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Case Report

Bilateral Mandibular and Bilateral Maxillary Cancer: A Rare Case of Tumor Metastases from The Breast

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ABSTRACT

Tumor metastases are very rare causes of jaw lesions. Previous reports have indicated occurrence in one or more quadrants, but metastatic lesions in all four quadrants are exceptionally rare. This study aimed to report an unusual presentation of metastatic carcinoma, in which metastatic deposits from the breast were found on the four quadrants of the jawbones. We present an unusual case of metastatic lesion from the breast to the mandible and maxilla bilaterally in a 46-year-old woman. The patient was admitted to a previous treatment of breast cancer only after several promptings. The clinical and microscopic features of the jaw lesions and the microscopic features of the breast lesion are presented with a review of the pertinent literature.

INTRODUCTION

Metastasis can be defined as the spread of diseased/pathological cells from one organ or part of the body to another that is not directly connected with it.[1] Metastasis occurs via different routes, which may include the blood, lymphatics, or detachment, and direct implantation of the diseased cell or the combination of the above[1]. The lungs, liver, brain, and bones are usually the recipient sites of metastatic cells[1], though metastasis can occur at any part of the body.

Metastasis from any site to the maxillofacial region comprises about 1% of all orofacial malignancies.[2] In the maxillofacial region, the majority (about 85%) of the metastatic tumors are located in jaw bones, with the molar and retromolar mandible as the most commonly affected sites (about 80-90%).[3] Fewer metastatic lesions are found in the soft tissues and salivary glands.[4] Few cases of mandibular metastasis of breast carcinomas have been reported, and cases of its metastasis to the maxilla are quite rare.[5,6]

Some of the signs and symptoms associated with tumors metastatic to the maxillofacial region include pain, swelling, altered sensation, halitosis, gingival irritation, ulceration, tooth mobility, exophytic masses of the soft tissues, trismus and, rarely, pathologic

fractures.[7, 8, 9, 10] Altered sensation in the lower lip and chin can also occur.[6]

This report is a case of metastatic lesions from the breast to the mandible and maxilla bilaterally after three years post-radical mastectomy. The purpose of the report is to present a very rare occurrence of bilateral mandibular and bimaxillary involvement in tumor metastases and the challenges of management in a resource-scarce environment.

CASE REPORT

A 46-year-old woman presented with a 6-month history of swelling of the right mandible and a two-month history of mobile upper left first and second molars. On examination of the right mandible, a firm, fleshy buccal mass was noted around the first to third molars buccally, it extended from parasymphyseal region anteriorly to the retromolar region posteriorly (Figures 1a &b).

On the left mandible was a palpable, bony hard, buccal swelling around the lower left second premolar and first molar (Figure 1c). Also noted was the swelling on the upper left jaw extending from the tuberosity to the level of the first molar on both the buccal and palatal sides, with areas of ulceration on the palate area adjacent to the second and third molars (Figures 2a, b & c).

Mobile teeth were 16, 17, 28, 36, 37, 38, 44, 45, 46, 47 and tooth 48. Mouth opening was adequate with an interincisal distance of 3.7cm. A craniofacial CT scan revealed destruction of the right mandible, an area of hypodensity on the left mandible, signifying a destructive process, an area of enhanced isodensity with areas of hypodensity on the upper left and right maxillae, signifying bone destruction, and areas of suspected intracranial metastasis (Figures 3a, b, c & d and 4a, b, c, and d).

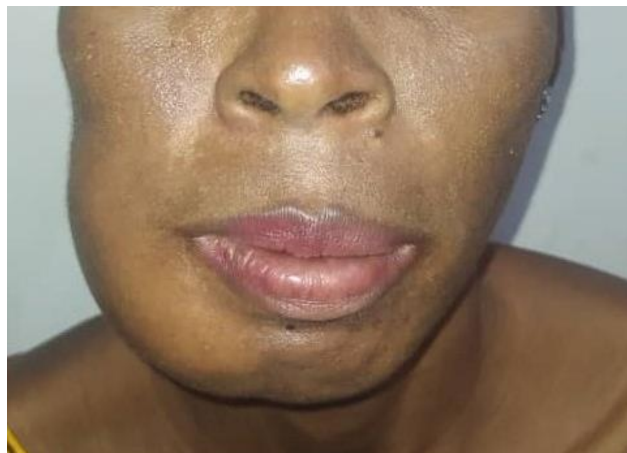

Figure 1a: Extraoral views of bilateral mandibular swellings (frontal view)

Figure 1b: Extraoral views of bilateral mandibular swellings (right lateral view)

Figure 1c: Extraoral views of bilateral mandibular swellings (left lateral view)

Figure 2: Intraoral swellings from metastatic tumor to the jaws (a), arrow shows lesions of left and right maxilla (b), arrow shows ulcerated mass of right mandible and (c).

