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Pathology in Focus

Hyoid bone tumour mass presenting with cervical nodal metastasis

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Abstract
This paper presents a quite unique case report of a patient presenting with the combination of cervical metastatic lymphadenopathy and a hyoid bone tumour mass. The differential diagnosis and treatment is discussed, with emphasis on the importance of immunohistochemistry and electron microscopy in the management of such a case.

Key words: Hyoid bone; Neoplasm metastasis, unknown primary; Lymph nodes; Neck

Case report

A 60-year-old man presented with a painless left lower cervical mass which had been noted (accidentally) one month prior to referral. He was otherwise well with no significant past medical history, and he had been a non-smoker for 30 years. Examination confirmed a left anteroinferior mobile cervical node, 3 cm in diameter. The rest of the head and neck examination was normal. In particular, there were no other palpable nodes in the neck, axillae or inguinal regions, no hepatosplenomegaly or skin lesions present.

Initial investigations including a routine blood screen and chest X-ray were normal: however, a fine needle aspirate of the neck lump suggested a poorly-differentiated malignancy. The patient underwent a normal panendoscopy, followed by an excision biopsy of the cervical node. Histology confirmed the diagnosis of a poorly-differentiated neoplasm, showing sheets of large pleomorphic cells (Figure 1), containing large irregular nuclei and prominent eosinophilic nucleoli. There was a moderately high mitotic rate and focal areas of necrosis. Immunohistochemical analysis demonstrated slight focal staining for epithelial membrane antigen. The malignant cells were positive for vimentin but negative for high and low molecular weight keratin. Anti-thryoglobulin antibody titres were performed and were not raised. The tumour was negative for common leucocyte antigen, L26, carcinoembryonic antigen, α-fetoprotein, α-1-antitrypsin, lysozyme, MAC387, neuron specific enolase, chromogranin, S-100 protein and placental alkaline phosphatase. Ultrastructural analysis showed basement membrane material and intercellular junctions compatible with epithelial differentiation (Figure 2). No evidence of glandular formation was apparent. Overall, pathological assessment failed to show specific features suggestive of the primary site of the tumour, but supported the diagnosis of metastatic carcinoma.

Further investigations to determine the primary site of the tumour included computed tomography (CT) scanning of the head and neck, thorax, abdomen and pelvis. In addition an abdominal ultrasound, barium swallow and small bowel follow-through X-rays were carried out. All of these investigations were normal apart from the surprising finding of a soft tissue mass eroding and expanding the hyoid bone (Figure 3), which was not apparent clinically.

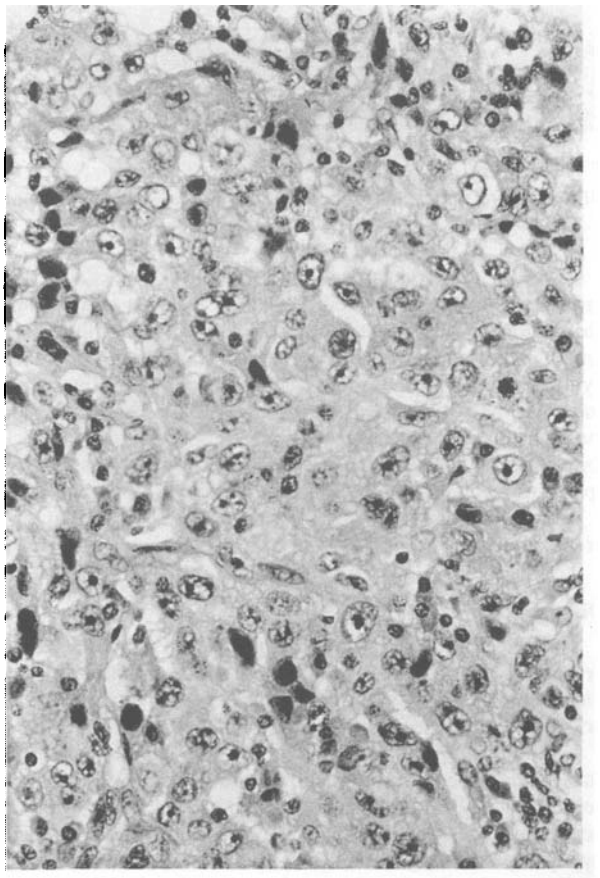

FIG. 1
Showing a poorly-differentiated neoplasm, comprised of pleomorphic cells containing large irregular nuclei.

