Rare Floret Like Tyrosine Crystals in Pleomorphic Adenomas of Parotid Gland

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Abstract The pleomorphic adenoma arising in the parotid gland is a benign neoplasm that is aptly named because of its histomorphological diversity. The stromal component can contain chondromyxoid material, amyloid, and elastic fibers, along with a few rare reports of crystalline structures present in this tumor. Especially, the crystalline components are rarely encountered or appreciated in routine pathology reporting. Here, we report a case of pleomorphic adenoma of the parotid gland, in which tyrosine-rich crystalloids were identified in abundance and were confirmed with special stains and electron microscopy. Though their exact source is not yet known, crystallization of the stromal or myoepithelial cell secretion has been hypothesized. This comprehensive report is to make the histopathologists aware of this rare morphological observation in pleomorphic adenomas so that more cases are identified and followed-up to reveal the impact of their presence in a subset of pleomorphic adenomas.

Keywords Pleomorphic adenoma · Tyrosine crystals · Tumoral stroma · Electron microscopy

Introduction

Pleomorphic adenoma accounts for about 60% of all salivary neoplasms and about 80% of them arise in the parotid, 10% in the submandibular gland and 10% in the minor salivary glands of the oral cavity, nasal cavity and paranasal sinuses and the upper respiratory and alimentary tracts [1]. Tyrosine rich crystalloids (TRCs) have been described in various neoplasms of the major and minor salivary glands, with isolated examples in ceruminous and lacrimal gland tumors and fibrous tissue in laryngectomy specimens [2]. Though, such oret-shaped orangeophilic extracellular TRCs measuring up to 100 μm in the greatest dimension were described long back in 1953, till now their exact source and significance are not known. Some suggestions for their origin from the stromal or myoepithelial cell secretions have been given [3].

Case History

A 38-year male patient presented with swelling over the right parotid region for 10 years. On examination, a 5 × 5 cm firm mass was seen in the right parotid region. Fine needle aspiration cytology from the swelling showed features suggestive of pleomorphic adenoma. The patient underwent right superficial parotidectomy weighing 33.6 grams and measuring 6.5 × 6 × 4 cm with an intact capsule. On serial slicing, a greyish-white lobulated tumor with myxoid areas was identified measuring 4 × 3.7 × 3.2 cm

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(Fig. 1a, b). Multiple sections examined from the tumor showed histomorphological features of pleomorphic adenoma with numerous TR crystals. The crystals were orangeophilic and showed a central hallow, surrounded by radiating club shaped crystals (Fig. 1c, d). Transmission electron microscopy (TEM) showed osmophilic tyrosine rich crystalloids composed of an electron-dense centre with radially arranged, blunt-ended, sometimes club-shaped to lobular osmophilic structures that often appeared to be continuous with a central core (Fig. 1e, f). No feature of malignancy or unusual clinical or pathological findings were noted in the index case.

Discussion

Pleomorphic adenoma is characterized microscopically by architectural and cellular characteristic features. Extracellular stroma is one of the defining components of this tumor, ranging from scant to abundant in amount. Epithelial and modified myoepithelial elements are seen intermingled with myxoid, chondroid, chondromyxoid, fibrillary, hyaline, fibrous, sclerohyaline and, very rarely,

osseous and adipose tissue components. Most pleomorphic adenomas, particularly the longstanding lesions present variable amounts of elastic fibres, which are uncommon in other salivary gland tumors [1–3].

Tyrosine crystals in salivary gland pleomorphic adenomas is an unexplained phenomenon, though it appears that the deposition of crystals may be related to the long duration of the tumor like the index case [4]. Though, TRCs may be demonstrated in around 1.5–5% of pleomorphic adenomas, in a study based on African patients the TRCs were identified in up to 21% of PAs [2–5]. Due to the abundance of tyrosine in various animal tissues, as hair cortex, thyroid colloid, paneth cell granules, and zymogen granules, they are likely components of the salivary secretions [6, 7].

They can be identified both in histological sections and cytological preparations but are not apparent macroscopically [2]. These crystal are refractile under polarized microscopy without any distinct characteristic feature. TRCs have also been identified in other tumors including adenoid cystic carcinoma, polymorphous low-grade adenocarcinoma, terminal duct adenocarcinoma of minor

Fig. 1 Gross photograph of the superficial parotidectomy specimen showing a homogeneous grey-white lobulated tumor (arrows) with peripheral rim of normal parotid gland (a). Fine needle aspiration cytology smear shows numerous crystals (arrows) in a background of pleomorphic adenomas (b) [×100]. Histophotomicrographs show

orangeophilic crypts (arrows) in a background of myoepithelial cells and myxoid background material (c) [×100], (d) [×200]. TEM images show the crystals with electron dense cores and radiating clubshaped electron dense crystals (arrows) characterizing Tyrosine crystals (e) [×2050], (f) [×2600]

salivary glands, malignant mixed tumor and tumors of ceruminous and lacrimal glands. They can also be found in non-neoplastic lesions like in parotid cysts, the benign connective tissue of the larynx, and bronchial connective tissue following surgery [3–6].

Interestingly, TRCs are commoner in mixed salivary gland tumors of people with black ethnicity than in whites, possibly related to imperfect pigment metabolism containing tyrosine. Disordered protein synthesis by tumor cells or local changes in the concentration of mucopolysaccharides and inorganic ions in the matrix have also been suggested [2–4]. It is also a question why in some of the salivary lesions only the TRCs are found, not in all? It may be also possible that the crystals are simply a manifestation of disordered protein synthesis by the tumor cells in tumors which are present over last couple of years [5–7]. But, importantly, till now there is no clinical significance of the salivary gland tumors TRCs have been published. Possibly, they do not have any impact on patients' prognosis. However, our overall knowledge is limited.

In summary, numerous extracellular floret like shiny tyrosine crystals can be identified in salivary gland lesions in up to one-fifth of all cases with characteristic appearance under polarized light and with TEM; however, their exact clinical impact is not yet known.

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Compliance with ethical standards

Conflict of interest The author declares that they have no conflict of interest.

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