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Basal cell adenocarcinoma of parotid with metastases to lung and thigh

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Abstract

Background: Basal cell adenocarcinoma (BCAC) of parotid is a rare malignancy with extremely rare citations of distant recurrences in medical literature.

Case description: A 76 year old male presented with swelling on the left parotid region for 1.5 years. He underwent superficial parotidectomy and the specimen showed Basal cell adenocarcinoma (BCAC) of parotid. He was treated with adjuvant radical radiotherapy and he developed lung and thigh metastases (proven by fine needle aspiration cytology) nearly 3 years after radiotherapy. Palliative radiotherapy to metastatic sites offered some relief but eventually there was disease progression.

Conclusion: This is the first case of BCAC presenting with lung and thigh metastases simultaneously. Oncologists should be aware of this rare clinical entity.

Keywords

basal cell adenocarcinoma of parotid, lung and thigh metastases, rare

Introduction

Basal cell adenocarcinoma (BCAC) is a rare malignant tumor of the salivary gland that was included in the 1991 World Health Organization (WHO) classification [1] . Due to rarity of incidence only limited information is available from case reports. It is characterized by minimal capsular invasion at places in its periphery conferring a low invasive and malignant potential [2] . Hence metastatic recurrences after surgery and post-operative radiotherapy have been reported to be exceedingly rare [3]. We present here a case of BCAC of left parotid gland in an elderly male who presented with metastases to lungs and thighs nearly 3 years after post-operative radiotherapy.

Case Description

A 76 year old male from the outskirts of Chandigarh presented, with an insidious onset lump in the left parotid region slowly increasing in size for the last 1.5 years. There was no associated history of pain / dysphagia or any neural palsy. Fine needle aspiration cytology (FNAC) from the lump pointed to a diagnostic dilemma over Basal cell adenoma (BCA) vs. BCAC. Contrast enhanced computed tomography (CECT) neck in August 2013 showed a heterogeneously enhancing soft tissue lesion in superficial lobe of left parotid measuring 3.6 X 2.8 X 4cm (Antero-posterior X Transverse X Craniocaudal) and causing

external contour bulge (Figure 1a). Medially it had indistinct fat planes with left masseter muscle. Deep lobe of parotid gland and neurovascular bundle were normal. There was no significant cervical lymphadenopathy. Other structures in CT slices were also normal. He underwent superficial parotidectomy in February 2014 at the department of ENT in PGIMER, Chandigarh. The intraoperative findings revealed a firm lesion with cystic component in the superficial lobe of parotid gland measuring 8 X 6 cm in size.

Histopathological analysis of the specimen showed jigsaw-puzzle like infiltrative lobules of tumour cells tumor cells arranged in solid cords, clusters and islands separated by abundant eosinophilic extracellular basement membrane like material. The cells were characterized by coarse chromatin, inconspicuous nucleoli and scanty cytoplasm. Focal areas of capsular and perineural invasion were also detected (Figure 2a). On immunohistochemical analysis, the tumor cells stained positive for CK5/6, SMA and BCL2 but negative for EMA (Figure 2b). A diagnosis of solid type of basal cell adenocarcinoma of left parotid was given. Subsequently he was referred to our radiotherapy clinic for adjuvant radiotherapy.

We treated him with radical radiotherapy by 3-D conformal technique to a dose of 60 Gy in 30 fractions over 6 weeks. Following treatment completion in May, 2014 he was kept on periodic follow-up on OPD basis. He was in complete clinical remission till December 2016 when he developed firm, fixed and slightly painful swellings over anterolateral and medial aspects of right thigh. FNA smears from thigh lesions showed large clusters of basaloid cells with high nucleo-cytoplasmic ratio, inconspicuous nucleoli and scanty cytoplasm (Figure 2c and 2d). The features pointed to a diagnosis of metastatic BCAC. Whole body CECT was done in February 2017 for evaluating metastatic spread of the disease and multiple heterogeneously enhancing soft tissue lesions were noticed in bilateral lungs, suggestive of lung metastases (Figure 1b). Additionally, heterogeneously enhancing hypoechoic mass lesions were detected in right thigh (13.2 X 5.7 cm in anterolateral aspect and 8.5 X 4.2cm in medial aspect) (Figure 1c).

The general condition of the patient was not fit for palliative chemotherapy; hence palliative radiotherapy to a dose of 20Gy in 5 fractions was given to the painful thigh lesion and largest lung lesion. The lesions regressed partially after radiotherapy. His last follow-up on 18th April, 2017 showed evidence of disease progression as increase in size of the larger lump on right thigh to 14 X 9cm (Figure 3). The poor prognosis of the disease has been explained to the patient and his care-givers and the patient is being planned for further palliative radiotherapy to the new painful sites. The patient has, however remained loco-regionally disease free since radiotherapy completion as confirmed in the latest CECT neck on 21st April, 2017 (Figure 1d).