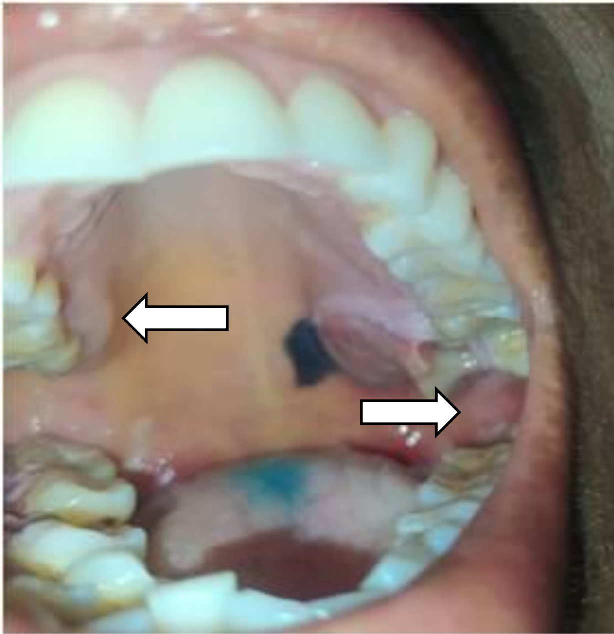

Figure 2a: Arrows show lesions of left and right maxilla

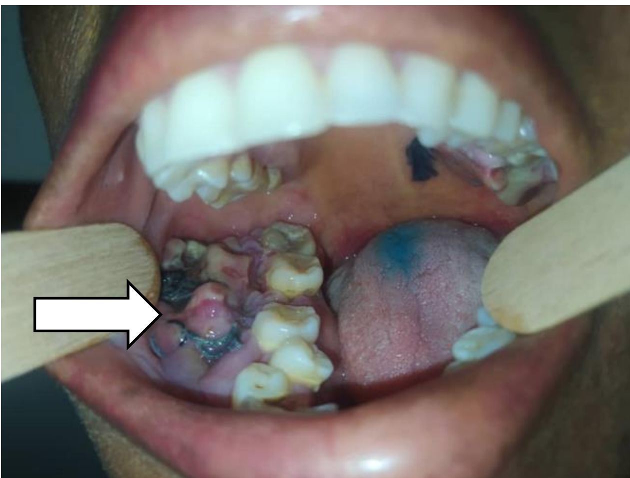

Figure 2b: Arrow shows ulcerated mass of right mandible

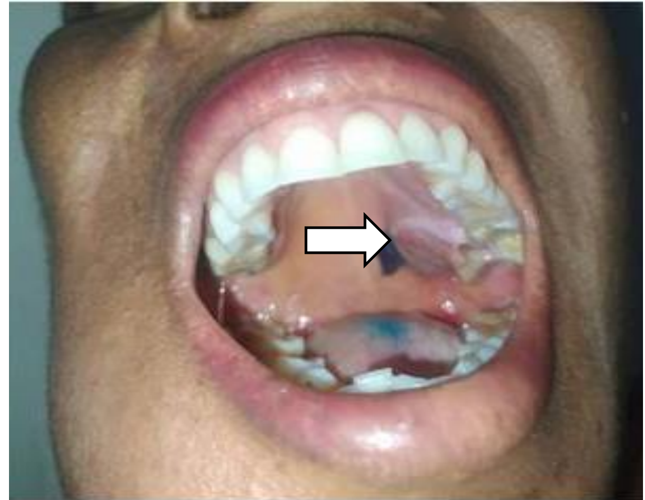

Figure 2c. arrow shows ulcerated mass of right maxilla

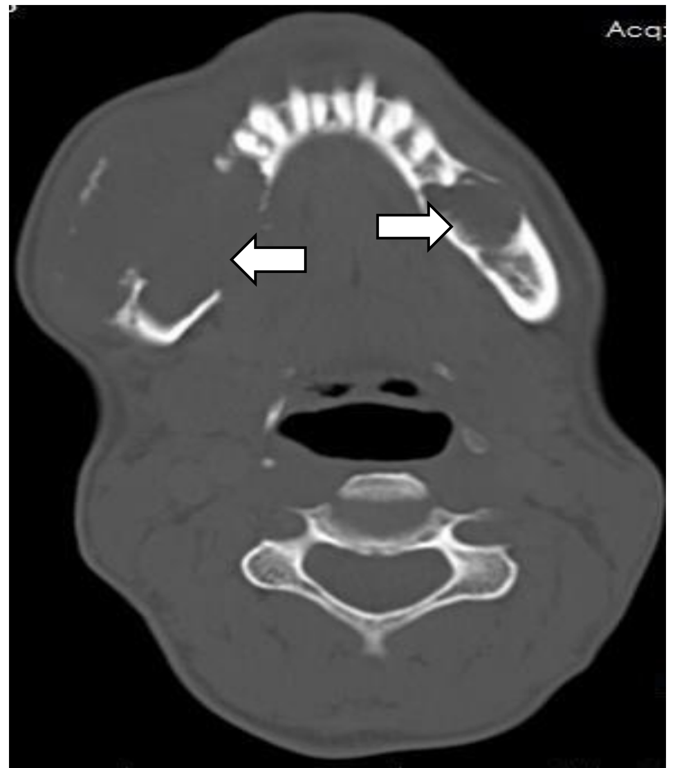