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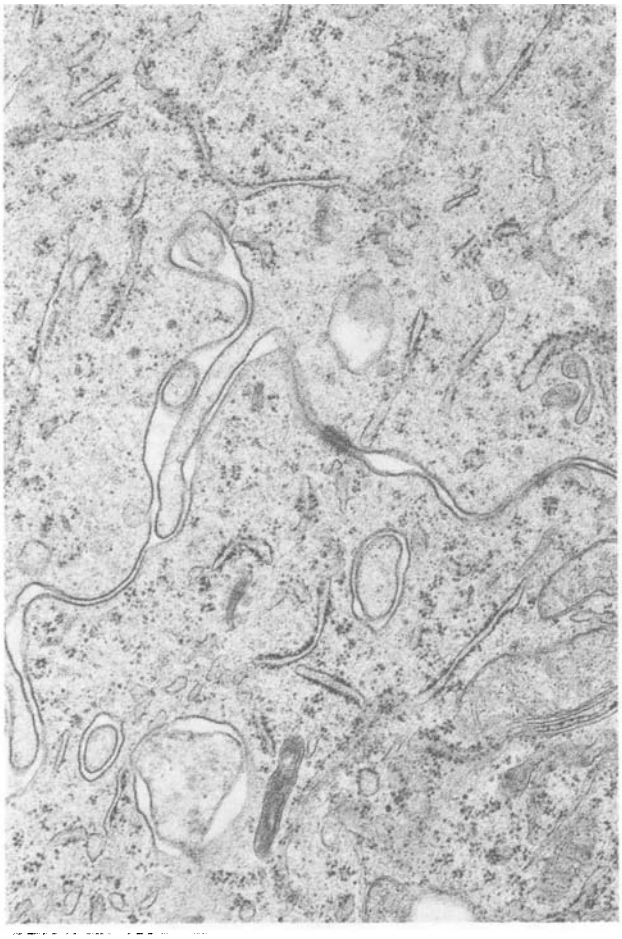

FIG. 2
Ultrastructural appearance showing intercellular junction formation compatible with epithelial differentiation. (× 26, 600).

At neck exploration the presence of a hyoid mass was confirmed and excised. Histological and electron microscopic characteristics were identical to that obtained from the cervical lymph node, and showed malignant cells infiltrating through the hyoid bone and adjacent soft tissue. After multidisciplinary consultation, the patient was referred for external beam radiation, the fields including the nodal regions of both sides of the neck, together with the tonsils, and tongue base. The nasopharynx was spared to lessen radiation-induced xerostomia.

Close follow-up of the patient has shown no evidence of local or distant recurrence three years after diagnosis.

Discussion

This paper presents a quite unique case report of a patient presenting with cervical nodal metastases from an undifferentiated tumour, which on further investigation was found to either originate from or secondarily involve the hyoid bone.

In patients presenting with high or mid-neck cervical metastatic nodal disease, more than two-thirds will have squamous cell carcinoma, originating from primary sites in the head and neck, particularly the nasopharynx, tongue base, tonsil and hypopharynx (Wang *et al.*, 1990). Nodal metastases in the low neck may arise from a primary site below the clavicle, particularly from the lung, stomach, pancreas or kidney (Didolkar *et al.*, 1977), and are most often adenocarcinomas. Other diagnoses to consider include neoplasia originating in the lymphoreticular system and mesenchymal tumours. Management of these patients includes a comprehensive examination followed by fine needle aspiration, and if cytological diagnosis is unhelpful, triple endoscopy proceeding to excision biopsy.

When a poorly-differentiated tumour is diagnosed, as in this case, a comprehensive pathological examination is crucial, and must include immunohistochemistry and electron microscopy evaluation. For our patient, in spite of the negative-staining reaction for keratin and the apparent anomalous staining for vimentin, which is characteristically expressed by mesenchymal neoplasms, a diagnosis of metastatic poorly-differentiated carcinoma was favoured. Coexpression of keratin and vimentin in epithelial neoplasms, or sole expression of vimentin in poorly-differentiated epithelial tumours, is now well recognized (Azumi and Battifora, 1987). This preference was based on the light microscopic appearance of the malignancy, its presence in a lymph node, the positive-staining reaction for epithelial membrane antigen, and the ultrastructural findings.

