A basaloid tumour of the parotid gland

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Abstract

Basal cell adenocarcinoma is a malignant basaloid tumour most seen in the parotid gland. This case report and subsequent discussion highlights the key diagnostic findings seen in this rare tumour.

Keywords Basal cell adenocarcinoma; beta-catenin; cytology; salivary gland tumour

Case report

A 64 year old female presented with a slowly growing parotid mass over the preceding 1–2 years. Ultrasonography showed a 16 mm well defined lesion in the superficial lobe of parotid gland. Fine needle aspiration cytology was undertaken which showed a moderately cellular sample comprising three dimensional aggregates of basaloid cells (Figure 1) amounting to a Milan Category IVB (Salivary Gland Neoplasm of Uncertain Malignant Potential (SUMP)). Surgical excision of the superficial lobe of parotid was undertaken which histologically showed a relatively well circumscribed, biphasic salivary neoplasm (Figure 2). The tumours comprised of both epithelial and myoepithelial cells arranged in tubules and trabeculae with areas of basement membrane like material (Figure 3). There were occasional mitotic figures present but no cytologically high grade areas or areas of necrosis were identified. The lesion focally breached its capsule into the surrounding fat (Figure 4). Immunohistochemistry highlighted the biphasic nature of the tumour (Figure 5) and focal nuclear positivity with beta-catenin was seen (Figure 6). The diagnosis of basal cell adenocarcinoma was made. The tumour was clear of the surgical margins. A subsequent CT scan did not show any lymphadenopathy or distant metastasis. No further intervention was required and on 6-month surgical follow-up, there was no evidence of recurrence.

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Discussion

Basal cell adenocarcinoma (BCAd) of the salivary glands is a rare tumour. One retrospective study of cases from the American National Cancer Database found that of the 36,224 cases of salivary gland tumours recorded between 1998 and 2012 only 509 (1.4%) were classified as BCAd.[1] This study found the tumours occurred equally among males and females and that they often appeared in the sixth or seventh decade. Of these 509 cases, 88% occurred in the parotid glands, with the remaining 11.2% in the submandibular and 0.8% in the sublingual glands.[1] The 5-year survival rate has been reported as between 42 and 79%.[1,5]

One study reporting on the cytological features of 7 BCAds showed all samples to be cellular with no necrosis seen. The majority of these containing large clusters of predominantly small and round neoplastic cells.[2] Distinguishing between basal cell adenoma and BCAd on fine needle aspiration cytology is challenging and histologic evidence of capsular invasion is required to make a definitive diagnosis. However certain features can point towards a diagnosis of malignancy. In one cytological case series of 15 salivary basal cell tumours, the authors found that cases with nuclear pleomorphism, large three-dimensional cellular clusters, mitotic figures, and/or necrosis were diagnosed as BCAd on subsequent histologic examination.[3]

Immunohistochemistry shows a classic biphasic pattern with the epithelial components staining positive for example with CK, EMA, CEA, c-kit and the basal/myoepithelial component staining with myoepithelial markers eg p40.[4]

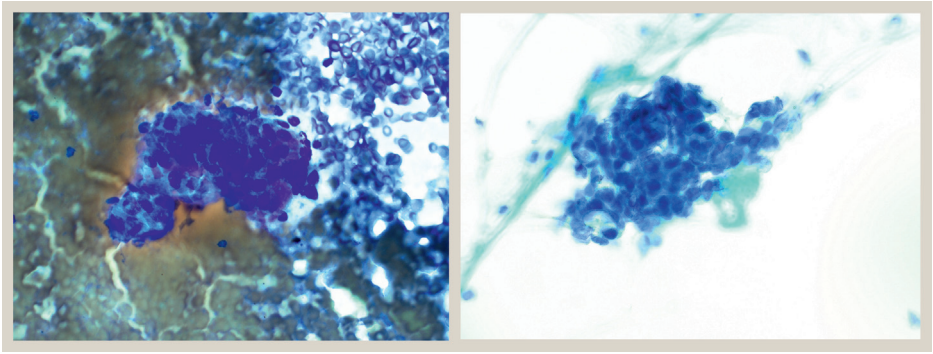

Figure 1 FNA cytology of the lesion. Staining after Giemsa (left) and Papanicolaou (right) show three dimensional clusters of cells with relatively bland, round hyperchromatic nuclei and scant cytoplasm giving a basaloid appearance.