Figure 3a: Axial CT scan of mandible showing destructive lesion (bone window)

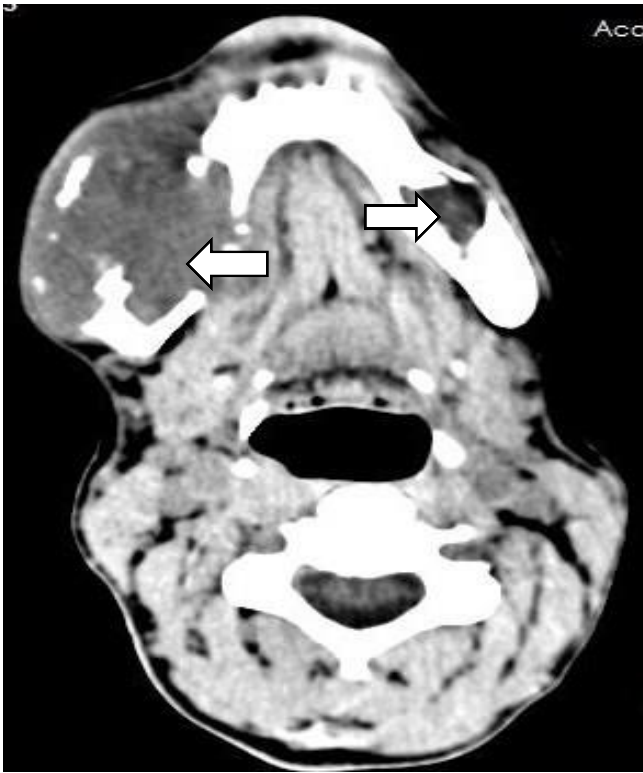

Figure 3b Axial CT scan of mandible showing destructive lesion (soft tissue window)

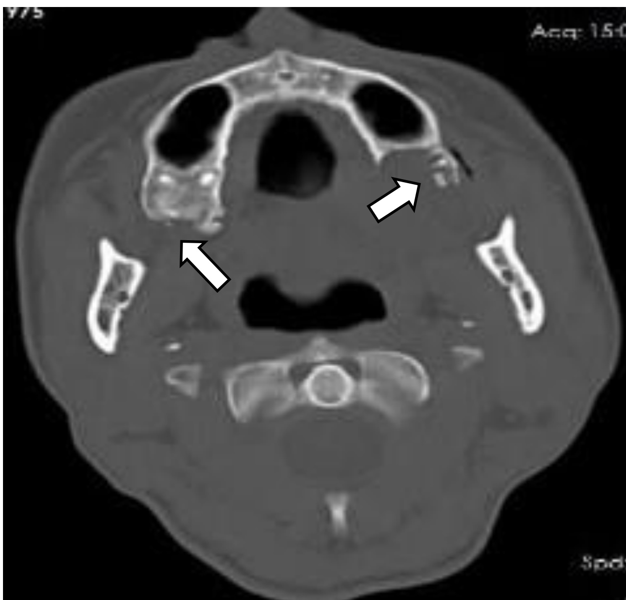

Figure 3c. CT showing bony destruction of right maxillae (bone window)

