

Intestinal-type Adenocarcinoma Presenting as a Mandibular Mass: An Unusual Presentation

Abstract

Intestinal-type adenocarcinoma is an aggressive malignancy with the rare possibility of metastatic spread to the mandible. We represent a case of a 30-year-old female patient who reported with a chief complaint of a right mandibular mass for 4 months. Orthopantomogram X-ray observed an ill-defined lytic lesion with severe periosteal reaction. Incisional biopsy revealed “colonic variant of intestinal-type adenocarcinoma.” Expression of cytokeratin 20 and caudal type homeobox transcription factor 2 (CDX-2) markers was seen on immunohistochemistry. Biochemical markers such as serum cancer antigen 125, serum alkaline phosphatase, and serum carcinoembryonic antigen were raised. Contrast-enhanced computed tomography of the thorax and abdomen suggested cavitory lesions in the bilateral lungs, right adrenal gland, and mediastinum. Positron emission tomography revealed multiple lesions in the body, hence confirming the diagnosis. The patient was kept on palliative chemotherapy. It is concluded that prompt diagnosis and initiation of treatment increases the chances of survival in such cases.

Keywords: Adenocarcinoma, intestinal adenocarcinoma, mandibular metastasis, metastasis

Introduction

Intestinal-type adenocarcinoma is a subset of sinonasal adenocarcinoma that occasionally resembles intestinal carcinomas/adenomas and is often mistaken for the same.^[1] It is an aggressive lesion that can metastasize to various parts of the body including upper airways, lungs, ethmoid sinus, nasal cavity, maxillary antrum, orbit, and skull base, but is infrequently seen involving the mandible.^[2] Various malignancies can metastasize to oral region, but intestine is not the most common primary site. The spread of such lesions intraorally is an indicator of its poor prognosis. A PubMed literature search yielded only five case reports till date presenting intestinal adenocarcinoma in the mandible.^[3-7] Hence, we report an unusual case of a 30-year-old female who initially presented with a mandibular mass which was later found to be metastatic from primary intestinal-type adenocarcinoma of colon. A series of investigations and development of specific markers are required for its prompt diagnosis, which can help in initiation of early treatment and hence prolonging its survival.

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Case Report

A 30-year-old female patient presented with intraoral soft tissue growth of the right posterior mandible [Figure 1]. She was apparently normal 4 months back when extraction of three teeth was done in the same region from a local clinic due to their Grade 3 mobility which interfered with chewing ability of the patient, following which the growth developed suddenly on the same day. There was no associated pain, swelling, or discharge prior to the extraction of the said teeth. Growth was pea shaped initially, which gradually increased to the present size. It was associated with occasional pain, and no episode of fever was reported. Medical history and general physical examination revealed no significant abnormalities. On inspection, an ulcerative lesion of size 4 cm × 1.3 cm × 1.5 cm was observed extending from the distal to the right lower second premolar till the right anterior border of ramus, causing buccal vestibule obliteration and mild expansion of lingual cortical plate in the same region. Right mandibular nerve paresthesia was present. Palpation elicited tenderness and confirmed the inspectory findings. No associated

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Figure 1: Intra-oral picture showing growth with respect to right body mandible

blood or pus discharge was observed. Right submandibular lymph node was palpable, one in number, 1 cm × 1 cm × 1 cm in size, not completely mobile, with no tenderness. Routine blood investigations were done, which were within normal limits (total leukocyte count: 7.64 thousand/mm³, red blood cell count: 3.79 million/mm³, hemoglobin: 8.35 g/dL, hematocrit: 29.29%, mean corpuscular volume: 77.21fL, mean corpuscular hemoglobin: 22.02 picograms, platelet count: 314.2 thousand/mm³, prothrombin time: 13.4 s, international normalized ratio: 1.18, urea: 21.4 mg/dL, serum creatinine: 0.68 mg/dL, sodium: 138.6 mEq/L, potassium: 4.2 mEq/L, chloride: 100.8 mEq/L, calcium: 9.0 mg/dL, uric acid: 2.7 mg/dL, phosphorous: 3.8 mg/dL, total serum bilirubin: 0.4 mg/dL, alanine transaminase: 17.7 U/L, aspartate transaminase: 27.1 U/L, serum proteins: 7.30 g/dL, serum albumin: 4.50 g/dL, globulin: 2.8 g/dL) and (viral markers, i.e., HIV, hepatitis B virus, and hepatitis B antigen: all negative).

Orthopantomogram X-ray and noncontrast computed tomography of the face was done which depicted mixed radiolucent-radio-opaque lesion involving right posterior mandible, extending from distal of second premolar till third molar region, with radiolucency predominantly in the third molar region [Figure 2a]. Incisional biopsy of the mass revealed infiltration of the submucosa with tumor cells arranged in glands with nuclear stratification and intracytoplasmic mucin suggestive of colonic variant of intestinal-type adenocarcinoma [Figure 3]. Immunohistochemistry of the sample expressed cytokeratin 20 and caudal type homeobox transcription factor 2 (CDX-2) markers which are specific for the above diagnosis.

Further, colonoscopy was carried out depicting ulcerated and narrowed illeo-caecal valve along with necrotic area which was biopsied for confirmation of the diagnosis. Biochemical markers like serum cancer antigen 125 (98.20 U/ml; normal value: <35 U/ml) and serum carcinoembryonic antigen (34.37 ng/ml; normal value: <5 ng/ml) were found to be raised.

