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Clinical Record

Interdigitating dendritic cell sarcoma of the parotid gland

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Abstract

Interdigitating dendritic cell sarcomas (IDCSs) are extremely uncommon tumours that arise predominantly in lymphoid tissue. We report a case of an IDCS arising in the parotid gland of a 73-year-old man. Clinically, a primary salivary gland tumour was suspected but fine needle aspiration cytology suggested a soft tissue tumour. A diagnosis of IDCS was made on histopathological examination of the resection specimen, with subsequent confirmation by electron microscopy. Given the extreme rarity of this tumour at this site, it is unlikely to be a common diagnostic problem, but the importance of multiple diagnostic modalities is emphasized. The findings of cytology, histology, immunohistochemistry and electron microscopy have not previously been described together in a single case report of this tumour.

Key words: Parotid Neoplasms; Sarcoma; Dendritic Cells

Introduction

Interdigitating dendritic cell sarcomas (IDCS) are rare neoplasms arising from interdigitating dendritic cells. These cells are normally present in the T-cell areas of the peripheral lymphoid tissue and play an important role in antigen presentation to T-cells. Interdigitating cell sarcomas represent one of the four groups of tumours of dendritic or accessory cell origin in the classification system recently proposed by the International Lymphoma Study Group, based on their immunophenotypic characteristics.[1] These tumours usually arise in lymph nodes, although there are a few reports of tumours arising in extranodal sites such as the testis[2] and spleen.[3] Here, we report a case of an IDCS in the parotid gland with involvement of ipsilateral cervical lymph nodes, in a 73-year-old man.

A PubMed search was used to identify relevant articles published in English, using the terms 'interdigitating dendritic cell sarcoma', 'parotid' and 'reticulum cell sarcoma'.

Case report

A 73-year-old, previously fit gentleman presented with a six-month history of a gradually enlarging lump over the angle of the left mandible. On examination, a 20 × 20 mm, firm, non-tender, mobile lump was noted. It appeared to arise from the parotid gland. There was no evidence of facial nerve weakness and the rest of the patient's upper aerodigestive tract was normal. Pre-operative fine needle aspiration of the lesion revealed scattered single pleomorphic cells with wispy cytoplasm and free nuclei. Immunocytochemistry performed on the cell block showed the cells to stain positive with vimentin and negative with cytokeratins. A diagnosis of soft tissue tumour was suggested and excision advised. A left partial parotidectomy was performed. Peri-operatively, the lower branches of the facial nerve were seen to be involved with the tumour. Furthermore, lymphadenopathy was noticed in the ipsilateral level I, III and V nodes. The operation was therefore extended to include a selective neck dissection. Post-operatively, the patient underwent radical radiotherapy to the left side of the neck. Fifteen months post-operatively, he remained disease-free.

The resection specimen, therefore, comprised the left parotid gland and a selective neck dissection (levels I, II, III and V). A solid, grey-white, 30 × 30 × 70 mm tumour with areas of necrosis was noted on sectioning the parotid gland. Macroscopically, the capsule of the parotid gland appeared intact. Histology showed a tumour which, although circumscribed, had a pushing margin and was present in amongst salivary gland acini. The tumour was composed of pleomorphic, spindle cells arranged in sheets, as well as interlacing fascicles and whorls. The cells possessed large, vesicular nuclei and prominent nucleoli and were mitotically active. An intense neutrophilic infiltrate was a prominent feature throughout the tumour, rather than the usual lymphocytic infiltrate.

Immunohistochemically, the spindle cells were strongly positive with S-100 and vimentin and showed patchy positivity with CD68. The spindle cells were negative for cytokeratin (Cam 5.2 and MNF116), desmin, CD45, CD30, vascular markers (CD31 and CD34), melanoma markers (HMB-45 and melan-A) and CK5/6. Markers for a follicular dendritic cell tumour, such as CD4, CD21, CD35 and CD1a, were all negative.

On electron microscopy, the tumour cells showed lobulations and deep invaginations of the nuclear membrane. The tumour cells, which lacked desmosomal junctions and Birbeck granules, showed numerous cytoplasmic processes on their surface, which interdigitated with those of adjacent tumour cells.

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On the basis of the morphology and immunophenotypical characteristics, a diagnosis of an IDCS was made. Two lymph nodes, representing level I and level III nodes, also showed involvement by a similar tumour, the largest involved lymph node measuring 20 mm in maximum dimension.