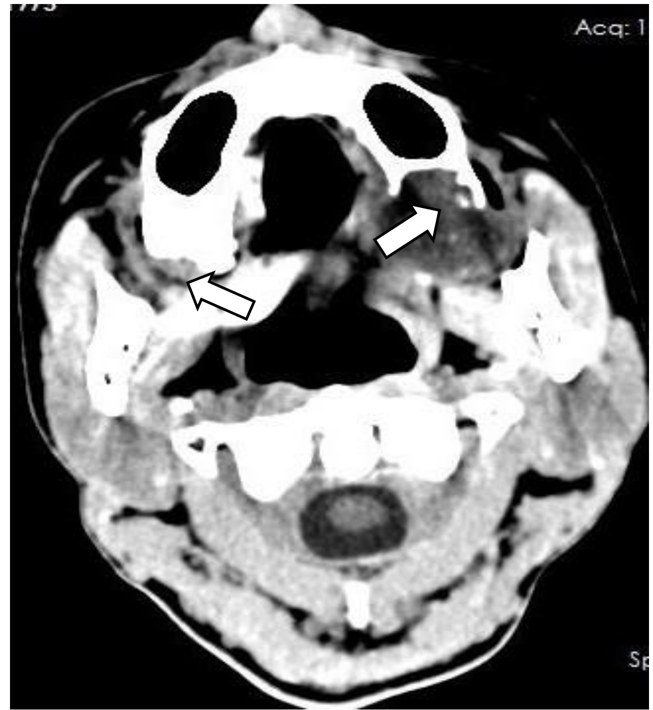

Figure 3d. CT showing bony destruction of right maxillae (Soft tissue window)

HISTOPATHOLOGY

The histopathology of pieces from the oral lesions revealed well-differentiated parakeratinized squamous epithelium with an underlying connective tissue infiltrated by mixed inflammatory cells. Within the connective tissue was a tumor mass disposed in a trabecular and ductal pattern. Some islands of tumor cells had central cystic spaces, individual tumor cells were small and round with scanty cytoplasm and hyperchromatic nuclei. Clear cell differentiation was observed in areas and hyalinization of the periphery of the tumor. A diagnosis of intraosseous mucoepidermoid carcinoma (intermediate grade) was pronounced with the proviso to rule out the possibility of a metastatic lesion. Specimens from all quadrants showed similar features.

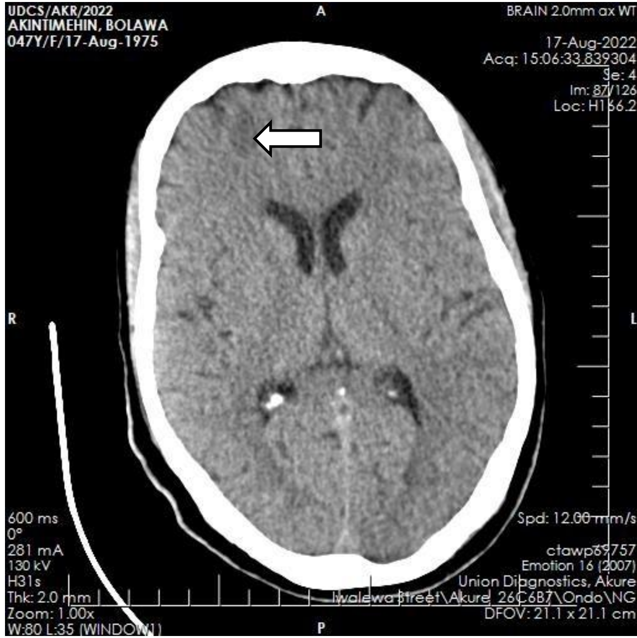

Figure 4a: Possible tumor metastases to the brain (temporal region)

