



## Case of Metastatic Spinal Disease with Unusual Primary

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**Abstract**

*We present the case of a 60-year-old lady admitted with lower back pain subsequently diagnosed as metastatic spine disease. Although the patient had a background history of breast cancer and basal cell carcinoma of the skin in the past, a thorough search for primary revealed an unusual cause for her metastatic disease. A review of the patient's known primary sites was negative but interestingly had an oral ulcer that was histologically confirmed to be small cell carcinoma. It is easy to assume the metastasis was from the breast. However, a thorough history and focussed examination of the oral cavity helped us pick up the oral ulcer, which turned out to be small cell carcinoma, a rare cause for metastasis and spinal spread. The case highlights the importance of oral examination in a patient with metastatic disease and consideration of unusual primary.*

**Background**

Studies have shown that the spine is the third most common metastatic site. In up to 15 to 20% of patients, a primary is not identified (1). The most common primary sites include the lungs, breast, prostate, kidneys and haematological system. Identifying a primary is essential to tailor treatment, which may influence the prognosis and survival. Current guidelines do not prescribe direct oral examination for metastasis of unknown primary disease (2).

Small cell carcinomas (SCC) are among the most aggressive forms of neuroendocrine carcinoma (3). They are most commonly seen in the lungs but can originate from the cervix, bladder, prostate and gastrointestinal tract. They are rare in the head and neck region, most commonly from the larynx, salivary glands and Sino-nasal region (4). Database studies demonstrate that only 0.3% of head and neck cancers are histologically small cells carcinomas.

**Case Presentation**

A 65-year-old female was admitted with a history of sudden onset lower back pain of three weeks duration and constipation. There were no symptoms suggesting cauda equina syndrome. The patient complained of intra-oral pain from the right side of her maxilla, severe enough to prevent her from wearing her dentures. The patient denied dysgeusia, intra-oral bleeding, dysphagia, otalgia, weight loss or night sweats.

Past medical history included severe eosinophilic asthma. The patient was under the regional severe asthma team, having been commenced on mepolizumab one year previously with good results. The patient also had type 2 diabetes mellitus, CKD 3, hiatus hernia, colonic polyps, osteoporosis, and basal cell carcinoma. In 2018, the patient was diagnosed with screen-detected right breast cancer (grade 1 cribriform ductal carcinoma), subsequently managed with tamoxifen. The patient was a lifelong non-smoker, drank alcohol rarely with no family history of cancer.

Neurological examination revealed loss of power in both legs. There was a slight facial swelling over the right side of the maxilla, and a right-sided neck lymph node was palpable at level III. Intra-orally, a raised erythematous lesion with rolled edges was noted, overlying the right maxillary alveolar ridge and extending to involve the buccal sulcus and palatal mucosa, overall measuring 4cm by 3cm. The patient was partially dentate, with only her upper and lower anterior teeth remaining.

### **Investigations**

Routine blood results were unremarkable. MRI of the spine revealed multiple diffuse bony metastases with no evidence of cord compression. An orthopantomogram revealed no dent alveolar pathology. The maxillofacial team carried out an incisional biopsy of the intra-oral lesion. Histological examination confirmed a small cell carcinoma with malignant cells showing strong positivity for synaptophysin and p63. They were negative for chromogranin, GATA3, CD3, CD5 and CD20. **(Fig 1 and 2)**

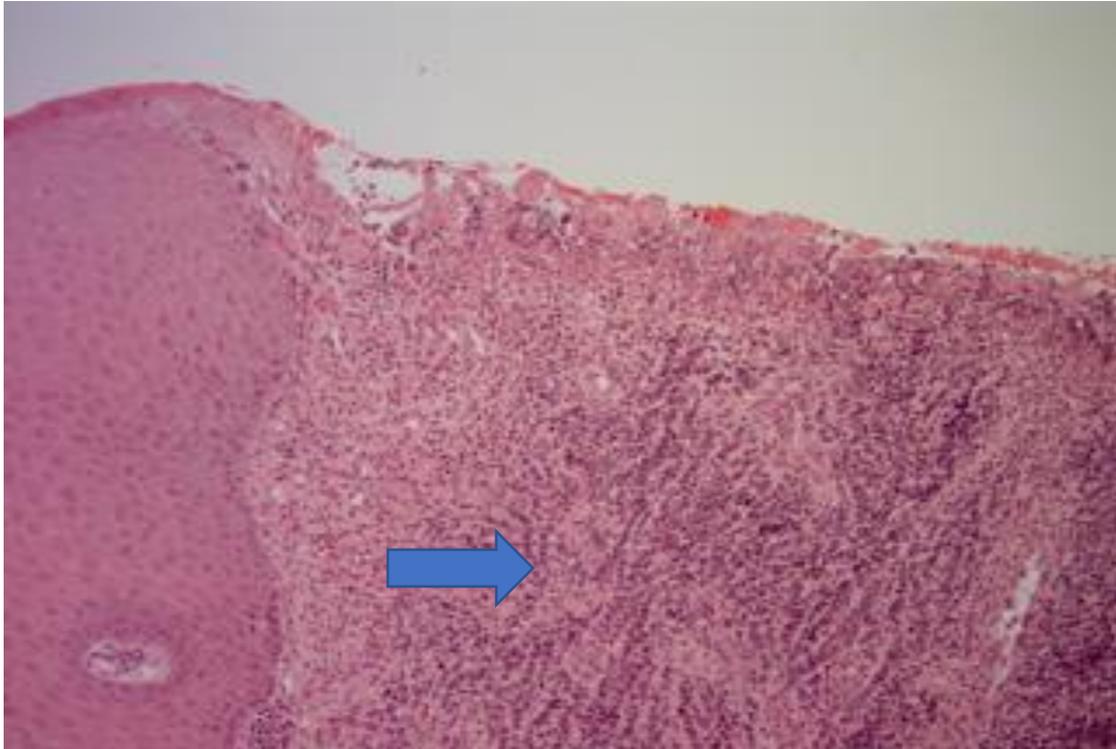
MRI of the head & neck revealed a malignant mass of the right maxilla, centred in the alveolar mucosa extending into the subcutaneous tissue of the right cheek and oral cavity and breaching the anterior and inferior bony wall of the maxillary sinus with right level III lymphadenopathy, staged at T3N1MX.

