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Case Report

A case of alopecic primary cutaneous adenoid cystic carcinoma successfully managed with the aid of prostate-specific membrane antigen-positron emission tomography imaging

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ABSTRACT

Primary cutaneous adenoid cystic carcinoma (PCACC) is a rare adnexal malignancy, most commonly affecting the scalp, and typically characterised by slow growth, marked local invasiveness and a high rate of local recurrence. Distant metastasis however remains uncommon. We report a case of a 58-year-old woman with a 6-year history of a progressively enlarging tender alopecic scalp lesion. Histopathology demonstrated a dermal basaloid neoplasm with tubular, nested and cribriform architecture and prominent perineural invasion, consistent with adenoid cystic carcinoma. Immunohistochemical studies highlighted a ductal cell component positive for cytokeratin 7 and CD117, and a myoepithelial cell component positive for smooth muscle actin and p40. Initial staging with ^{18}F-fluorodeoxyglucose-positron emission tomography/ computed tomography (^{18}F-FDG PET/CT) showed no significant metabolic activity or metastatic disease. Given emerging evidence of prostate-specific membrane antigen (PSMA) expression in adenoid cystic carcinoma, ^{68}Ga-PSMA PET/CT was performed and demonstrated moderate uptake confined to the primary scalp lesion without regional or distant disease. The patient underwent staged wide local excision with clear histological margins and split-thickness skin graft reconstruction. Six-month follow-up ^{68}Ga-PSMA PET/CT showed no evidence of residual or recurrent disease. To the authors' knowledge this is first documented case of PSMA-avid PCACC and suggests that ^{68}Ga-PSMA PET/CT may have a role in staging and surveillance of this rare tumour. Further studies are required to support these findings.

Keywords: Primary cutaneous adenoid cystic carcinoma, Prostate specific membrane antigen, PSMA-PET imaging

INTRODUCTION

Adenoid cystic carcinoma (ACC) is a rare malignancy most commonly originating in the salivary glands and is typically locally aggressive with a high risk of local recurrence and a propensity for late distant metastasis.[1] In contrast, PCACC is a much rarer and typically slow-growing adnexal tumour, first described by Boggio in 1975.[2] PCACC is thought to arise from eccrine sweat ducts, with some cases harbouring a t(6;9) MYB:NFIB gene fusion.[3] The scalp is the most affected site, however, other documented sites include the chest wall, abdomen,

back, eyelids and perineum.[1] Despite its tendency for local recurrence, occurring in approximately 50% of cases, metastasis in PCACC is extremely rare.[1] Due to the rarity of PCACC, there are no formal, widely accepted guidelines for treatment and follow-up specific to this tumour.

PSMA is a transmembrane protein over-expressed by prostate cancer cells and used in staging, re-staging and treatment of patients with prostate cancer. PSMA is also expressed in primary, recurrent and metastatic ACC of the salivary gland.[4] To the authors' knowledge, this is the first reported case of making ^{68}Ga-PSMA-positron emission tomography/computed tomography (^{68}Ga-PSMA PET-CT) uptake in PCACC, implying that PCACC also expresses ^{68}Ga-PSMA.

CASE REPORT

A 58-year-old female presented to dermatology with a 6-year history of a progressively enlarging and tender patch on the scalp. She denied any pruritus, bleeding or systemic symptoms. The presentation was delayed due to undisclosed personal reasons. Physical examination revealed a 7×8 cm erythematous patch on the right parietal area with overlying alopecia (Figure 1).

Punch biopsy from the scalp demonstrated dermal tumour infiltration comprised of uniform, bland, basaloid neoplastic cells variably organised in tubules, nests and cribriform configurations, with foci of perineural invasion (Figure 2). A ductal cell population was highlighted immunohistochemically using cytokeratin 7 and CD117, while a myoepithelial cell population was evident on smooth muscle actin and p40 stains. The tumour failed to express cytokeratin 20 and GATA3. The histological findings were compatible with ACC.

