

CASE REPORT

From esophagus to gingiva: A rare case of metastatic adenocarcinoma

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ABSTRACT

Localized gingival enlargement is often associated with systemic medications, abscess formation, trauma, or reactive lesions. Very few reports are available reporting enlargement of gingiva due to metastasis of adenocarcinoma of the esophagus. A case of localized gingival growth affecting right maxillary and mandibular gingiva was reported in a 60-year-old male patient in which excisional biopsy of the lesion was done. Histopathological examination showed metastatic signet ring cell adenocarcinoma, which on further examination showed lower one-third of esophagus as the primary site of metastasis.

Key words: Adenocarcinoma, carcinoma of esophagus, localized gingival growth, metastatic carcinoma, signet ring cell

INTRODUCTION

Malignant metastatic involvements of oral tissues have been infrequently reported only in 1% of all oral malignancies.^[1-4] Breast, lung, kidney, thyroid, and prostate are the most common primary sites. Furthermore, metastasis to soft tissues is less common compared with the osseous structures, and metastasis to gingiva is very rare^[1,5,6] and usually occurs as gingival hyperplastic or reactive lesions with clinical appearance of pyogenic granuloma or epulis.^[7-9]

A case of metastatic signet ring cell adenocarcinoma of the esophagus in a 60-year-old patient is reported here in which the patient had no complaint related to the primary site and presented as multiple localized gingival growths.

CASE REPORT

A 60-year-old male patient came to OPD of Karnavati School of Dentistry, Uvarsad, Gandhinagar, Gujarat, with the chief complaint of mobile teeth in lower right back teeth region since 1 year and he noticed swelling of gums in right side upper and lower back teeth region since 1 month with occasional bleeding tendency. He had a habit of tobacco chewing for

15 years. He had history of exfoliation of 11, 18, and 26 before 4–5 years.

Intraoral examination revealed exophytic sessile growth of gingiva of 3×3 cm in size in right mandibular posterior region, which was extending from right mandibular first molar to third molar and extending into lower vestibular mucosa. Overlying mucosa was pinkish red in color. On palpation, growth was firm, non-tender, with 47 and 48 grade III mobile. Bleeding on probing was also present.

We found another exophytic growth of 2×3 cm in size with pedunculated base in maxillary right posterior region extending from distal of 15 to distal of 16 [Figure 1]. Overlying mucosa was normal in color. On palpation, growth was firm and non-tender.

Orthopantomographic examination showed mild-to-moderate bone loss in all the teeth except 27, 36, 41, 47, and 48, which showed severe bone loss with root resorption and 11, 18 and 26 teeth were missing [Figure 2].

We made diagnosis of generalized periodontitis with pyogenic granuloma in right maxillary and mandibular posterior region with differential diagnosis of other reactive lesions like fibrous epulis, giant cell lesion, and neoplastic lesion involving gingiva. Excisional biopsy of the lesion was carried out along with the removal of teeth 47, 48 [Figure 3]. Specimen was sent for histopathologic examination, which showed features of signet ring cell adenocarcinoma [Figure 4]. The possibility of metastatic lesion was considered because it is extremely rare for a signet ring cell adenocarcinoma to develop in gingiva

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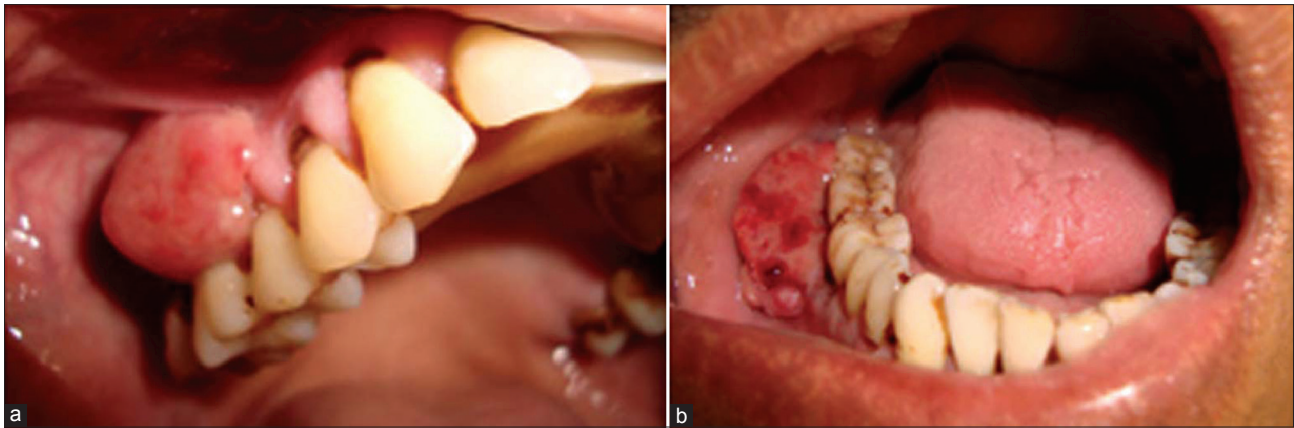