CT staging showed no pulmonary pathology, but extensive metastatic infiltration of the liver and left proximal humerus was identified.

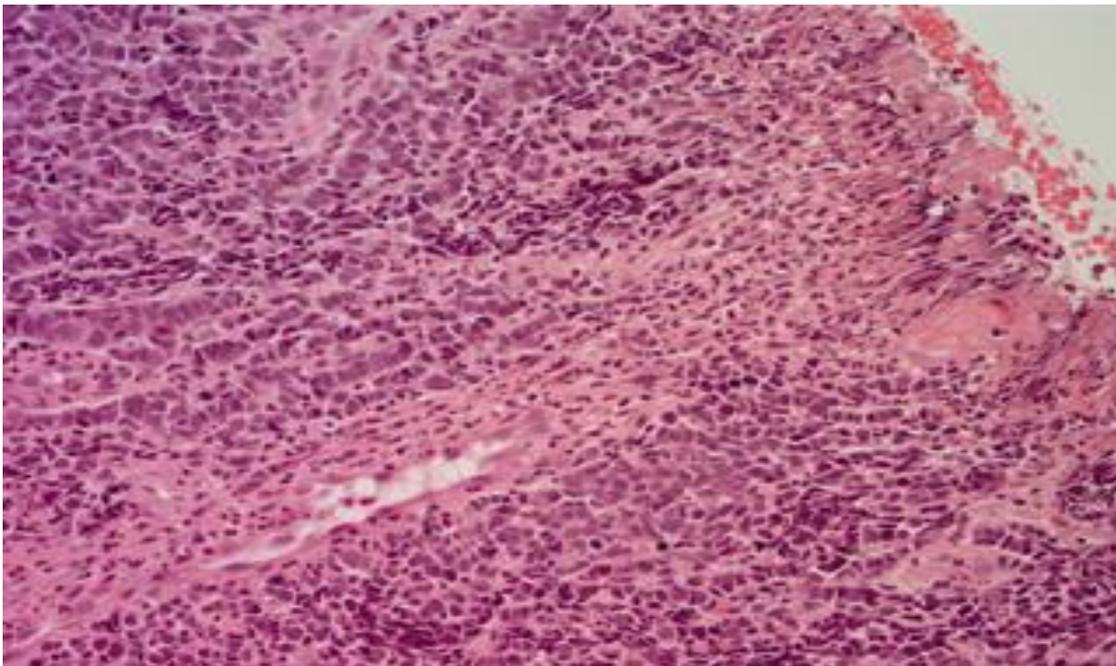
### **Fig 1 section of the cheek biopsy Fig 2 HP section of the cheek biopsy**

### **Outcome**

The patient was discussed at the regional head and neck MDT, where it was felt that the tumour was extensively disseminated for treatment with curative intent. Since the patient had a WHO score of 4 and multiple comorbid conditions, MDT recommended the best supportive care. Unfortunately, the patient passed away three weeks later with palliative team support.



**Figure 1** Section of The Cheek Biopsy



**Figure 2** HP Section of The Cheek Biops

## Discussion

Identifying primary in a case of metastatic spine disease can be tricky and requires a detailed history, systematic review and examination. Investigation for identifying a primary can be exhaustive. Our junior doctors' thorough examination and history taking picked up this lady's diagnosis. Ours is one of the first cases of metastatic spinal disease arising from an oral small cell carcinoma reported in Western literature to the best of our knowledge. Previous reports and series (5,6,7) do not feature any spinal or liver metastatic disease, and we believe this case is unique. Current literature quotes only two other small cell cancer of the cheek and tonsils in World literature (8, 9)

The patient was investigated for a primary lung or the breast tumour's re-occurrence. The lung, breast, head and neck MDTs all concluded that the metastases were likely derived from the small, oral cell cancer. Since the patient was not fit for active oncological therapy, a further biopsy of the metastatic disease was not merited to confirm the diagnosis. The lack of biopsy of the metastasis is the only limitation of our report. However, the concerted opinion of the MDTs and the absence of any evidence of the recurrence of breast tumour or primary lung cancer makes the metastasis likely originating from an oral small cell lesion.

## Conclusion

This case adds to the literature that Oral small cell carcinoma can present as distant spinal metastases. The report highlights the importance of oral examination in metastatic spine disease.

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