Staging (^{18}F-FDG PET/CT) scan showed no significant FDG uptake in the primary tumour and no FDG avid loco-regional nodal or distant metastases. Recent immunohistochemical and PET imaging studies have demonstrated increased PSMA expression in ACC, making ^{68}Ga-PSMA-PET/CT a promising diagnostic tool.[4,5] Therefore, a ^{68}Ga-PSMA-PET/CT was performed to further stage the tumour. This showed moderately increased tumour related tracer uptake in the right fronto-parieto-temporal scalp region and no other PSMA avid lesions, confirming a diagnosis of PCACC.

Management was coordinated through an integrated multidisciplinary team. Mapping biopsies were taken to delineate the tumour, based on clinical examination, dermoscopy and ^{68}Ga-PSMA-PET/CT findings. Closure of the primary excision site was achieved using a split-thickness skin graft. Complete tumour excision was confirmed histologically following two wide local excisions, achieving a 1-2 cm clearance margin. The tumour measured 8 cm at its largest dimension and was histologically uniform, lacking areas of solid growth or features of high-grade transformation. Florid perineural invasion was confirmed. Although post-operative radiotherapy was recommended to reduce risk of local recurrence, the patient declined this option. Six months after diagnosis, a follow-up ^{68}Ga-PSMA-PET/CT did not identify any macroscopic PSMA-avid residual, recurrent or metastatic sites of disease (Figure 3). The patient will be followed up with clinical assessments and 6-monthly ^{68}Ga-PSMA-PET/CT.

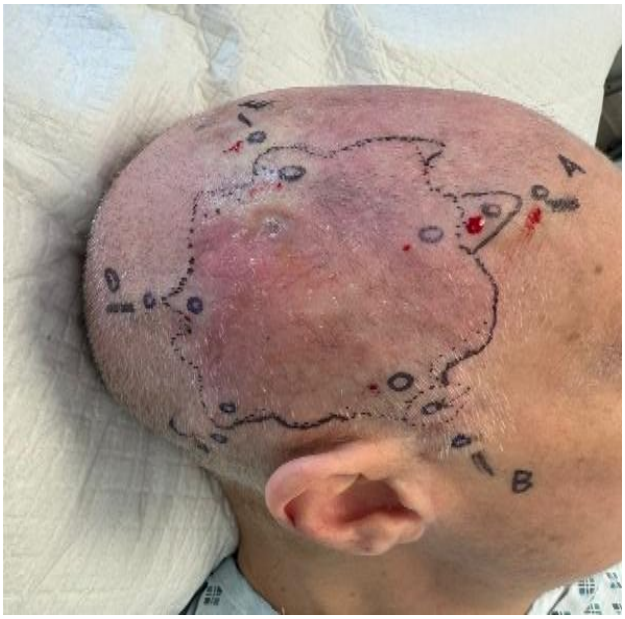

Figure 1: Clinical photo showing ill-defined erythematous patch on the right fronto-parieto-temporal scalp.

*The lesion and designated sites for mapping biopsies are outlined.

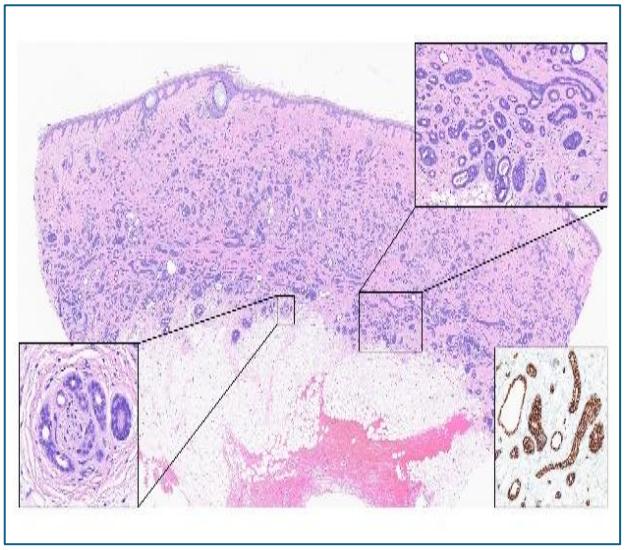

Figure 2: Skin punch biopsy from the scalp.

