Sir,

Ameloblastoma are benign locally aggressive tumors accounting for about 1% of all oral tumors. Metastasis occurs in 2-5% of patients and lung is approximately involved in 80% of cases. Solitary or multiple lung nodules are the common manifestations, cavitating nodules have never been reported. Here we report probably the first case in literature about multiple cavitating lung secondaries in a 38-year-old female, who was previously diagnosed 5 years back to have ameloblastoma of right mandible.

A 38-year-old female presented to pulmonary medicine outpatient department (CMC Vellore, Tamilnadu, India) with complaints of dry cough and exertional breathlessness of 3 months duration. There was no history of fever, loss of appetite, weight loss, hemoptysis, chest pain or symptoms suggestive of connective tissue disease (CTD) or vasculitis. Our patient had significant past history and 5 years back she was diagnosed to have benign ameloblastoma of right mandible [Figure 1a] and underwent total excision and repair with fibular graft [Figure 2a and b]. She was in regular follow-up and there was no signs of recurrence.

Her general examination, systemic examination and laboratory investigations were within normal limits. Chest X-ray revealed [Figure 3a] multiple bilateral lung nodules with cavitation, and the same was confirmed by computed tomography [Figure 3b]. Her old chest X-ray taken 5 year back [Figure 3c] was normal. Her routine biochemistry and serology for CTD and vasculitis were negative. Bronchoscopy was done and the cultures were negative for mycobacteria and malignant cells were negative in cytology. Computed tomography (CT) guided transthoracic needle biopsy was done from one of the nodules and the histology showed features suggestive of metastasizing ameloblastoma [Figure 1b]. Local recurrence was ruled out with Mandibular radiographs. Metastasis to other organs were ruled out with appropriate investigations. She was referred to oncology department for further treatment. Patient was advised palliative chemotherapy, the same was refused by her and was lost to follow-up.

Ameloblastoma are uncommon slow-growing benign tumor arising from odontogenic epithelium of jaw. Usually occurs in middle-age group (20-40) and the most common site is mandible (80%). These tumors are locally aggressive and rarely metastasize.

Metastasizing ameloblastoma is placed along with ameloblastic carcinoma under the general grouping of
odontogenic tumors in the classification proposed by the World Health Organisation. High-grade malignant potential, marked cellular atypia and increased mitotic activity are the characteristic features of ameloblastic carcinoma. Metastasis is not a definite phenomenon in ameloblastic carcinoma. Metastasizing ameloblastoma is the benign form which metastasizes and it lacks features of malignancy in the primary as well as in metastatic lesion.

Lung is the most common site of metastasis (upto 88%) followed by regional lymph nodes, vertebrae, pleura, skull, parotid gland, diaphragm and liver. Long duration of case, extensive initial disease, mandibular focus of primary tumor, multiple surgical and radiation procedure are associated with high likelihood of developing metastasis. The exact mechanism of metastasis is unclear, the proposed mechanisms are hematogenous route, lymphatic route or due to aspiration of tumor cells in to lung.

Radiologically lung metastasis in ameloblastoma presents as solitary or multiple lung secondaries. Ameloblastoma presenting as cavitating lung secondaries probably not reported previously in literature and the common cause of cavitating lung lesions are primary lung malignancies with squamous histology followed by adeno- and large cell carcinoma. Multifocal bronchoalveolar carcinoma can present as multiple cavitating lesions. Metastatic squamous cell carcinoma arising from uterine cervix, larynx, pharynx, and esophagus, adenocarcinoma of colon or rectum, transitional cell carcinoma of bladder and lymphoma especially primary pulmonary are the common causes of lung secondaries with cavitation.

The non-malignant cause of cavitating lung lesions include infections like tuberculosis, atypical mycobacterial infection, Nocardia, Staphylococci, Klebsiella and histoplasmosis. Non-malignant non-infectious causes include immunological conditions like rheumatoid arthritis and Wegener’s granulomatosis. We are unable to offer an explanation to this rare phenomenon of cavitation in ameloblastoma and the matter is left open to the medical fraternity.

Management of lung metastasis is disappointing and the various treatment options available are surgery, chemotherapy and radiotherapy. Surgical removable of operable lesions perhaps often the best treatment especially for peripheral lung metastasis. The optimal surgical approach is controversial, the various options are cryotherapy, enucleation, curettage, electrocautery, wide local excision and marsupialization. The role of chemotherapy and radiotherapy is not well defined. Response to radiotherapy is unpredictable and it is usually used as a palliative modality for patients with inoperable metastatic deposits. While chemotherapy, especially combination chemotherapy had showed reduction in tumor size in limited reports, but in most no improvement was noticed.

In conclusion, we would like to emphasize two observations, first is ameloblastoma should be considered as on the differential diagnosis in patients with pulmonary cavitatation and secondly these patients should be kept in close follow-up so that lung dissemination can be detected early and the current treatment of choice (surgery) can be initiated.

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