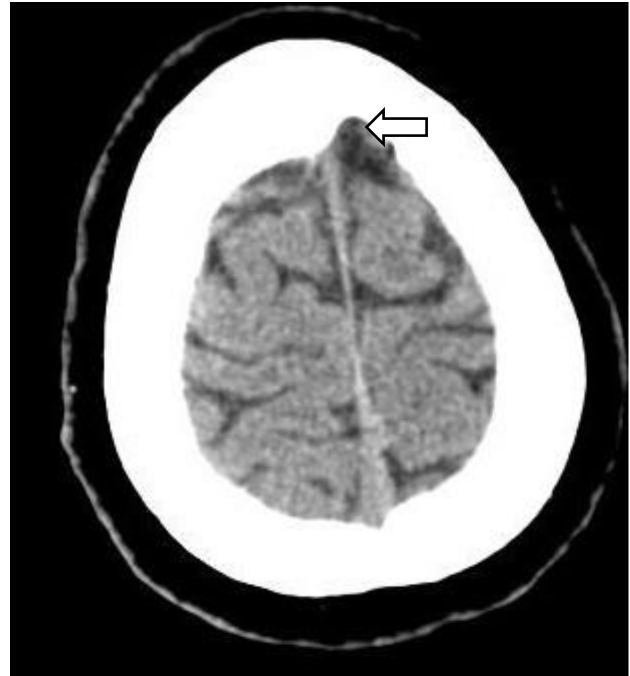

Figure 4c: Possible tumor metastases to the brain (frontal region)

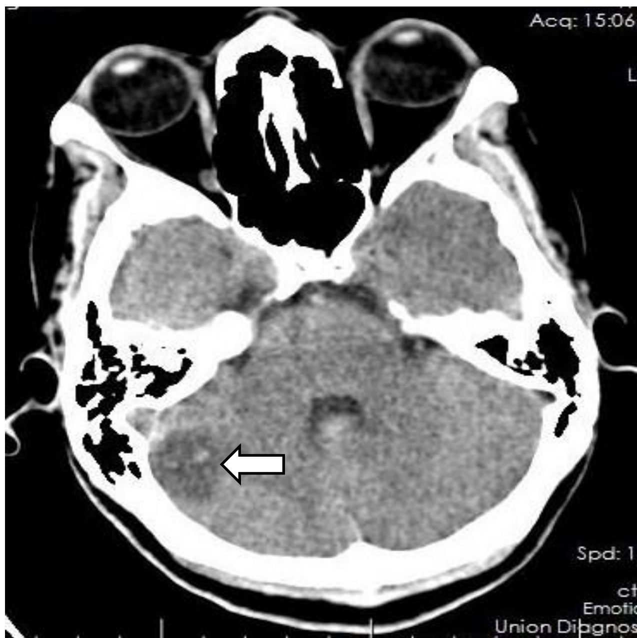

Figure 4b: Possible tumor metastases to the brain (temporal region)

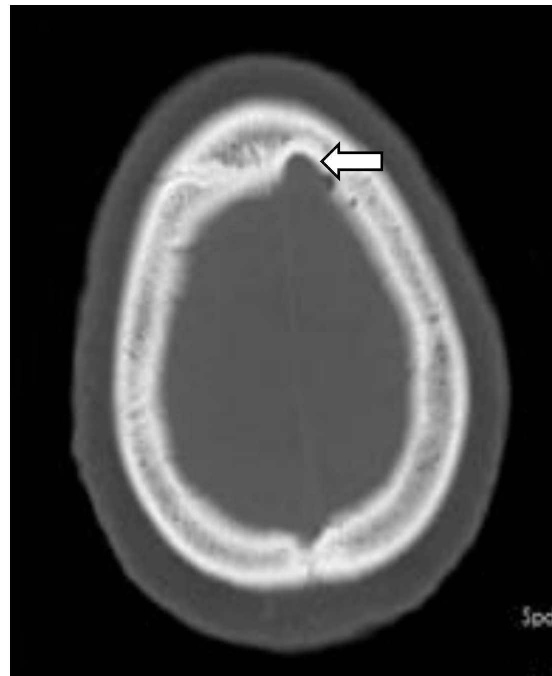

Figure 4d: Possible tumor metastases to the brain (frontal region)

