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Case Report

Carcinoid Tumour of the Parotid Gland: Conservative Treatment

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Abstract

Carcinoid tumours are a unique and relatively uncommon subset of neuroendocrine tumours. We present a 70-year-old female with end stage chronic obstructive pulmonary disease with a left parotid gland lump associated with facial discomfort. She was diagnosed with bronchial carcinoid tumour a year prior to this presentation. The parotid lump was confirmed to be a carcinoid tumour of the parotid gland on cytology. This unique case is presented with discussion on the management of this condition together with literature review.

Keywords: Carcinoid Tumour; Parotid Gland; Bronchial Carcinoid Tumour; Management

Abbreviations

FNAC: Fine Needle Aspiration Cytology; MRI: Magnetic Resonance Imaging; CT: Computed Tomography; PET-CT: Positron Emission Tomography-Computed Tomography; PC: Pulmonary Carcinoid; ICC: Immunocytochemistry; CD56: Neural Cell Adhesion Molecule

Introduction

Carcinoid tumours are a unique and relatively uncommon subset of neuroendocrine tumours. Common sites of origin for carcinoid tumour are gastrointestinal and respiratory tract [1,2]. Primary carcinoid tumour in the parotid gland is extremely rare and most commonly represent a metastatic deposit [3]. There are two main types of carcinoid tumours: atypical which behave aggressively while the typical variant is benign. The proposed general treatment consensus for any parotid tumour, including primary carcinoid tumour, is surgical excision.

We present a case of a carcinoid tumour in the parotid gland, discussion of its management and literature review of this unique pathology.

Case Report

A 70-year-old Caucasian female, life-long smoker with end stage chronic obstructive pulmonary disease, presented with a lump in her left parotid gland along with some facial discomfort. She proceeded to have an ultrasound scan of her parotid gland and ultrasound guided fine needle aspiration cytology (FNAC) which confirmed a solid mass in her left parotid gland. The cytological findings were in keeping with atypical carcinoid tumour. A year prior to this, she had undergone a bronchoscopy examination which revealed an abnormal tissue observed at the origin of her left upper and lower lobes. The biopsy from this had revealed carcinoid tumour.

As part of the workup to stage the patient's disease, she underwent an MRI scan of her parotid glands (Figure 1), CT chest, and a full body PET-CT scan. Due to her significant co-morbidities, she

was found to be unfit for any surgical intervention for her parotid atypical carcinoid tumour. The decision to treat her conservatively was made by the multidisciplinary team and following discussion with patient. At 18 months, she remained clinically stable with supportive care.

Figure 1: MRI scan of the left parotid gland showing a mass measuring 22 x 23 x 22 mm in the inferior aspect of the deep lobe of the parotid gland.

Discussion

Salivary gland tumors account for 3 - 6% of head and neck adult neoplasms and 70-85% are within the parotid gland [3]. Carcinoid tumour is a distinct and relatively uncommon subset of neuroendocrine tumors but this itself represents a wide spectrum of neoplasms with vast array of secretory products [2]. It rarely occurs outside the gastrointestinal tract but has been reported in the larynx, middle ear and pancreas [2]. For all sites, age-adjusted incidence rate was 4.48 per 100,000 population per year; highest in black males though carcinoids in the lung is less prevalent among the black population than expected and an overall female predominance [2]. Approximately 10% of these tumours secrete bioactive mediators that may engender various characteristics of carcinoid syndrome [4]. In most cases, a parotid lesion is assumed to have been a metastatic tumour from other sites rather than a primary carcinoid.

Diagnostic investigations include physical examination, CT and fine needle aspiration cytology (FNAC). The overall sensitivity and specificity of FNAC for detection of salivary gland tumours is reported between 87 - 94% and 75 - 100% respectively [5-7]. Magnetic resonance imaging can assist in assessing the degree of soft tissue involvement and perineural invasion. In bronchopulmonary carcinoid (PC), somatostatin receptor imaging can visualise up to 80% of primary carcinoid tumors and are more sensitive for metastatic disease [10].