*Diffuse dermal infiltration by a basaloid neoplasm organised in tubules, nests and cribriform structures (top right inset), exhibiting perineural invasion (bottom left inset) and positive staining for CD117 immunohistochemistry (bottom right inset).

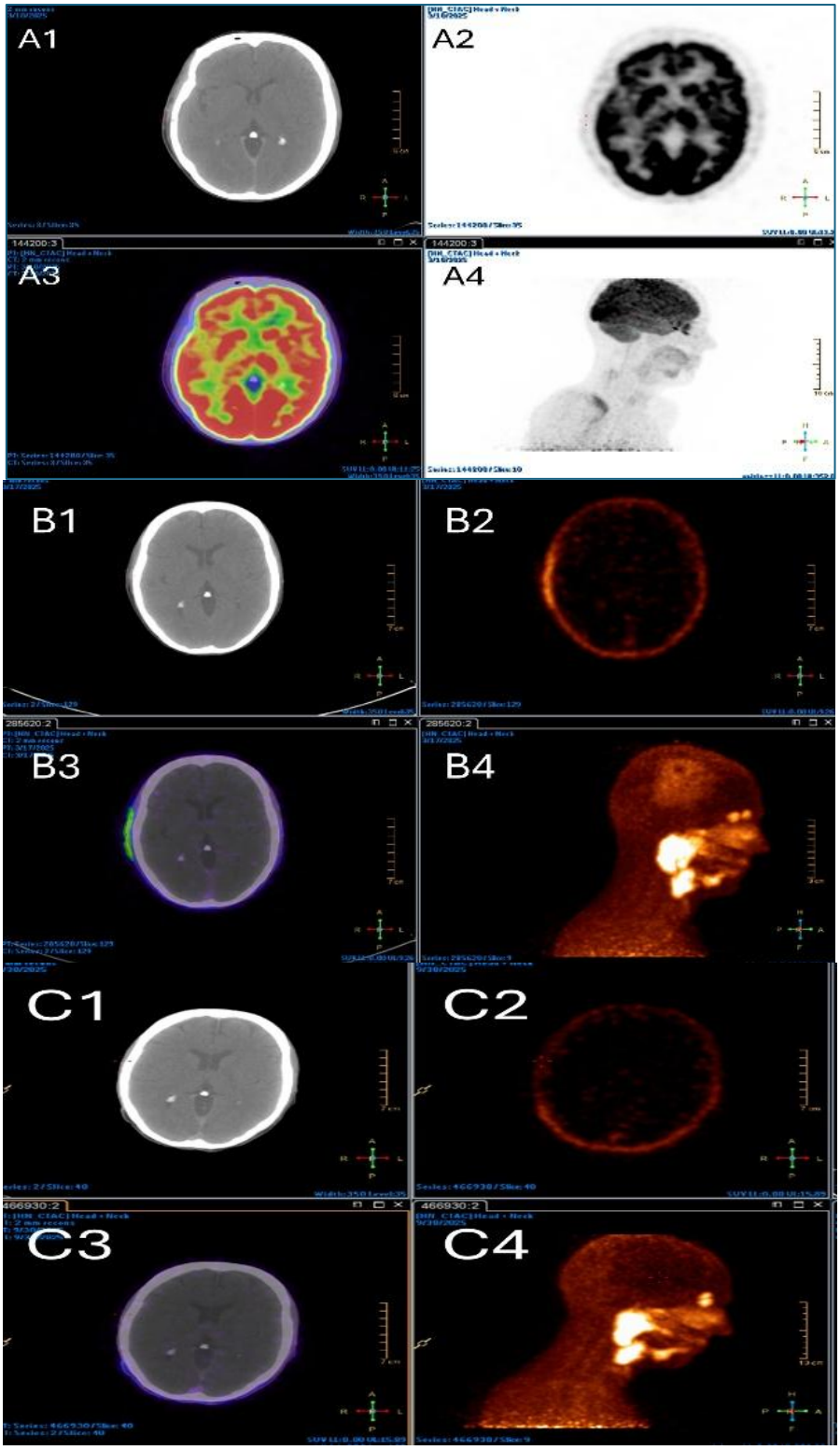

Figure 3 (A-C): Pre-and post-operative nuclear medicine imaging. A: Selected images from the staging ^{18}F-FDG PET/CT showing no significant FDG uptake in the primary tumour. 1) Low dose axial CT, 2) Axial PET images alone, 3) Fused axial PET-CT images and 4) Sagittal maximum intensity projection. B: Selected images from the staging ^{68}Ga-PSMA-PET/CT showing moderately increased tumour related tracer uptake in right fronto-parieto-temporal scalp region: 1) Low dose axial CT, 2) Axial PET images alone, 3) Fused axial PET-CT images and 4) Sagittal maximum intensity projection. C: Selected images from the post-therapy ^{68}Ga-PSMA-PET/CT showing treatment response with no residual uptake in right fronto-parieto-temporal scalp region. 1) Low dose axial CT, 2) Axial PET images alone, 3) Fused axial PET-CT images and 4) Sagittal maximum intensity projection.

DISCUSSION

ACC is a rare neoplasm that arises mainly in salivary glands of the head and neck and represents approximately 1 percent of all head and neck tumours. Unfortunately, ACC has a predilection for perineural invasion and recurrence. Distant metastases to lung, bone and liver are the leading cause of mortality. The 10-year survival rate for ACC is between 37 to 65 percent.[6] PCACC is a rarer variant with approximately 200 reported cases in the literature.[7,8] PCACC is considered locally infiltrative, with local recurrence occurring in over 50% of cases following treatment, however metastatic spread is rarely observed.[7,8] Due to its rarity, data on the incidence, clinical behaviour and management of PCACC are largely limited to published case reports.

PSMA is a type II transmembrane glycoprotein of the prostate secretory acinar epithelium and is upregulated in prostate carcinoma and its metastasis.[9] Highly specific ligands for PSMA have been developed, and for imaging purposes can be labelled with radioisotopes such as Gallium-68 or Fluor-18.[9] For prostate cancer ^{68}Ga-PSMA-PET CT significantly identifies more tumour lesions than other imaging modalities such as ^{18}F-FDG-PET.[9] The application of PSMA radioligand therapy is also typically guided by the ability to detect PSMA-positive tumours via PET-CT.[9]