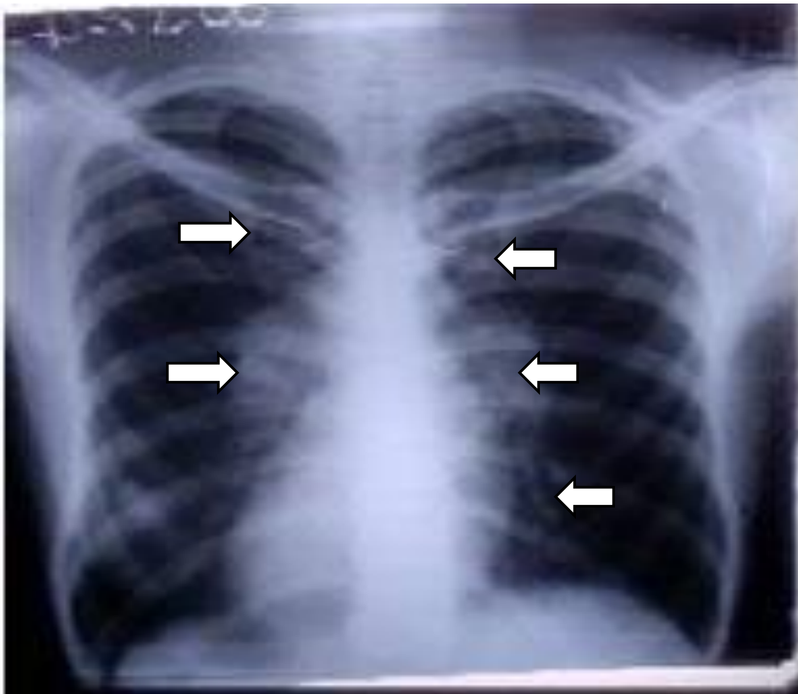

Figure 5: Antero-posterior radiograph of Chest showing prominent hilar marking and a nodule in the lower right chest suggestive of cannon balls

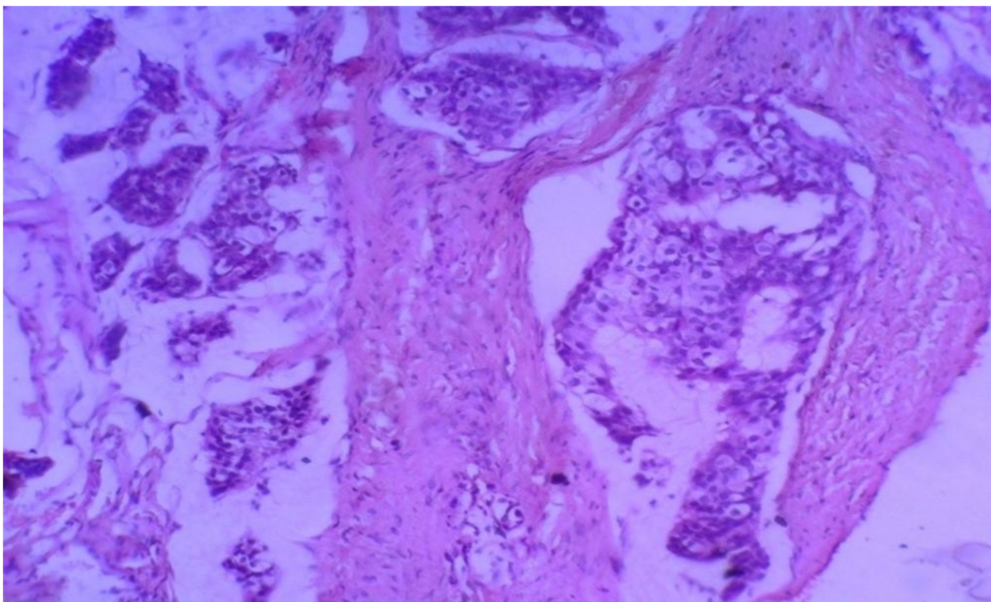

Figure 6: Photomicrograph of jaw swelling showing solid nests of squamous epithelial cells intermixed with mucus-producing cells (Hematoxylin and eosin stain at ×100)

Comparing the histopathologic findings with the breast lesion histopathology, similarities were observed. A review of the breast tissue histopathology revealed proliferating malignant ductal epithelial cells with few areas of ductal formation. Malignant ducts were seen floating in pools of mucin. There were areas of necrosis and infiltration by chronic inflammatory cells with the diagnosis of infiltrating ductal carcinoma (mucinous variant), Nottingham grade II (Figure 7).