Discussion

Prior to its present day nomenclature, BCAC was reported under various names like malignant basal cell tumor, basaloid salivary gland carcinoma, atypical monomorphic adenoma, hybrid basal cell adenoma, etc. It received the status of a distinct entity in WHO classification after the seminal work of Ellis and Wiscivitch on the clinicopathological features of 29 cases of this tumor [4] . BCAC comprises 1.6% of all salivary gland neoplasms and 2.9% of malignant salivary gland neoplasms [5]. Nearly 90% of the cases arise from the parotids and rest are occasionally observed in submandibular and minor salivary glands [6]. In the study by Jung et al on basal cell adenomas, the median age group of presentation was reported to be 60.6 years (range 51-79 years) and all cases of BCACs were located in the parotid gland

with a predilection for left side (6:2) [7]. Patients usually present with salivary gland enlargement with mild or no pain [8]. The index case also presented in mid 7th decade with a painless progressive enlargement of his left parotid gland.

The histology and cytomorphology of BCAC is quite similar to its benign counterpart basal cell adenoma (BCA) except for the features of capsular and perineural invasion that makes it more aggressive than the later [9]. BCAC exhibit a mixture of growth patterns like tubular, trabecular, solid, membranous, and cribriform, out of which solid (62.5%) and cribriform (25%) are predominantly observed [7]. The histopathology in index case also showed the solid pattern of growth. Although a limited immunohistochemistry panel was used in our case as the histopathological features were strongly suggestive of BCAC, the detection of SMA positivity confirms the myoepithelial differentiation that is frequently associated with this tumor [7]. Even the FNA smears from the thigh lesions show cohesive sheets of basaloid tumor cells with hyperchromatic nuclei again confirming them to be sites of metastatic BCAC [7].

Most studies recommend wide local excision (WLE) for the tumor and neck dissection is only indicated in cases of significant cervical lymphadenopathy. Adjuvant radiotherapy is usually given in cases of positive/close margin status or in recurrent cases [2,10,11]. However, superficial parotidectomy was done in our case owing to the substantial size of the tumor. Additionally, we delivered postoperative radiotherapy as capsular and perineural invasion are traditionally considered to be high risk factors for local recurrence. The absence of loco-regional recurrence testifies to the benefit of our approach.

Although local recurrences have been described, only one case of distal recurrence in lung has been reported till date [4]. Our case is the second case of lung metastasis from BCAC of parotid. Additionally, the presence of thigh muscle metastases is the first of its kind to be reported in literature. These features make this case truly unique and warrants reporting as a new clinical entity.

Figures

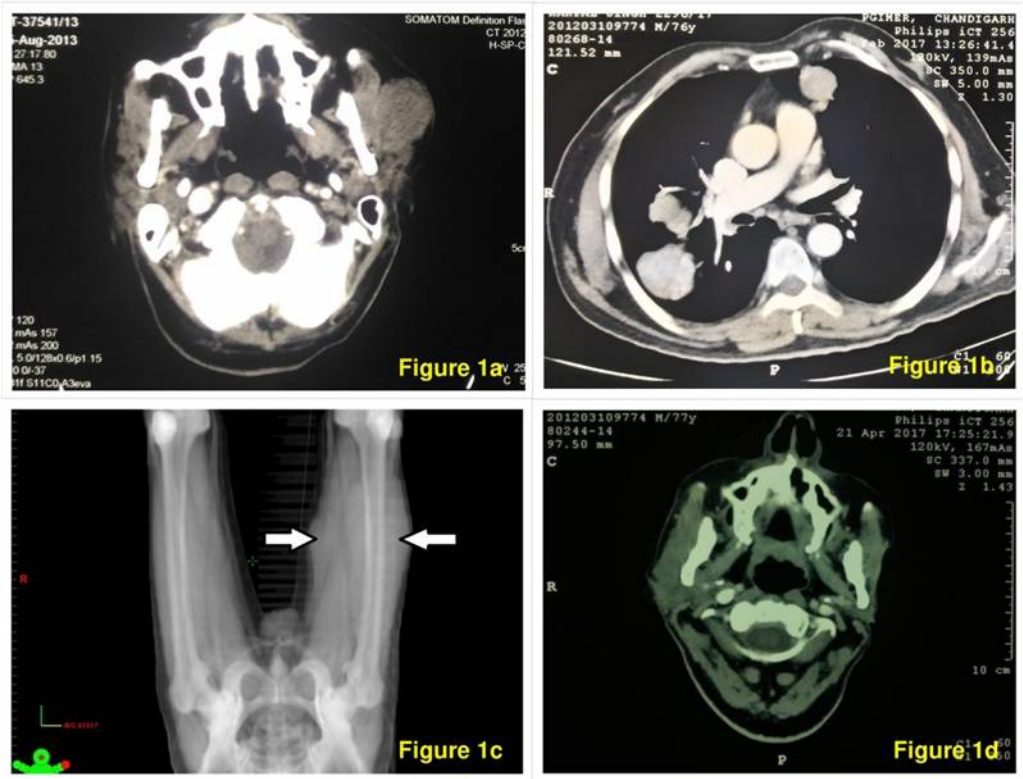

Figure: ***Figure 1a:*** Preoperative CT image of parotid tumor; ***Figure 1b:*** CT slice showing lung metastases; ***Figure 1c:*** CT of limbs (thigh metastases shown by white arrows); ***Figure 1d:*** Latest CT image showing no evidence of recurrence in primary site.