Histologically, the tumour comprises a dispersed population of malignant cells with round to oval nuclei, granular nuclear chromatin, a small but distinct nucleolus and minimal cytoplasm. The immunocytochemistry (ICC) shows that these cells stain positively for cytokeratin AE1/AE3 and for neuroendocrine markers CD56, chromogranin and synaptophysin (Figure 2). Primary salivary gland carcinoid tumour consists of nests and sheets of round to spindle-shaped argyrophilic elements containing small dense-core granules at ultrastructural level in direct contact with ductal structures lined by exocrine cells similar to those with normal salivary gland ducts [1,8]. Pulmonary carcinoid (PC), on the other hand, exhibits a paraganglioid structure, with small non-argyrophil cells and scanty Congo red-negative stroma [1]. Therefore, these two types are structurally, cytologically and histochemically different to suggest that two independent endocrine tumours can co-exist in the same patient. Plasma chromogranin A can be increased in PCs [10].

Metastases of bronchial carcinoids to the head and neck region are thought to be very rare. Koraitim., *et al.* report a case of widespread metachronous carcinoid tumour metastases to 6 different sites in the head and neck region involving both parotid glands, submandibular gland, upper lip and thyroid gland [11]. Other documented cases in the literature are parotid gland [12], submandibular gland [13], thyroid gland [14]. As our patient has a history of bronchopulmonary carcinoid tumour diagnosed a year ago and the rarity of primary carcinoid tumour in the salivary gland, it was necessary to establish whether the tumour in her parotid gland was a metastatic deposit or a synchronous tumour.

Surgery has been advocated the treatment of choice for both PCs and parotid carcinoid tumour with the aim of removing the tumor with preservation of the facial nerve if the nerve was functionally intact before surgery. Resection of metastases should be considered when possible with curative intent [10]. Somatostatin analogs are the first line treatment modality for carcinoid syndrome and could be considered as first-line systemic anti-proliferative treatment in unresectable tumour, particularly of low-grade typical and atypical carcinoid tumours [10]. Intensity-modulated radiation therapy may be considered for metastatic disease. Chemotherapy can be considered for progressive carcinoid tumour, though this has demonstrated limited effects. The most commonly used cytotoxic combination is etoposide and platinum. Temozolomide, however, has shown most clinical benefit [10]. In our case, however, no active intervention was instituted due to patient's significant comorbid state and patient's choice.

The overall 5-year survival rate for PC is 89% for all stages [15] but less is known for primary parotid carcinoid tumour. Soga and Yakuwa [16] described a series of 1875 tracheal and bronchopulmonary carcinoids within a Japanese carcinoid registry; significantly different 5-year postoperative survival rates were found between patients with typical and atypical carcinoid types (93.3% and 68.8%, respectively). This showed the 5-year survival rate is significantly worse for patients such as ours with atypical carcinoid tumour. At 18-month from the diagnosis of parotid gland atypical carcinoid tumour and 30-month for her PC, our patient was still alive on supportive care.

Conclusion

Parotid gland carcinoid tumour is a rare condition and should always be included in the differential diagnosis of a parotid lump. Radiological and cytological investigations are required to confirm the diagnosis and to establish whether this is either a primary tumour or metastatic deposit from the gastrointestinal and respiratory tracts. Surgical intervention in the form of partial or total parotidectomy is the mainstay of treatment. Chemoradiotherapy may be useful if surgical intervention is contraindicated and for metastatic and more advanced disease. Management of carcinoid disease should always include the awareness of possible metastatic spread to other locations. Early recognition and intervention are crucial for successful treatment outcome.

Figure 2: Fine needle aspiration cytology and histopathology. (a) Papanicolaou stain- specimen comprises a dispersed population of malignant cells with round to oval nuclei, granular nuclear chromatin, a small but distinct nucleolus and minimal cytoplasm. A fragment of salivary ductal epithelium is present in the top corner. (b) Immunocytochemistry (ICC) shows that these cells stain positively for cytokeratin AE1/AE3 and for neuroendocrine markers CD56, Chromogranin and Synaptophysin. (c) Carcinoid tumour diagnosed previously on bronchial biopsy (medium power photomicrograph).

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Conflict of Interest

None declared by all authors.

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