Pleomorphic Adenoma of the Upper Lip: Some Clinicopathological Considerations

Sir,

We read with great interest the article by Kataria *et al*.[1] regarding pleomorphic adenoma (PA) of the upper lip. Although this report reveals useful information, there are some areas for discussion upon which we would like to expand, based on our previous publications on PA of the upper lip[2] and the parotid gland.[3]

First, most of the PAs occur at the parotid gland (85%) and the submandibular gland (8%). Only 7% of the cases are intraoral PAs, which commonly involve the palate, upper lip or buccal mucosa. This tumour can be found at the tongue, retromolar trigone, floor of the mouth, jaw bone, tonsillar fossa, oropharynx, internal and middle ear, lacrimal gland, sinonasal mucosa and cervical lymph nodes.[2] Differential diagnosis of a clinically benign mass of the upper lip can be classified into three lesion groups: (1) salivary gland tumours such as PA, canalicular and mucoepidermoid carcinomas; (2) mesenchymal tumours such as lipoma, leiomyoma, nerve tumours (neurofibroma, neurilemmoma/schwannoma), benign fibrous histiocytoma, oral focal mucinosis and granular cell tumour; and (3) infections such as tuberculosis, syphilitic gumma (if the lesion presents as an ulcerated mass) and deep fungal infections (histoplasmosis, cryptococcosis, blastomycosis, coccidioidomycosis).[2]

Second, 3–15% of the cases develop dysplasia or intracapsular carcinoma. Malignant transformation of PA is suspected when the tumour grows rapidly, presents with pain or irregular borders or local neurological disturbances such as anaesthesia, paraesthesia, or it is covered by ulcerated/irregular mucosa or skin. Multiple recurrences also increase the risks of malignant transformation of PA. Studies have shown the relationship between malignant transformation and some histopathological features of the tumour, including micronecrosis, tumour invasion of the surrounding tissues, increased vascularity, dystrophic calcification, atypical meiosis, excessive hyalinisation and cell-rich variant of the tumour.[2] Once these clinical or histopathological features are found, the attending clinician and pathologist should pay more attention on malignant transformation. Long-term follow-up of the patient is recommended.[2]

Third, several factors are associated with recurrences of PA. These include incomplete removal, incomplete capsulation with pseudopenetration or satellitosis of the tumour, intraoperative opening of the tumoral capsule and spillage of tumoral mucoid because of intraoperative burst of the tumour.[2] Some histopathological features have also been found to be linked to recurrences of this tumour, including predominantly cellular or extremely cellular presentation, or hypocellular tumour in abundant myxoid or chondromyxoid stroma.[2] Incomplete resection of the tumour and intraoperative opening of the capsule can result in recurrence as multiple discrete foci within the previously operated salivary gland, surrounding tissues or scar. Hence, incisional biopsy should be avoided if PA is suspected because it can increase recurrence risks.[2]

Fourth, microscopically the epithelial component of PA comprises ductal epithelium and myoepithelial cells. However, it is often difficult to ensure the presence of myoepithelial cells within this tumour by using a light microscope alone. Over the past decade, an immunohistochemical analysis has played an important role in diagnosing or confirming the presence

of myoepithelial cells. Myoepithelial cells present immunoreactivity to cytokeratin, S-100 protein, glial fibrillary acidic protein (GFAP), actin and vimentin, whereas ductal epithelial cells and solid cellular nest with tubular structure are strongly immunoreactive to cytokeratin and moderately immunoreactive to epithelial membrane antigen (EMA) and carcinoembryonic antigen (CEA). In certain cases where the surgical specimen is small, it may be difficult to histopathologically segregate PA from basal cell adenoma and tubular variant of adenoid cystic carcinoma. Anti-GFAP antibody can be used because it reacts only with epithelial and stromal components of PA.[2]

Lastly, the treatment of choice of PA is complete excision of the tumour and its capsule. PA of the lip merits a localised V-wedge excision. Resection of one-third of the lower lip or one-fourth of the upper lip can be closed primarily without interference of cosmetic results.[2] Enucleation (simple "shell-out") and extracapsular dissection (ECD: Tumour extirpation in a plane 3–4 mm peripheral to its capsule) of the lesion should be avoided since PA has incomplete capsule and tumour pseudopenetration outwards is common. Data from recent meta-analyses have shown that enucleation and ECD of PA of the parotid gland produce 9 and 10 times higher rates of recurrence compared with superficial parotidectomy.[3] Radiotherapy is not recommended for PA because it does not reduce recurrence rates, a recurrence-free interval and the size of safety margins needed intraoperatively. Moreover, irradiation can create tissue ischaemia and fibrosis, and osteoradionecrosis, as well as can retard the recovery of the facial nerve.[2,4] Twenty-year survival rate after the complete excision is 96.5% if there is no malignant transformation.[2]

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