CASE REPORT

A rare case of late solitary vertebral metastasis from an adenoid cystic carcinoma of the lacrimal gland

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SUMMARY

Adenoid cystic carcinoma of the lacrimal gland is one among the common malignancies affecting the lacrimal gland. However, overall, it is a rare condition. It has a rather poor prognosis with local recurrence and distant haematological metastasis which are invariably multiple. We present a rare case of a 51-year-old woman who presented with localised lower thoracic pain with collapse of the T10 vertebral body, which turned out to be a solitary late metastasis from her previously treated lacrimal gland tumour.

BACKGROUND

Adenoid cystic carcinoma (ACC) of the lacrimal gland is the the most common non-lymphoid malignancy of the lacrimal gland,¹ constituting 13.4% of lacrimal gland cancers² and about 1% of all head and neck malignancies.³ They are tumours which have a propensity to spread locally and to metastasise via the haematogenous route to the lungs, brain and bone in a decreasing order.^{4–8} Metastases are usually widespread and multiple.^{9–10}

We report the case of a 51-year-old woman who presented with a solitary vertebral metastasis 4 years after initial treatment of an ACC of her lacrimal gland.

CASE PRESENTATION

A 51-year-old woman was referred to the spinal surgery department of Southampton General Hospital in January 2015 with a history of vague back pain for about 3 months associated with progressive difficulty in walking and mobilising. She had previously been diagnosed with an ACC of the lacrimal gland of the left orbit which had been

Figure 2 T2 sagittal MRI.

debulked in September 2011 at the Moorfields Eye Hospital, London. She subsequently received radiotherapy with a total dose of 60 Gy in 33 fractions over a period of 6½ weeks which was tolerated well apart from some orbital swelling. She has had no local recurrence since. On examination, she had normal tone, power and sphincter control. She had reduced sensation to pin prick below T10 dermatome and an up-going plantar response bilaterally. There was no clinical evidence of recurrence of tumour in her orbit.

INVESTIGATIONS

She was investigated with a CT of the chest, abdomen and pelvis and an MRI scan of the whole neural axis which showed isolated involvement and collapse of T10 vertebral body (figures 1–3). There was evidence of abnormal soft tissue replacing the T10 vertebral body, infiltrating the pedicles, transverse processes and spinous process of T10, and early involvement of the adjacent T9 pedicles and T11 body. There was extension into the spinal

Figure 1 T1 sagittal MRI.

Figure 3 T2 axial MRI.

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Figure 4 Biopsy of orbital tumour showing features of adenoid cystic carcinoma (H&E x20 objective).

canal causing concentric spinal canal stenosis and evidence of cord compression. There was no evidence of metastatic spread elsewhere. She had a Spinal Instability Neoplastic Score of 16 suggesting significant instability requiring consideration of stabilisation. Her prognostic revised Tokuhashi Score was 11.

She subsequently underwent a CT-guided biopsy. Histology showed metastatic carcinoma with well-circumscribed islands of cells and features consistent with adenoid cystic morphology. The tumour cells were immunoreactive for cytokeratin (MNF116+, CK7+, CK20–) confirming their epithelial nature and were TTF-1 negative (figures 4–5). The histology from the orbital tumour was reviewed, and a primary lacrimal gland adenoid cystic carcinoma was confirmed which had essentially identical histological features to the spinal metastasis.

DIFFERENTIAL DIAGNOSIS

Treatment

The patient underwent decompression at T10 and posterior stabilisation of T8–L1 (figures 6–7) and was subsequently mobilised fully weight bearing. The immediate postoperative period was uneventful, and she went on to receive radiotherapy (30 Gy in 10 treatments over 2 weeks).

Figure 5 Biopsy of spinal lesion showing similar adenoid cystic morphology (H&E x20 objective).

Figure 6 Postoperative anteroposterior radiograph following instrumented stabilisation.

Figure 7 Postoperative lateral radiograph following instrumented stabilisation.

OUTCOME AND FOLLOW-UP

At the 6-month follow-up, she was mobilising independently, and there was no evidence of recurrence of the disease. At further follow-up in 2016, over a year after her treatment, she continued to do well and did not show any evidence of recurrence at either site.

DISCUSSION

ACC of the lacrimal gland is a rare tumour constituting about 1% of all head and neck malignancies³ and tending to have a poor prognosis overall.¹¹ In general, ACCs are slowly growing tumours with local recurrence in 3.5 years (mean) and a mean survival of 5 years.¹² Gamel and Font⁴ found a 5-year survival of 21% for a basaloid variety of ACC and 71% for a non-basaloid ACC.

Solitary metastasis of ACC is rare. There have been a few reported cases of isolated metastasis to the liver¹³ and kidney¹⁴; however, we could have identified only one other reported case of solitary vertebral metastasis of an ACC of the lacrimal gland.¹⁵

This case highlights the critical importance of thoroughly investigating a patient with atypical back pain who has a history of malignancy, even if it is a slow-growing tumour with low tendency to metastasise.

Contributors All persons who meet authorship criteria are listed as authors, and all authors certify that they have participated sufficiently in the work to take public responsibility for the content, including participation in the concept, design, analysis, writing or revision of the manuscript. AA is the lead author and contributed to management of patient, information gathering, writing of the draft and analysis of

Learning points

- We would like to highlight and reiterate the importance of thorough clinical evaluation and early investigations in patients, presenting with atypical back pain with a history of any malignancy, even when it may be a slow-growing tumour with low risk of metastasis.
- Metastasis from lacrimal gland tumours could be solitary, and early diagnosis and intervention could potentially offer better outcomes.

data. SK-RG is the corresponding author and contributed to the review and revision of draft, analysis and input of data and coordination of submission to the journal. SH contributed to writing of draft, analysis and referencing. JN is the senior supervisor providing guidance through the process and involved with the care of patient, providing histopathological input and pictures of specimen analysis.

Competing interests None declared.

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