Figure 1: Lesions in (a) maxilla and (b) mandible

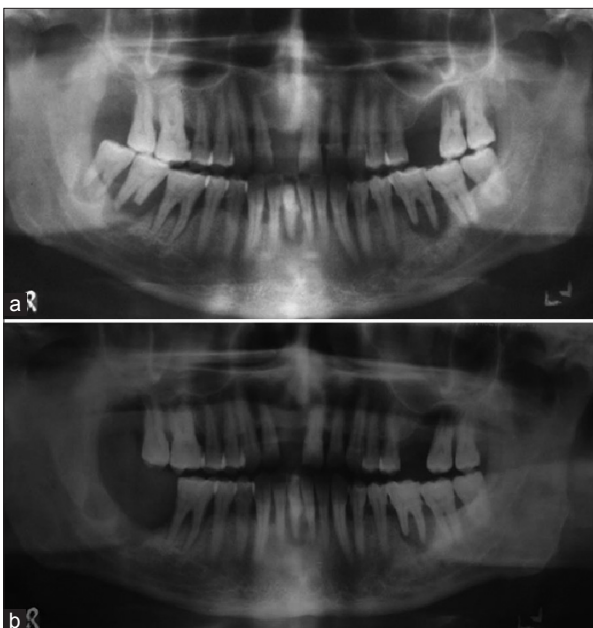


Figure 2: (a) Pre-treatment and (b) post-treatment OPG showing moderate-to-severe interdenal bone loss

as the primary site. Immunohistochemistry was performed to know more about tumor cells, which was positive for EMA (Epithelial Membrane Antigen), CK7 (Cytokeratin), and CK15 markers [Figure 5] and negative for CK20, TTF-1 (Thyroid Transcription Factor), and PSA (Prostate Specific Antigen) markers – suggestive of the GI tract as the possible primary site. The patient did not have any complaints related to any other region of the body like difficulty in swallowing or hematemesis.

The patient was referred to Gujarat Cancer Research Institute, Ahmedabad, for further examination. Barium swallow was performed, which showed 8 cm long defect in lower one-third of the esophagus. Endoscopy of the same region performed, which also confirmed esophageal involvement. Computed tomography (CT) scan of thorax was performed, which showed malignant lesion involving lower esophagus, gastro-esophageal junction, and fundus of the stomach [Figure 6].

A biopsy from esophagus was taken, which also showed histopathological features of signet ring cell adenocarcinoma. CT scans of other regions were taken to rule out metastasis but did not show any such involvement. The patient was advised for surgical treatment and chemotherapy but patient refused to take any treatment and he expired within 5 months of the final diagnosis.

As esophageal signet ring cell adenocarcinoma had not been recognized at the time of diagnosis of oral lesion. The oral lesion was identified as a metastatic signet ring cell adenocarcinoma with lower third of esophagus identified as the primary site.

DISCUSSION

There are two types of adenocarcinoma: signet ring cell type and mucinous type. Primary signet ring cell adenocarcinomas are mainly associated with the gastrointestinal tract, breast, or lung and have a more aggressive behavior.^[10] Squamous cell carcinoma has remained the most common esophageal malignancy between 1989 and 2004. It is unlikely that it will be surpassed by adenocarcinoma in number in the near future. The highest reported increase in esophageal adenocarcinoma has been reported in USA at 10% per annum. Among the Asian countries, China and Singapore have reported an increasing number of esophageal adenocarcinoma.^[7] Esophageal adenocarcinoma is the 15th most common cancer in developed country and the 4th in the developing world.^[7]