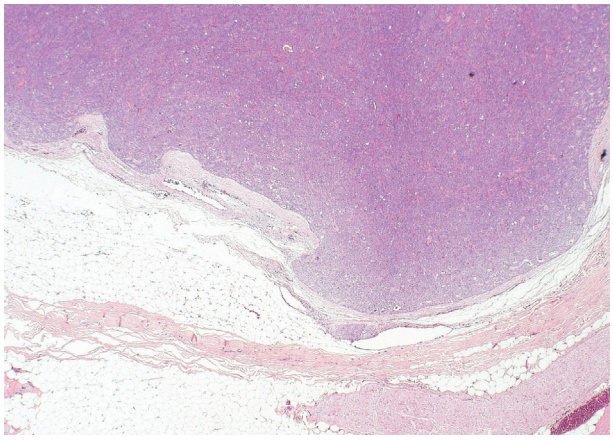

Figure 2 H&E at ×20 magnification showing an irregular, but relatively well-circumscribed lesion.

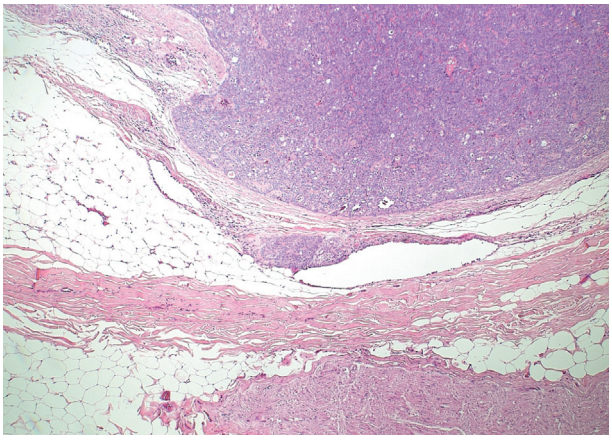

Figure 4 H&E at ×40 magnification shows an area of capsular breach with invasion into fat.

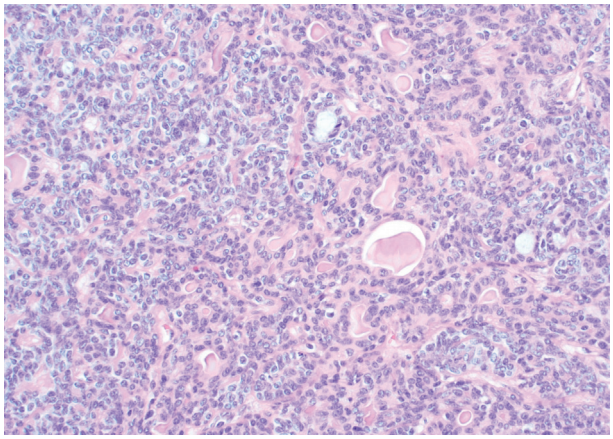

Figure 3 H&E at ×200 magnification shows a biphasic salivary neoplasm comprising epithelial and myoepithelial cells arranged in tubules and trabeculae with areas of basement membrane like material.

Mutations in the genes *CYLD*, *PIK3CA*, and *CTNNB1* have been previously reported. Mutations in *CTNNB1* form the basis for the nuclear beta-catenin positivity seen on immunohistochemistry in 33–100% of cases of both basal cell adenoma and BCAd[5]. Co-expression of LEF-1 (Lymphoid enhancer binding factor 1) has been shown in BCAd, however LEF-1 can also be positive in pleomorphic adenoma.[5,6]

When the differential diagnosis lies between BCAd and adenoid cystic carcinoma (AdCC), FISH testing for the *MYB-NFIB* or *MYBL1-NFIB* fusion genes which are frequently identified in AdCC can help.[4] No specific molecular alteration has been shown for basal cell adenocarcinoma.[6]