Discussion

Interdigitating dendritic cell sarcomas are rare neoplasms, with only 33 cases reported prior to this case. There is only a single other report of an IDCS arising in salivary gland associated lymphoid tissue of the parotid gland.[4] In our case, the tumour was clearly within the parenchyma of the parotid gland, although we surmise that it had an origin in an intraparotid lymph node or lymphoid tissue within the parotid gland. However, no histological evidence of this remains. Two ipsilateral cervical lymph nodes, much smaller in size than the main tumour mass, were involved by the tumour, with the same histological appearances.

On the basis of the morphological and immunocytochemical characteristics of the tumour, as described above, a diagnosis of soft tissue tumour was suggested and excision advised. To the best of our knowledge, this is the first case in which cytology was performed prior to excision and histological diagnosis.

In the resection specimen, the appearance of a poorly differentiated spindle cell lesion raised several differential diagnoses, including undifferentiated spindle cell carcinoma, malignant soft tissue tumours, a myoepithelial tumour, spindle cell melanoma, and tumours of dendritic or accessory cell lineage. Immunocytochemistry helped exclude the first four, while the patchy positivity with CD68 was suggestive of the last. However, the cells were negative for the markers of follicular dendritic cell tumour – CD21, CD35, CD4 and CD1a. Therefore, a diagnosis of an IDCS was made by exclusion and subsequently confirmed by electron microscopy (see Figures 1 and 2), which was entirely in keeping with the ultrastructural appearances of this tumour described in previous reports.[5–8]

These tumours behave in a variable fashion clinically, ranging from widespread metastasis and early death in some patients[9] to a more benign course in others. The patient presented here remained disease-free after 15 months of follow up.

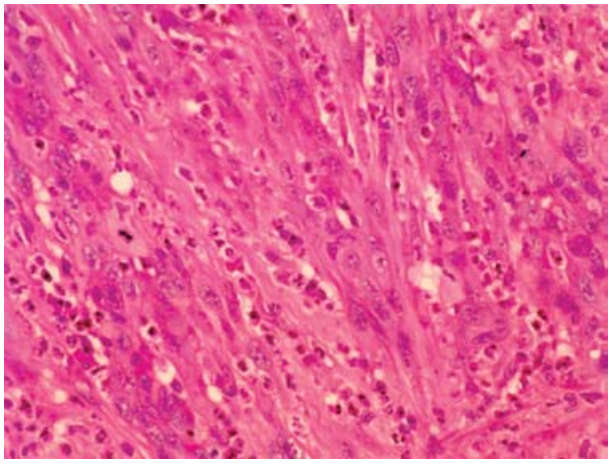

FIG. 1

The tumour comprises pleomorphic spindled cells arranged in fascicles with a prominent infiltrate of neutrophils. A mitotic figure is also seen (H&E; ×400).

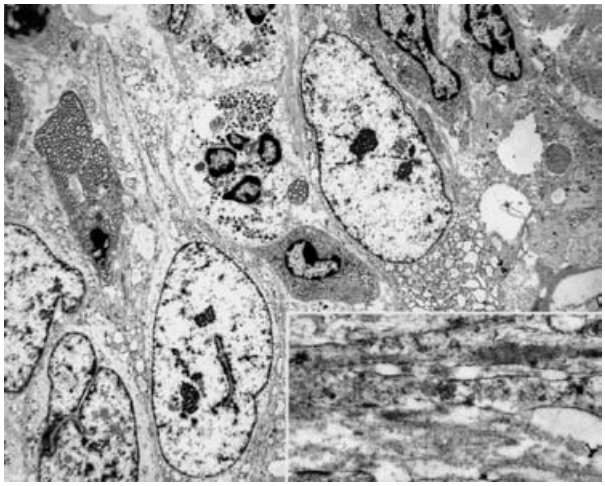

FIG. 2

Electron micrograph (EM) of the tumour; the tumour cells have large, lobulated nuclei. Inflammatory cells, including neutrophils, are seen in amongst the tumour cells (EM; ×3300). Inset: slender, interdigitating cytoplasmic filaments are evident on electron microscopy (EM; ×31 000).

Conclusion

This report presents an IDCS arising in the parotid gland, an extremely rare site. This is the only report to discuss the cytological, histological and electron microscopic appearances of this uncommon neoplasm.

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- **Interdigitating dendritic cell sarcomas are extremely uncommon tumours that predominantly arise in lymphoid tissue**
- **This paper describes an interdigitating dendritic cell sarcoma within the parotid gland**
- **The clinical, cytological, histological, immunohistochemical and ultrastructural findings have not previously been reported on a single case**

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