Esophageal adenocarcinoma tends to grow locally, invading surrounding tissues, which may lead to death.^[7] Most common invasion of esophageal adenocarcinoma is into tracheo-esophageal fistula/broncho-esophageal fistula. Mediastinum and lung are secondarily involved; lymphatic, liver, and spinal cord can be involved, rarely involving supraclavicular organs.^[2] Gastric signet ring adenocarcinomas have a lower rate ($\leq 8\%$) of lymph node metastasis than the non-signet ring cell type.^[10] Cases of gingival metastasis are usually found in the later stages of tumor growth and are most often associated



Figure 3: (a) Maxillary and (b) mandibular specimen of excisional biopsies with extracted teeth

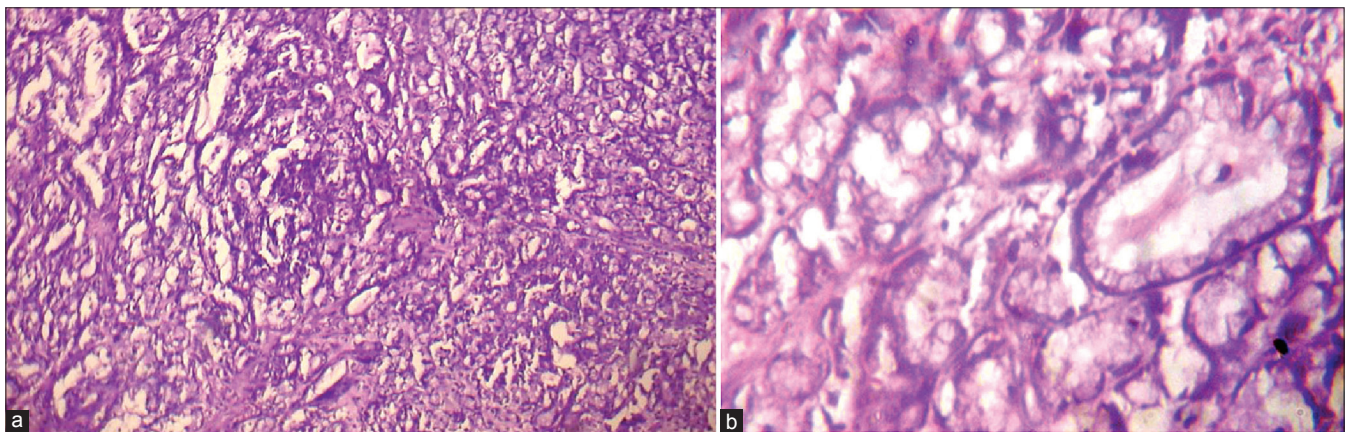


Figure 4: Histopathologic features of metastatic signet ring cell adenocarcinoma [H and E stain in (a) 10× and (b) 40×]

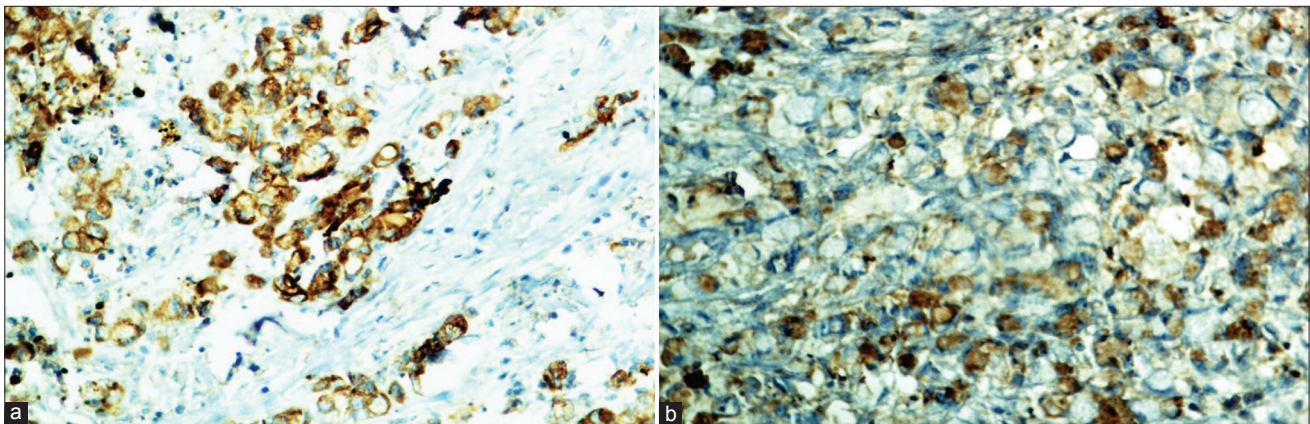


Figure 5: Immunohistochemistry stains positive for (a) CK7, (b) CK15, and EMA, 10× and 20× respectively

with metastatic deposits in other organs. Therefore it has a poor prognosis, leading to death of the patient within a few weeks or months.^[10] In this case, patient died within 5 months of diagnosis of metastatic signet ring cell adenocarcinoma. Despite this, an early diagnosis is must for successful treatment and prevention of other complications like tissue destruction and pain.^[5]