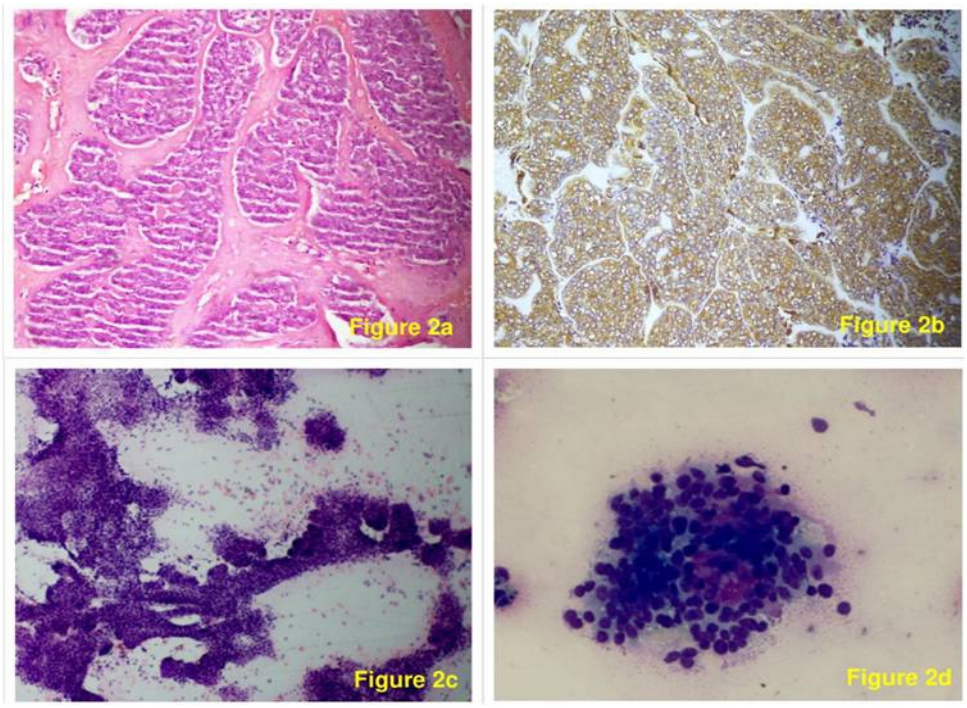

Figure 2: ***Figure 2a:*** Photomicrograph of jigsaw-puzzle like infiltrative lobules of tumour cells in basal cell adenocarcinoma (H&E, x400); ***Figure 2b:*** Photomicrograph showing CK5/6 positivity in tumour cells (CK5/6 immunostain, x200); ***Figure 2c:*** Smears showing cohesive sheets of tumor cells with peripheral palisading as well as singly scattered cells in the background. H&E; 20X. ; ***Figure 2d:*** Aggregate of basaloid tumor cells associated with eosinopihilic globule. Cells are of high N:C ratio with mildly pleomorphic hyperchromatic nuclei, inconspicuous nucleoli and scanty cytoplasm. MGG; 200X.

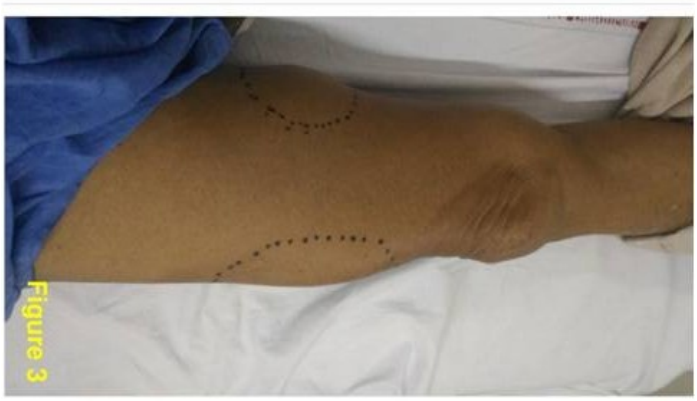

Figure 3: Photograph of right thigh of patient with metastatic lumps encircled with blue dots marked on skin

Conclusion

Being a rare entity the knowledge on clinical behaviour of BCAC has been derived mostly from case reports/series. It is usually considered to have a favourable prognosis owing to its low malignant potential. However, this report serves to remind that once a while BCAC may behave atypically and present with extensive distal recurrences despite adequate loco-regional control. Hence, oncologists should remain vigilant about the existence of aggressive BCAC, should there be another such case encountered in clinical practice.

Acknowledgements

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