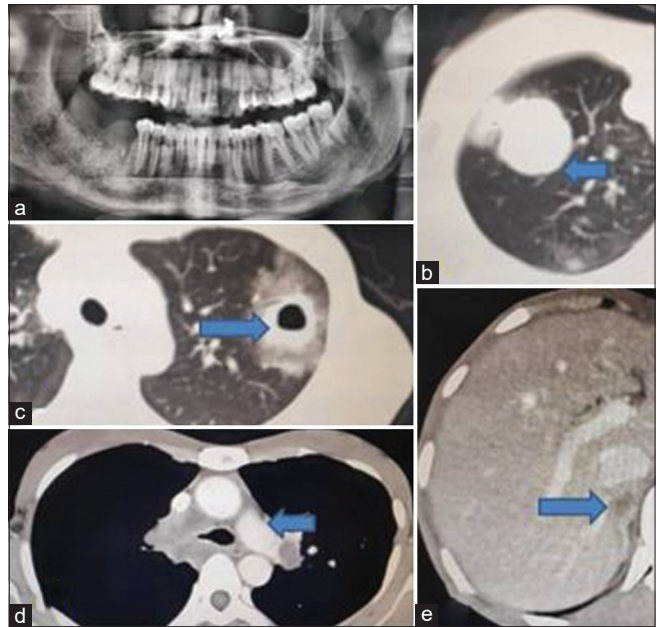


Figure 2: Orthopantomogram X-ray depicting mixed radiolucent to radio-opaque lesion with respect to right posterior mandible region. (a) Contrast-enhanced computed tomography of the thorax and abdomen depicting cavitary lesions in bilateral lungs. (b and c) Mediastinum. (d) Right adrenal gland (e)

In order to assess distant tumor metastasis, contrast enhanced computed tomography of thorax and abdomen was done which was suggestive of cavitary lesion in bilateral lungs, mediastinum, right adrenal gland and right femoral neck [Figure 2b-e]. Positron emission tomography confirmed metabolically active lesions in above-mentioned regions along with subcutaneous and intramuscular deposits and; multiple skeletal lesions including right hemi-mandible.

All the above investigations substantiated the diagnosis of metastatic colonic variant of intestinal-type adenocarcinoma of colon. The patient was kept on palliative chemotherapy (5-fluorouracil and cisplatin) due to its widespread dissemination representation, including the mandible.

Discussion

Metastatic spread to oral cavity occurs due to various primary neoplasms. It might involve soft tissues of the oral cavity or jaw bones.^[8] Oral soft tissue sites involved in metastasis are mainly gingivae (55%) and tongue (30%).^[9] Most common primary site of carcinoma causing metastasis to jaw bones are breast in women and lungs in men. Intestinal-type adenocarcinoma rarely causes metastasis to mandible.^[2] Metastatic spread can occur through hematogenous route, transcoelomic permeation, lymphatic route, local infiltration or a combination of the above.^[10] Mandible is more commonly metastasized as it usually spreads through hematogenous route; and involves the posterior mandible which is rich in marrow

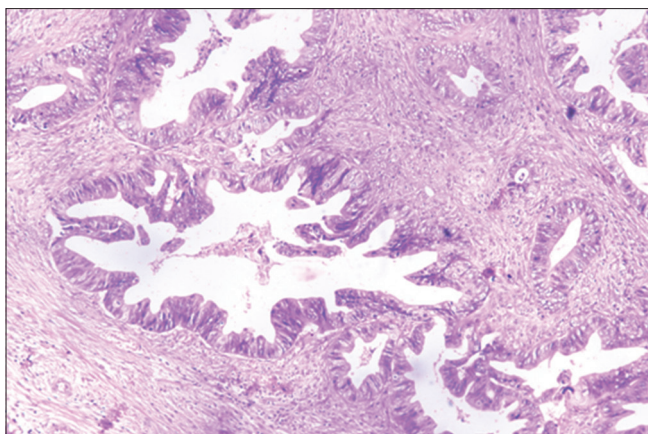


Figure 3: Histopathology report shows tumor cells arranged in glands suggestive of colonic type of intestinal-type adenocarcinoma

spaces.^[3] Adenocarcinoma cells are seen to be positive for CK20 and caudal type homeobox transcription factor 2 (CDX2) in immunohistological analysis. CDX2 is considered to be a specific marker for intestinal type adenocarcinoma.^[11] Histologically, there are five variants of intestinal-type adenocarcinoma: colonic, papillary, mucinous, solid and mixed.^[1]

According to literature, symptoms at the site of secondary deposits might occur prior to the diagnosis of the primary tumor; which can be indicative of progressive disease state.^[12] Appropriate treatment is selected carefully after evaluating the tumor dissemination.^[13] In case of widespread metastasis having poor prognosis, palliative radiotherapy and chemotherapy is recommended which is equivalent to our case report where widespread metastatic sites were observed and hence resection of the intestinal lesion was not possible.^[13] The mean survival rate is found to be 7 months in cases showing oral metastasis, similar to our reported case where the patient survived for just 5 months.^[12]

Although adenocarcinoma of colon is frequently encountered, its intestinal-type metastasis to mandible is a rare presentation.^[14] Primary cancerous site might be diagnosed after the identification of its metastatic lesions. Orofacial symptoms should not be overlooked as they can be sign of distant tumor metastasis; which is an indication of a widespread disease having poor prognosis. A series of investigations including radiographs, biochemical and immunohistological markers, computed tomography, positron emission tomography, colonoscopy, and complete blood picture are required for confirming the diagnosis. Development of specific markers for differentiation of intestinal carcinomas can promote early diagnosis. Palliative treatment must be considered in case of advanced metastasis. Also, prompt diagnosis and initiation of treatment increases the chances of survival in such cases.

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.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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