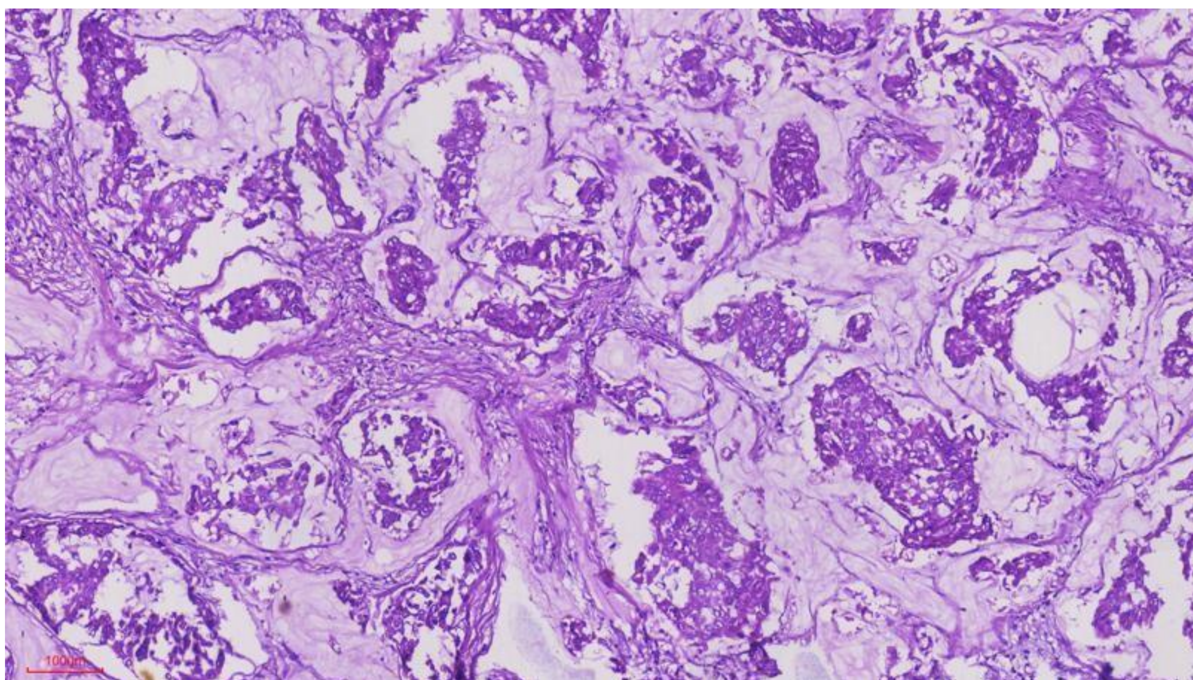

Figure 7. Photomicrograph of the primary breast lesion showing proliferating malignant ductal epithelial cells with few areas of ductal formation (Hematoxyline and eosin stain at ×40)

The final diagnosis of bilateral mandibular-maxillary glandular carcinoma secondary to metastatic breast carcinoma was made. The patient and caregiver were counselled and advised to enrol for palliative care. However, the patient preferred to be discharged from the hospital. One week after discharge from the hospital, she presented with altered sensorium, and was readmitted for care, but her condition deteriorated, and she died after two days of stay in the hospital.

DISCUSSION

Metastasis of a tumor to the jaw and oral cavity is uncommon when it arises; it poses a diagnostic challenge. This lack of specific signs and symptoms means that clinical presentations vary, may sometimes be vague or innocuous and may mimic dental infections. Some cases of metastasis to the jaws are asymptomatic.[8, 11, 12, 13, 14]

Pain is said to be the most common symptom associated with metastatic jaw cancers.[11] Other features include swelling, intraoral mass, mobile or extruded teeth, cortical expansion, regional lymphadenopathy, gingival irritation, mucosa ulceration, exophytic growth, halitosis, paresthesia of the lower lip and trismus.[2, 14, 15, 16] Pain was the major reason for presentation in this case.

Differences in sex at birth determine the primary site of origin of metastasis to the jaws. In women, breast, genitourinary or gynecologic sites, kidney, and colorectal region are the most common primary sites, while in men, they may originate from the lung, kidney, liver, and prostate.[3, 17] Breast cancer has been described as the most common cause of cancer deaths in females worldwide, accounting for 15% of deaths from cancer.[18] Mortality has been attributed to late presentation and inadequate access to comprehensive cancer care, especially in low and middle-income countries where mortality has remained high.[19, 20, 21] In a Dutch study, all metastases to the jaws among women originated from the breast, [17] which is not different from our findings. This present case affected the hard tissues of all the jaw quadrants, indicating that metastasis to the mandible or maxilla is more likely than to the soft tissue of the oral cavity.[22] While breast cancer metastasis to the maxilla is very uncommon.[21] This patient exhibited bilateral maxillary and bilateral mandibular involvement.