This case report is unique however because, in addition to the cervical metastatic disease, our patient also had a focus of carcinoma infiltrating his hyoid bone. Very little information exists in the literature relating to a tumour arising from, or involving, the hyoid bone.

When bone is invaded by malignancy, possibilities entertained include local or distant spread from the primary neoplastic focus. In the former case, the possibility of a tumour arising from local structures such as the thyroid gland, parathyroid gland or thyroglossal duct remnant has to be considered. Other potential sites include a primary gastro-respiratory tract tumour or tumour arising from epithelial tissue within the hyoid bone. The only paper published so far dealing with carcinomatous involvement of the hyoid bone describes 11 patients with clinically advanced squamous cell carcinomas invading the hyoid bone (Timon *et al.*, 1992). Importantly, all 11 cases originated from the larynx, pyriform fossa, vallecula or tongue base; in contrast these potential sites were free of neoplasm on examination and endoscopy in the present case report. Also, neither clinical, histological or radiological evidence of a thyroid or parathyroid neoplasm was detected.

Distant bony metastases from a head and neck squamous cell carcinoma is a rare phenomenon in the clinical setting. However,

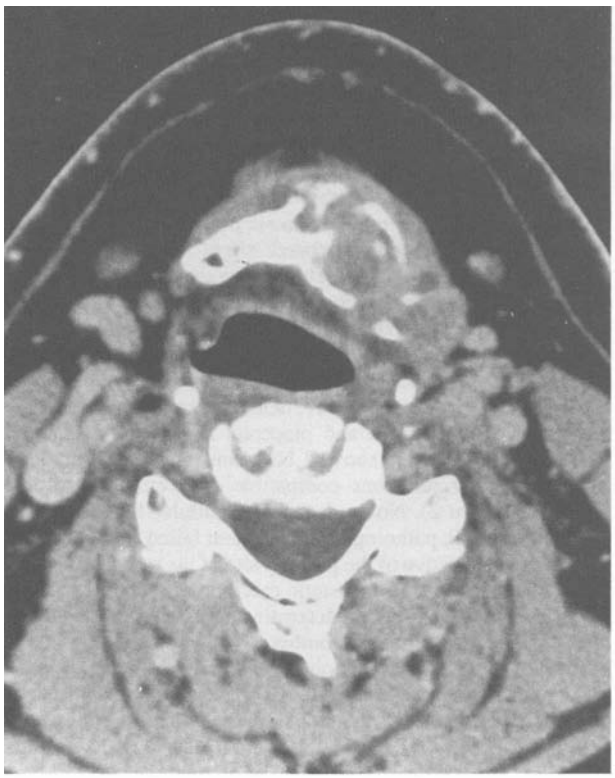

FIG. 3
Axial computed tomography (CT) of the hyoid bone showing a soft tissue mass eroding and expanding the hyoid bone.

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occult bony metastases are found at autopsy in approximately 20 to 25 per cent of patients with advanced disease (Dennington *et al.*, 1980). Despite this, there is no report in the literature of squamous cell carcinoma resulting in distant hyoid bone metastases. As regards as infraclavicular source of bone metastases, the most likely candidate is lung cancer, with other possible sources including breast and prostatic primaries (Didolkar *et al.*, 1977; Nottebaert *et al.*, 1989).

Metastases tend to spread to skeletal parts with active red bone marrow, such as the vertebrae. The body of the hyoid contains active marrow in adults, although in small amounts, and therefore metastases theoretically may appear here. To date, only Jacobsson and Bergstedt (1988) have reported a hyoid bone metastasis. This was described as a 'hot' spot in the hyoid bone on bone scanning, interpreted as a bony metastasis in a patient with breast carcinoma and generalized skeletal metastases. In contrast, in our patient, tumour involvement was confined to a cervical node and the hyoid bone.

Our patient was treated, on an empirical basis, as a poorly-differentiated squamous cell carcinoma arising from the head and neck, which carries a relatively good prognosis with radiotherapy and/or surgery (Bataini *et al.*, 1987; Wang *et al.*, 1990). This policy appears to have been justified, since close follow-up of the patient has shown no evidence of recurrence or of another primary tumour over a three-year period.

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