CASE REPORT

Colonic metastasis in mucoepidermoid carcinoma of the parotid: a rare occurrence

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SUMMARY

We present a case of intermediate-grade mucoepidermoid carcinoma of the parotid with late local recurrence and colonic metastasis. A 69-year-old man who had undergone right total conservative parotidectomy followed by adjuvant radiotherapy for intermediate-grade mucoepidermoid carcinoma 10 years prior, presented with a recurrent swelling in the postoperative site and cardiac failure. On evaluation, he was found to have severe anaemia with positive stool occult blood. Colonoscopic evaluation revealed a globular submucosal bulge with erosion 40 cm from the anal verge, the biopsy of which was consistent with mucoepidermoid carcinoma. The presentation, diagnostic details and management of this rare case are discussed.

BACKGROUND

Malignant salivary gland tumours account for less than 5% of all head and neck malignancies, and mucoepidermoid carcinoma (MEC) constitutes 35% of all salivary gland malignancies.¹ MEC, in particular, exhibits a wide spectrum of behaviour: high-grade tumours display aggressive behaviour and those of low grade usually have a benign nature with good prognosis. The most common site of origin is the parotid gland.² Distant metastases occur most frequently in the lung, bone and liver.³ A single institutional report on MEC of major and minor salivary glands showed a 5-year overall survival rate of 62.3%, and a 5-year relapse-free survival rate of 57.2%. Local recurrence was found in 6 of the 43 patients and regional lymph node recurrence without distant metastasis in four. Distant metastasis to the lung was present in five patients, to the liver in two patients and to the brain in one patient.²

CASE PRESENTATION

A 59-year-old man presented with a painless progressive firm 5×5 cm swelling in the region of the right parotid, lifting the right ear lobe, and fixed and deep to the parotid fascia, with no skin involvement. There were no palpable neck nodes. Fine-needle aspiration cytology revealed atypical features. The patient was planned for surgical excision of the lesion. Intraoperatively, the tumour was found at the upper pole of the parotid, infiltrating the underlying muscle, part of the temporomandibular joint capsule and the upper trunk of the facial nerve up to the facial trunk. He underwent total right conservative parotidectomy with cable grafting of the zygomatic branch of the facial nerve and right lateral tarsorrhaphy in October 2005. He

then received adjuvant radiotherapy, using 6 MV photons to the postoperative bed, 66 Gy in 33 fractions by three-dimensional Conformal radiotherapy technique, which was completed in January 2006. The ipsilateral anterior neck was treated to a dose of 46 Gy in 23 fractions. The patient had been on regular follow-up but almost 10 years after the initial diagnosis and treatment, in April 2015, he presented with recurrent swelling in the postoperative site in the right parotid region, along with anaemia and cardiac failure.

INVESTIGATIONS

Blood investigations revealed severe anaemia (haemoglobin 3.5 g%) and positive stool occult blood. Evaluation with colonoscopy revealed a globular 3×2 cm submucosal bulge with erosion 40 cm from the anal verge. The biopsy from the colonic lesion was consistent with MEC. It revealed colonic mucosa with submucosa infiltrated by a tumour, composed of small and large nests, and sheets of medium sized cells with round to oval nuclei with fine chromatin, occasional visible nucleoli and scant to moderate eosinophilic cytoplasm. There were a few larger cells with abundant vacuolated cytoplasm containing mucin (periodic acid–Schiff diastase-positive). The mitotic activity was inconspicuous. On immunohistochemistry, the tumour cells were positive for CK 5/6 and negative for neuroendocrine markers. The MIB 1 proliferation index is <1% (figures 1–3). A whole body positron emission tomography CT revealed abnormal fluorodeoxyglucose uptake in the right mastoid region (standardized uptake value (SUV) 4.31), right parotid region (SUV 12.34) and left adrenal (SUV 5.66), with corresponding CT images suggestive of local recurrence and nodular thickening of adrenals, likely metastasis. Biopsy from the recurrent swelling at the postoperative site was consistent with recurrent MEC.

DIFFERENTIAL DIAGNOSIS

On follow-up, when the patient presented with cardiac failure secondary to anaemia, he was evaluated for all the common causes of anaemia including stool occult blood, which was positive, hence he underwent colonoscopy, revealing a submucosal lesion, which, later, on histopathological examination, proved to be metastasis.