While PSMA-PET imaging is well-established in the management of prostate cancer, its application is expanding due to the recognition of PSMA expression in the neo-vasculature of a wide variety of non-prostate tumours.[10-12] For example, the role of PSMA/PET-CT in the diagnosis and restaging of advanced renal cell carcinoma is being assessed in several early-stage trials.[13] Studies are also currently examining the role of PSMA radioligand therapy in the treatment of high-grade gliomas, since PSMA is also highly expressed in the neo-vasculature of these tumours.[6]

More than 90% of recurrent or metastatic ACC tumours are detected on PSMA-PET CT, with histology demonstrating greater than 50% of primary tumour cells and up to 92% of metastatic cells expressing PSMA.[14] This offers potential for alternative targeted therapeutic approaches such as PSMA radioligand therapy, particularly since the response rate to chemotherapy and targeted inhibitors (e.g. anti-VEGF) is suboptimal in the setting of progressive/metastatic disease.[6] Studies also report the application of PSMA-PET CT in the staging and restaging of metastatic ACC.[14]

To the author's knowledge we report the first case of PCACC demonstrating avidity on PSMA PET/CT, when initial conventional imaging with ^{18}F-FDG PET/CT imaging was negative. Follow-up PSMA PET/CT imaging in this case will therefore enable accurate assessment of any local recurrence, which is common in this variant of ACC, and assess for any potential distant metastasis.

CONCLUSION

In conclusion, ^{68}Ga-PSMA-PET/CT may be a useful investigation in the workup of PCACC, including staging and identification of residual or recurrent disease on follow up scans, particularly when conventional imaging modalities such as ^{18}F-FDG PET/CT are inconclusive. ^{68}Ga-PSMA-PET/CT could also have implications for targeted radioligand therapies in PCACC, especially in cases where treatment options are limited. Further research is warranted to histologically confirm PSMA expression in PCACC and to explore these potential clinical applications.

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