Metastatic tumor presentation in the posterior part of the jaws, in this case, is similar to those reported in the literature, in which the posterior mandible and maxilla are the affected sites. This has been attributed to the ability of those parts of the jaws to contain red marrow with the ability to continue producing blood cells and blood vessels.[23, 24] The presence of many blood vessels, and reduced velocity of blood flow favour tumor seeding and cell growth.[23, 24]

The radiographic appearance of metastatic disease in the jaws varies from well-defined to poorly defined radiolucency, often described as a ''moth-eaten'' appearance.[24]Metastatic carcinomas from the breast and prostate might stimulate bone formation; hence, these might appear as mixed radiopaque radiolucent lesions.[8] Sometimes, there may be no radiographic changes.[8, 22] In the present case, the radiographic findings were that of a destructive lesion of the jaws, as evidenced by areas of hypodensity [Figures 4 and 5]. This is to improve the patient's quality of life[21] and may include radiotherapy, chemotherapy, hormone therapy, and, rarely, surgical intervention. [24, 25]

The management of patients with distant metastases to maxillofacial bones and other bones is usually and primarily palliative[21, 24, 25] This is to improve the patient's quality of life[21] and may include radiotherapy, chemotherapy, hormone therapy, and, rarely, surgical intervention. [24,25] Management of some of the associated orofacial complications, such as discomfort during mastication, odynophagia, and halitosis, is also of paramount importance.[21] Adequate attention should be given to pain management and avoidance of possible infections, fractures, or hemorrhage.[21,24]

Opioid analgesics and other non-pharmacological approaches can be used for chronic neuropathic pain.[21] Furthermore, assessment and management of presenting physical symptoms such as pain, breathlessness, fatigue, and delirium and psychological symptoms such as anxiety and depression cannot be overemphasised.[21] Patients with advanced breast carcinomas with psychological symptoms may benefit from anxiolytics, antipsychotics, and supportive psychotherapy (with cognitive therapy).[21] While caring for these terminally ill patients, their relatives (family caregivers) are also considered in the anxiolytics, antipsychotics, and supportive psychotherapy, as was done in this case.[26]

Generally, the prognosis of metastatic oral cancer is poor due primarily to delay in the detection and diagnosis of the lesions.[12] The average survival time for patients with metastatic tumors in the oral cavity is six to seven months, with approximately 70% of patients dying within a year of diagnosis[12, 17, 22] as in the presented case when the patient died within seven months after the presentation of the swelling of the mandible. Most patients with oral metastases have already developed generalised metastases by the time of diagnosis, such as the case being presented, with intracranial and possible pulmonary involvement.[12]

There are challenges in managing patients with distant metastases to maxillofacial bones in a resource-scarce environment like ours, as many factors come into play. These include delays in patient presentation to the maxillofacial surgeon due to non-specific features of the condition, delays in the diagnosis due to a low index of suspicion by clinicians and paucity of resources for investigations. Late presentation limits treatment options, while patient factors such as lack of funds for prescribed treatment due to low health insurance coverage also affect optimal management.

In this patient, the decision to opt for discharge rather than go for palliative care probably had no implication on the ultimate outcome of the disease, as metastatic carcinoma is usually associated with poor prognosis. The migration and mass exodus of health professionals from low to middle-income economies like Nigeria [23] also impairs the early diagnosis and optimal treatment of patients with malignancies such as breast cancer metastasis to the jaws.

CONCLUSION

Metastases to the jaws have been reported in the literature, but reports of metastasis to all the quadrants of the jaw in a breast cancer patient are rare. This case shows the importance of an adequate history of a patient's health status towards diagnosis. The benefit of good record keeping cannot be underestimated, as the previous hospital provided the tissue specimen that shows the initial cancer diagnosis. It is not known whether the patient was aware of the implications of the initial cancer diagnosis necessitating a follow-up. With more resource allocation to health, tumor recurrence would be detected early via the availability of a whole-body scan.

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