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Case study

Monoclonal origin of primary unilateral multifocal pleomorphic adenoma of the parotid gland☆

Sylvia L. van Egmond MD[a,*], Wendy W.J. de Leng PhD[b], Folkert H.M. Morsink BA[b], G. Johan A. Offerhaus MD, PhD, MPH[b], Lodewijk A.A. Brosens MD, PhD[b,*]

[a]*Department of Otorhinolaryngology and Head & Neck Surgery, University Medical Center Utrecht, The Netherlands*
[b]*Department of Pathology, University Medical Center Utrecht, The Netherlands*

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Summary Primary multiple pleomorphic adenomas in a unilateral parotid gland in previously untreated patients is a rare finding, and little is known about the etiology and pathogenesis. Here, a highly unusual case of a primary multifocal pleomorphic adenoma consisting of 15 individual nodules is presented. It is shown that all nodes are clonally related and thus share a common cell of origin excluding an independent multifocal pathogenesis. Most likely, multifocal pleomorphic adenoma represents parasitic nodules that have been detached from a main nodule, which may have been the result of undisclosed trauma.
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1. Introduction

Pleomorphic adenoma is the most common benign salivary gland tumor and is most frequently found in the parotid gland although it can also arise in other salivary glands. Histologically, pleomorphic adenoma is characterized by typical myxoid to chondroid stroma with a variable admixture of epithelial elements forming tubular or solid structures and stromal areas containing myoepithelial cells [1]. It has previously been shown that the epithelial and stromal elements are monoclonal and are therefore derived from the same precursor cell [2,3].

Multiple tumors arising in one salivary gland is rare in previously untreated patients. In particular, primary unilateral multifocal pleomorphic adenomas in a parotid gland is very rare. Most cases reported in literature describe 2 pleomorphic adenoma nodules in one gland although one patient with 4 separate nodules has been described [4]. Little is known about the etiology and pathogenesis of multifocal pleomorphic adenoma, and clonal relationship has only been studied in one patient presenting with 2 unilateral pleomorphic adenomas [4].

We present a patient with primary unilateral multifocal pleomorphic adenoma consisting of 15 separate nodules ranging from 0.1 to 1.6 cm. In addition, it is shown that these separated tumors are clonally related supporting a common origin of these lesions.

2. Case report

2.1. Clinical history

A 55-year-old woman presented with multiple painless masses in the parotid region on the right side. On further inquiry one swelling was already present for 20 years. Subsequently, multiple swellings arose in the same region

☆ Conflicts of interest: none.

* Corresponding authors.
E-mail addresses: S.L.vanegmond@umcutrecht.nl (S. L. van Egmond), l.a.a.brosens@umcutrecht.nl (L. A. A. Brosens).

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over the last few years. There was no pain, discomfort, malaise, facial nerve weakness, night sweats or unintentional weight loss. Importantly, there was no history of trauma, fine needle aspiration, parotid surgery or radiation to the head and neck.

Past medical history revealed right sided endoscopic sinus surgery for sinusitis with dental origin and a cerebellar disorder due to intermittent alcohol abuse for 20 years.

Physical examination revealed four separate firm, mobile and non-tender masses in the parotid region. The largest lesion was located preauricular and measured approximately 1.5 cm. The other palpable masses were located in the inferior portion of the parotid gland. Facial nerve function was intact and no weakness was noted. Further physical examination was unremarkable.

Magnetic resonance imaging confirmed the presence of multiple nodules in the right parotid gland, including one larger mass (1.6 cm) and at least 5 smaller separate masses (Fig. 1).

Fine needle aspiration of the largest lesion showed myoepithelial cells intermingled with stroma and extracellular matrix, compatible with pleomorphic adenoma. A superficial parotidectomy with facial nerve dissection and preservation was performed. A temporary partial facial nerve paresis due to postoperative swelling recovered completely.