Conclusion

This rare tumour with often subtle features of cytological atypia should remain in the differential diagnosis of a basaloid tumour upon FNA of a salivary gland mass, particularly from the parotid gland. Positive immunohistochemical nuclear beta-catenin

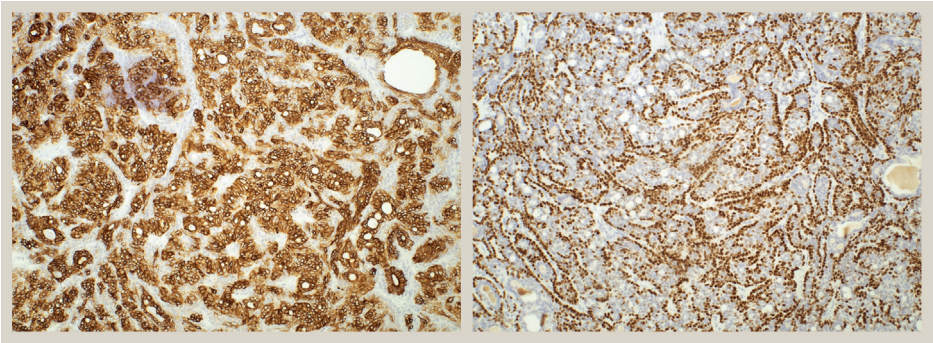

Figure 5 Immunohistochemistry with CAM5.2 (left) and P40 (right) highlighting the biphasic nature of the tumour.

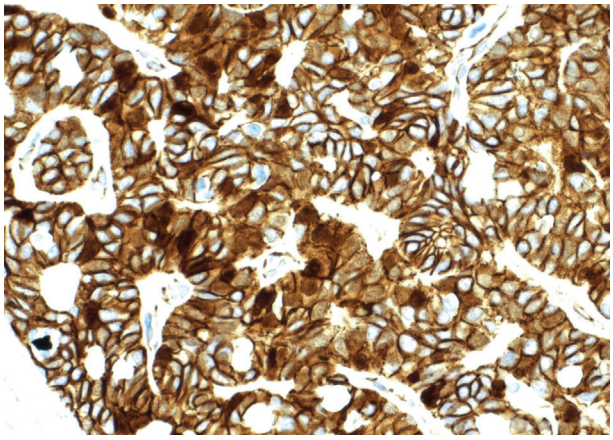

Figure 6 Immunohistochemistry showing focal nuclear beta-catenin positivity.

staining along with negative FISH testing can aid separate between similar basaloid entities. Surgical excision is required to definitively classify as either basal cell adenoma or basal cell adenocarcinoma. ◆

Practice points

- Basal cell adenocarcinoma is a rare tumour with often bland cytological features regularly showing three-dimensional aggregates of basaloid cells on FNA cytology
- The differential diagnosis includes basal cell and pleomorphic adenoma along with adenoid cystic carcinoma and polymorphous adenocarcinoma
- Immunohistochemistry identifies a biphasic tumour comprising of epithelial and basal/myoepithelial components
- Positive nuclear beta-catenin staining can help distinguish between this and other basaloid neoplasms
- Surgical excision is required to definitively classify as either basal cell adenoma or basal cell adenocarcinoma

Self-assessment MCQs

1. Which of the following is false regarding basal cell adenocarcinoma?

A. Basal cell adenocarcinoma is most often seen in the parotid gland
B. Basal cell adenocarcinoma occurs most often in the 6th or 7th decade
C. Basal cell adenocarcinoma is more common in males

Correct answer: (C).
This tumour is found equally in males and females.

2. Which of the following is false regarding basal cell adenocarcinoma?

A. LEF-1 is a specific immunohistochemical marker
B. Nuclear beta-catenin is seen in both basal cell adenoma and basal cell adenocarcinoma
C. No specific molecular alteration has been identified for this tumour

Correct answer: (A) LEF-1 is non-specific and can also be positive in pleomorphic adenoma.

3. Which of the following is not a reported cytological feature of basal cell adenocarcinoma?

A. Nuclear pleomorphism
B. Clusters of basaloid cells
C. Occasional mitotic figures
D. Granulomas

Correct answer: (D)

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