Like in the present case, clinically early gingival metastatic

lesions may imitate benign reactive lesions such as pyogenic granuloma, giant cell granuloma, or peripheral ossifying fibroma. The only difference can be rapid and progressive growth of the lesion. Definitive diagnosis requires histologic verification.^[10] Possible mechanism of gingival involvement of metastatic adenocarcinoma is not yet clear. Gingival metastases may be through the blood invasion of tumor cells. Batson proposed valveless vertebral venous plexus as the mechanism for bypassing filtration through the lungs in an

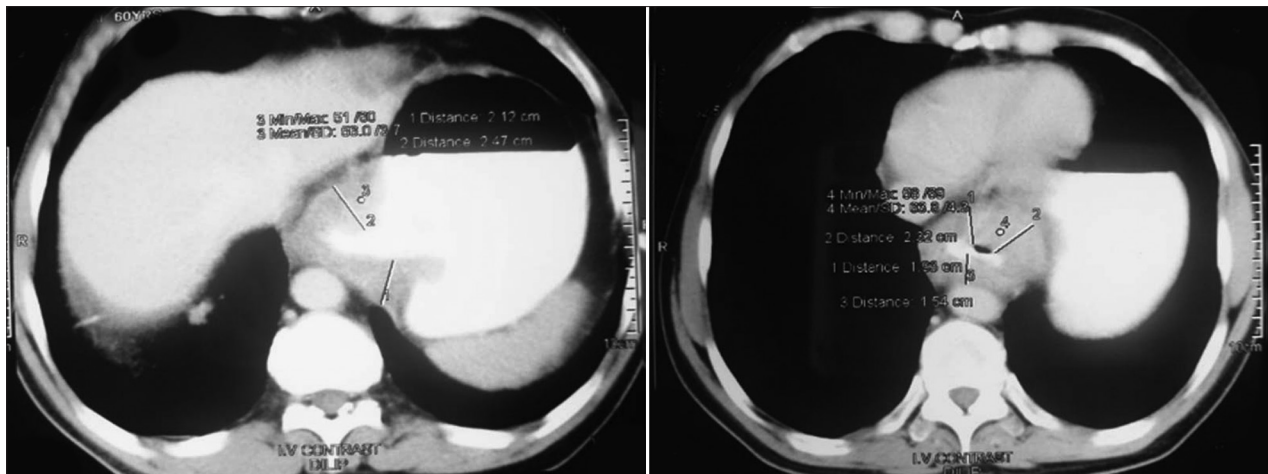


Figure 6: CT scan of thorax showing lesion involving the lower one-third of esophagus and gastro-esophageal junction

attempt to explain the metastasis to the oral region from a primary tumor of the lower part of the body, particularly in the absence of lung metastasis. Once circulating malignant cells reach the oral region, they can be trapped by the rich capillary network of the chronically inflamed attached gingiva.^[3,10]

The criteria for diagnosis of a metastatic neoplasm are: (1) primary lesion must be histologically verified, (2) histologically, the metastatic lesion must be the same subtypes as the primary lesion, (3) the possibility of direct extension from a primary lesion must be excluded.^[2,9,10] In the present case, gingival lesion was diagnosed before detection of the primary lesion (esophagus). According to Bhasker, approximately 33% of oral secondary tumors are an initial indication of the existence of a primary tumor.^[7] Histopathologically, gingival and esophageal lesions were same in terms of subtypes and morphology of tumor cells, so our final diagnosis was consistent with the above-mentioned criteria.

The treatment modalities are limited to conservative and palliative therapies intended to improve the quality of life of these patients, and include local resection, radiotherapy, or chemotherapy. Most of patients die within the first year of diagnosis with a survival rate at 4 years of 10% cases.^[3]

CONCLUSION

In the patients with the history of malignant tumors or with a rapidly growing oral soft tissue mass, possibility of the metastatic tumor should be considered in differential diagnosis. Detailed and careful oral examination is must to determine if there are any metastatic lesions that could manifest in the oral cavity, with or without swelling, pain, and looseness of the teeth.

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