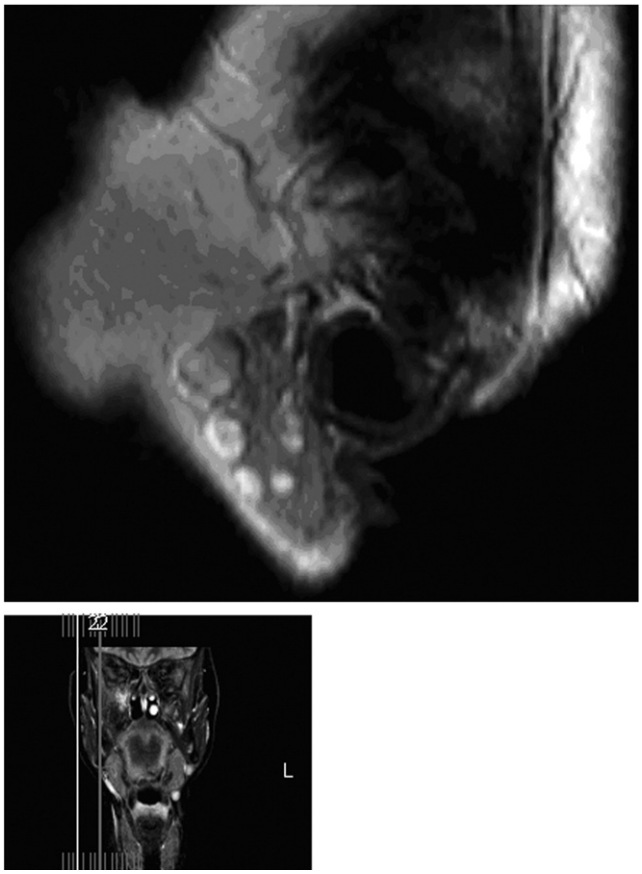

Fig. 1 MRI; T2 Lateral view with reference image showing multiple masses in the right parotid gland.

2.2. Pathological findings

Gross histopathological examination revealed nine separate nodules in the parotid gland ranging in diameter from 0.4 to 1.6 cm. Microscopic examination revealed a total of 15 separate pleomorphic adenoma nodules ranging in diameter from 0.1 to 1.6 cm. In addition, one small Warthin tumor was noted (diameter 0.1 cm). The resection margins were free and there were no signs of malignancy (Fig. 2).

3. Materials and methods

Formalin-fixed paraffin-embedded tissue from nine of the 15 pleomorphic adenoma nodules in the resection specimen was obtained by microdissection (Fig. 2). Genomic DNA was isolated by standard methods. Because the stromal and epithelial cells in a pleomorphic adenoma are clonally related, total DNA of the individual nine nodules was isolated, instead of separate microdissection of the stromal and epithelial cells [2,3,5].

Clonal relation between these nine nodules was assessed by a clonality assay based on random X-chromosome inactivation in females and amplification of the *human androgen receptor* (*HUMARA*) gene on the X-chromosome as described previously [2,3,6].

Briefly, polymerase chain reaction (PCR) amplification of the *HUMARA* gene was performed on tumor DNA before and after incubation with the methylation sensitive restriction enzyme HpaII (New England Biolabs, Ipswich, MA). Enzymatic digestion with HpaII followed by this PCR will

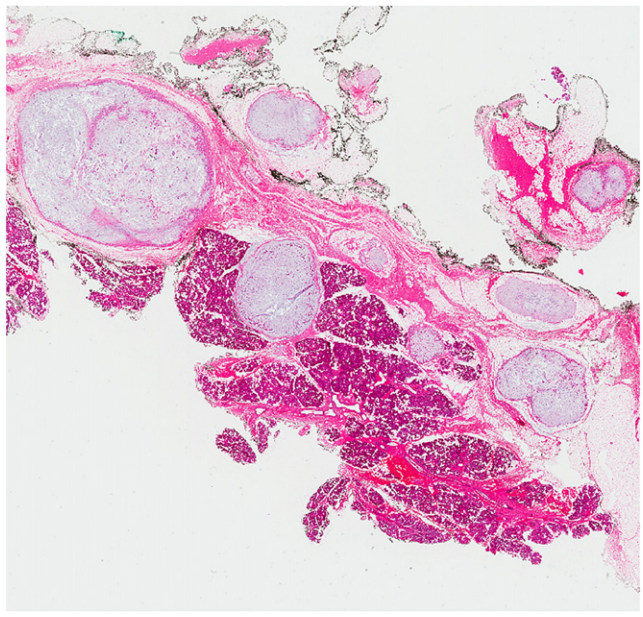

Fig. 2 Histological section showing multiple nodules of pleomorphic adenoma surrounded by normal parotid gland tissue (Hematoxylin and eosin staining).

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