirshberg A et al (1993 & 2008), van der Waal RIF et al (2003) and Muttagi SS et al (2011), clearly mentioned that the major primary sites presenting oral metastases were the lung, kidney, liver and prostate for men and breast, female genital tracts, kidney and colorectum for women.^{3,5,7,8}Most of the lung primaries metastasizing to oral cavity are small cell carcinoma, adenocarcinoma, non-small cell carcinoma and squamous cell carcinoma.^{9,10,11,12} In present case, histopathological confirmation of lung lesion was not possible as patient was unwilling to comply and eventually succumbed to disease. However clinico-radiologic features were in favor of metastatic lesion in the oral cavity from unknown or most probably lung primary. Histopathological features of intraoral lesion confirmed the diagnosis of low grade adenocarcinoma.

IV. Conclusion

Although the oral cavity is not a common site for metastatic colonization, it is an interesting site because of the various tissues that may be involved in the metastatic process. In any case where the clinical presentation is unusual, especially in patients with a known malignant disease, biopsy is mandatory. Metastasis should be considered in the differential diagnosis and appropriate investigations should be carried out to rule out the possibility of metastatic lesion.

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Figure 1. Approximately 3.5 X 3 cm, firm extraoral swelling extending from parasymphysis to angle of mandible antero-posteriorly and crossing the inferior border of mandible.



Figure 2.Intraoral swelling obliterating buccal vestibule and extending posteriorly to retromolar area with ulcerated surface and indurated margin.



Figure 3. Islands of epithelial cells forming ductal patterns of varying sizes and shapes (arrows) mixed with moderate degree of inflammatory infiltrate. (10X, H&E).



Figure 4. Ductal structures lined by cuboidal or columnar cells with a pseudostratified arrangement in places and showing dysplastic features (40X, H&E).



Figure 5. Soft tissue lesion of 3.1×2.7 cm in the left mandibular region with multiple cervical node involvement (arrows) on CT neck.



Figure 6. Lobulated soft tissue density lesion of 3.7×2.5 cm at the hilum of left upper lobe bronchus causing distal complete collapse of lung on CT chest.