TREATMENT

The patient was discussed in the multidisciplinary tumour board and in view of his advanced age, the local recurrence and distant metastasis, he was

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Figure 1 H&E $\times 10$ and H&E $\times 40$: colonic epithelium infiltrated by tumour composed of sheets and nests of medium sized cells with round to oval nuclei with fine chromatin, visible nucleoli and scant to moderate eosinophilic cytoplasm.

offered and treated with three cycles of palliative chemotherapy with cisplatin and paclitaxel. Following the chemotherapy, he presented with pain, ulceration, discharge and bleeding from the swelling in the right parotid region. With the intent to palliate the local symptoms, he was given re-irradiation to the local recurrence, using 9 MeV electrons, 30 Gy to 85% isodose in 10 fractions.

OUTCOME AND FOLLOW-UP

The patient maintained haemoglobin levels and did not have symptoms related to anaemia, following the chemotherapy. With the re-irradiation, there was reduction in pain, discharge and bleeding from the local recurrence. He was advised metronomic chemotherapy with oral methotrexate.

DISCUSSION

Stewart *et al*⁴ first described MECs in 1945. According to the Armed Forces Institute of Pathology grading, they are classified into three histological grades (low, intermediate or high), based on presence of intracystic components, neural invasion, necrosis, anaplasia and level of mitotic activity. The clinical behaviour

Figure 2 CYTOKERATIN 5/6, $\times 20$: these tumour cells are positive for CK 5/6.

Figure 3 PASD $\times 40$: a few intermixed occasional larger cells with abundant eosinophilic to vacuolated cytoplasm with PASD-positive mucin. PASD, periodic acid–Schiff diastase.

and prognosis correlate with the histological grading.⁵ Metastases in MECs occur by lymphatic or haematogenous spread. Metastases from MEC of the parotid are commonly found in the lung, bone and liver.³ There are a few reports of MECs of lung with metastasis to the bone, skeletal muscle, pericardium, kidney and adrenal.^{6–7} There is a single report of MEC of lung with symptomatic colonic metastasis at presentation.⁸ Ileal metastasis from parotid carcinoma expleomorphic adenoma, presenting with ileal perforation, has been described.⁹ MEC of the parotid with distant metastasis to the colon has never been reported, and this case report highlights, for the first time, the extreme rarity of the condition.

Advanced histological grade is an independent predictor for disease free survival and overall survival for patients with MEC in the head and neck region.⁵ There are conflicting reports, mostly from single institutions, on the prognosis of intermediate-grade MEC, with some studies indicating that they behave more as low-grade MEC,¹⁰ while others suggest that prognosis is similar to that of high-grade MEC.¹¹ Kokemueller *et al*¹² reported that grade, stage and margin status affected prognosis, however, the Mayo Clinic experience found these factors to be less important.¹³

The incidence of regional and distant metastasis in MECs range between 3–16% and 6–15%, respectively.¹⁴ Metastases are found more frequently with high-grade tumours.⁹ Metastasis to the colon in this patient had likely occurred via the blood stream. Clinical findings in such a case can be varied, as in any colonic malignancy, and this patient presented with cardiac failure secondary to severe anaemia. Histology remains the gold standard and once it is confirmed the treatment intent is palliative. This patient was initially transfused blood to correct anaemia, and then had palliative chemotherapy with cisplatin and paclitaxel. He did not have further bleeding or anaemia. Distant metastases often have poor prognosis with median survival ranging between 4.3 and 7.3 months.⁹ Single agents or a combination thereof in a palliative setting include cisplatin, carboplatin, etoposide, doxorubicin, cyclophosphamide, ifosfamide, fluorouracil and paclitaxel.¹⁴ Distant metastasis can occur after a considerable time period from initial diagnosis, and hence it is recommended that follow-up continue once every 12 months for life.¹⁵

Learning points

- ▶ Colonic metastasis from mucoepidermoid carcinoma of the parotid is extremely rare.
- ▶ Late metastasis should be considered when there are new symptoms on follow-up, and appropriate specific investigations should be undertaken.
- ▶ In view of likelihood of late recurrence and metastasis, lifelong follow-up is necessary.
- ▶ Distant metastases carry poor prognosis and treatment intent is palliative.

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Competing interests None declared.

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