Benign Pleomorphic Adenoma in the Facial Nerve

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An otherwise healthy, 55-year-old ex-smoker presented in clinic with a 1-month history of moderate right facial nerve palsy and oral ulcers. This represented a grade III House–Brackmann score.

His relevant medical history included excision of a right benign submandibular salivary lump at age 17 (1977) of which details are vague. In 2007, he presented with a 6-month history of gradual onset of a 3 cm × 2 cm hard, immobile lump, intermittently tender, inferior to his right ear, with a palpable lymph node in the right anterior triangle. Operative findings confirmed a large benign mixed pleomorphic adenoma (BMPA) in the deep lobe of the parotid gland and a small satellite lesion in the superficial lobe. He was treated with a right near total parotidectomy with facial nerve preservation due to concerns of tumor spillage from the original excision (1977).

In 2011, magnetic resonance imaging (MRI) was conducted after he experienced mild pain and discomfort in his neck, clinically suggesting a great auricular neuroma. The MRI showed no evidence of new lesions and a normal right facial nerve (Figure 1).

For his recent presentation, a virally induced facial palsy was initially suspected. This was treated conservatively with a short course of prednisolone and omeprazole. Over the subsequent 3 months, his symptoms progressed. On physical examination, he had a grade VI House–Brackmann right facial paresis. Otologic examination and flexible nasolaryngoscopy were unremarkable. Magnetic resonance imaging T2-weighted images revealed a contrast-enhancing irregular, solid mass of the right facial nerve measuring 1.4 cm (craniocaudal) × 0.9 cm (transverse), centered on the descending portion of the right facial nerve, proximal to the right stylomastoid foramen (Figure 2). The skull base was normal, with no evidence of recurrence within the parotid bed or cervical lymphadenopathy. A right transmastoid facial nerve biopsy confirmed a BMPA. Histology revealed no evidence of malignancy and no association with any nerve (Figure 3).

Following the original biopsy, a definitive cranial nerve VII excision and upper neck and mastoid dissection were undertaken with an immediate cable graft of the great auricular nerve. Operative findings confirmed unusual facial nerve involvement, with tumor extending from just proximal to the second genu, distally to the stylomastoid foramen. Anatomy was otherwise unremarkable and histology confirmed a BMPA.

Adjuvant radiotherapy was discussed as these are slow-growing tumors, but due to the significant side effects, the patient was not keen. He was referred to plastics for a canthoplasty, with gold weight insertion. Close, long-term follow-up is planned with otolaryngology. At 9 months postoperative, the nerve graft function has shown minimal improvement and the patient has been referred for dynamic facial reanimation.

This case describes a rare manifestation where a BMPA is centered in the mastoid bone, around the descending portion of the right facial nerve, proximal to the stylomastoid foramen.

Recurrence is a recognized feature of benign tumors that occur decades after the primary surgery due to breech of the tumor capsule, incomplete excision, or tumor spillage.1-4 Perineural involvement is an atypical characteristic in benign tumors. Of interest, Kunimoto et al describe a case of temporal

Figure 1. Postoperative follow-up MRI of the neck in 2011. This shows a follow-up MRI, postoperatively from 2007. There is no evidence of recurrence at the original site, no new lesion, and no lymphadenopathy noted. MRI indicates magnetic resonance imaging.

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bone invasion by BMPA, without malignant morphology. Similarly Jung et al describe a BMPA in the parotid gland with 2 foci of perineural involvement, despite this displaying malignant characteristics.  

Alternatively, small carcinoma ex pleomorphic adenomas (CXPA) can arise at onset as a malignant tumor or transform from a BMPA if long-standing. Park et al and Tsutsumi et al describe cases where CXPA demonstrate perineural involvement along the facial nerve to the internal auditory canal. Despite a differential, our histology makes this unlikely. Embryologically in the sixth week of prenatal development, epithelial buds on the cheek grow posteriorly toward the otic placoide of the ears, to form terminal ends that develop glandular acini near the developing facial nerve. Thus, residual tissue may arise near the duct that has the potential to develop ectopic salivate glands as documented in multiple sites in the head and neck. Peters et al report an isolated BMPA occurring in the middle ear and mastoid most likely arising from ectopic tissue. Although plausible, reports make this differential unlikely.

We present a clinical entity of a BMPA, originating within a bony canal, without malignant transformation, where adjacent spread is unlikely. Consequently facial nerve dysfunction can be a sequela of benign pleomorphic adenomas presenting in unusual locations where their biological behaviour cannot be explained. Any management regarding these tumors should be meticulous and close systemic follow-up maintained.

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References
1. O’Regan B, Bharadwaj G. Tumour recurrence after surgical removal of parotid pleomorphic salivary adenoma using a retrograde facial nerve dissection technique. Br J Oral Maxillofac Surg. 2012;50(5):417-419.
2. Peters BR, Maddox HE III, Batsakia JG. Pleomorphic adenoma of the middle ear and mastoid with posterior fossa extension. Arch Otalaryngol Head Neck Surg. 1988;114(6):676-678.
3. Colella G, Cannavale R, Chioldini P. Meta-analysis of surgical approaches to the treatment of parotid pleomorphic adenomas and recurrence rates. J Craniomaxillofac Surg. 2015;43(6):738-745.
4. Kunimoto Y, Di Lella F, Sivalingam S, De Donato G, Falcioni M, Sanna M. Temporal bone invasion by recurrent benign pleomorphic adenoma of the parotid gland. Int Adv Otol. 2013;9(1):140-143.
5. Jung SJ, Lee JC, Choi HJ, et al. Perineural involvement in benign mixed tumor, Letter to the editor. The Korean J Pathology. 2013;47:403-404.
6. Tsutsumi T, Nakajima N, Hirose T, Watanabe K. Total-length invasion of the facial nerve by parotid carcinoma ex pleomorphic adenoma. Auris Nasus Larynx. 2009;36(5):618-622.
7. Park KC, Choi HJ, Kwon JK. Carcinoma ex pleomorphic adenoma mimicking multiple facial nerve schwannoma. Auris Nasus Larynx. 2008;35(2):291-294.
8. Wang D, Chen Y, Guo C. Pleomorphic adenoma of the cheek: a case report arising from accessory parotid gland around Stensen’s duct orifice. J Oral Maxillofac Surg Med Pathology. 2016;28:341-343.
9. Sharma NK, Singh AK, Pandey A, Verma V. Pleomorphic adenoma involving soft tissue overlaying the anterior border of ramus of the mandible: a rare ectopic presentation. J Oral Biol Craniofac Res. 2016;6(suppl 1):S62-S64.