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Case Report

Pathology Section

Two Unusual Cases of Acinic Cell Carcinoma: Role of Cytology with Histological Corelation

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ABSTRACT

Acinic Cell Carcinoma (AcCC) is a slow growing, malignant tumour of salivary glands, predominantly found in parotid gland and rarely in submandibular gland or minor salivary glands. Rarely, the tumour can arise in Heterotopic Salivary Gland Tissue (HSGT) and can present bilaterally. Synchronous contralateral tumour or tumour arising in HSGT is easily missed clinically. Fine Needle Aspiration Cytology (FNAC) is an important preoperative diagnostic investigation in cases of AcCC. Sometimes its diagnosis on cytology is very difficult and it is easily misdiagnosed as benign, affecting the long term prognosis. Here, we present two unusual cases of AcCC. One developed in HSGT and the other was synchronous bilateral. Though the histological features of AcCC appear to be characteristic but clinical suspicion and cytological features have been described as equally important for preoperative diagnosis. Present cases highlight the importance of a cytopathologist who plays an important role in its preliminary diagnosis.

Keywords: Fine needle aspiration cytology, Heterotopic salivary gland tissue, Parotid gland, Salivary gland

CASE REPORT

Case 1

A 32-year-old female patient came for FNAC of a gradually increasing, painless, right sided, submandibular lump with a clinical diagnosis of submandibular lymphadenopathy [Table/Fig-1a]. On examination, the lump measured 1.0x1.0 cm, was firm, non tender and easily rolled over the mandible.

FNAC smears of the lump were markedly cellular with cells present in microacinar grouping. The cells had abundant finely vacuolated cytoplasm, minimal anisokaryosis and bland nuclear chromatin. Many stripped nuclei were seen in the background [Table/Fig-1b,c]. The case was diagnosed as Acmic Cell Carcinoma (AcCC) and advised for histopathological examination.

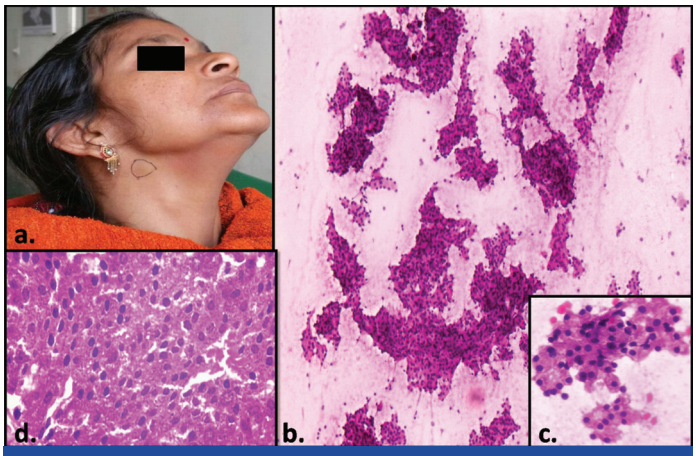
Grossly, excised specimen measured 1.5x1.0x0.6 cm with greyish cut surface. Histopathologically, a solid tumour composed of cells having basophilic finely granular cytoplasm and centrally placed bland nuclei with minimal cytological atypia was seen surrounded by fibro-fatty tissue [Table/Fig-1d]. No normal salivary gland or lymphoid tissue was seen. Patient was inquired for presence of any lump in the parotid or other salivary glands, but clinically and radiologically, no lump was detected. The case was diagnosed as primary AcCC, most probably developed in HSGT.

Case 2

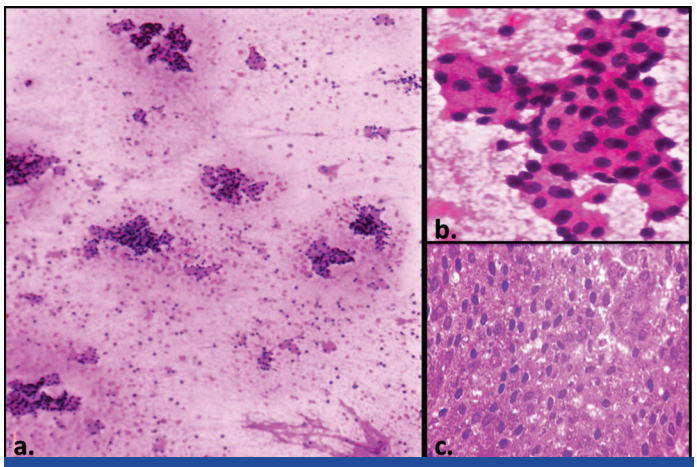
A 35-year-old female came for FNAC of gradually increasing, painless left retromandibular lump measuring 3.5x3.5 cm. Another very small lump was detected below her right ear lobule during examination. Both lumps were immobile and firm in consistency. FNAC smears from both the lumps showed similar pictures and the features were suggestive of AcCC. Both tumours were proven as bilateral synchronous AcCC of parotid glands on histopathology after surgical excision [Table/Fig-2a-c].

DISCUSSION

AcCC is a rare tumour of salivary gland, usually encapsulated and small in size measuring less than 3 cm. It is more commonly found in males in third decade with a rare familial preponderance. Cytogenetic alterations like 6q rearrangement, loss of Y chromosome and gain of chromosome 7 and 8 are related to



[Table/Fig-1]: Case 1. (a) Clinical image of case 2 showing right submandibular lump. (b) FNAC smear of AcCC showing richly cellular tumour with microacinar grouping of cells (H&E x 100). (c) Higher magnification of FNAC smear showing tumour cells with abundant finely vacuolated cytoplasm and bland nuclei (H&E x 400). (d) Histopathology of same tumour showing solid pattern of growth, composed of cells having basophilic cytoplasm and centrally placed monotonous nuclei (H&E x 400).



[Table/Fig-2]: Case 2. (a) FNAC smear of AcCC showing tumour cells with microacinar grouping (H&E 10X). (b) Higher magnification of FNAC smear showing tumour cells with abundant finely vacuolated cytoplasm and bland nuclei (H&E 40X). (c) Histopathology of same tumour showing solid pattern of growth, composed of cells having basophilic granular cytoplasm and centrally placed monotonous nuclei (H&E 40X).

occurrence of familial AcCC [1]. Other predisposing factor is radiation exposure [2].

AcCC in Heterotopic Salivary Gland Tissue (HSGT) is an extremely rare occurrence and very few such cases are reported till now. Heterotropia refers to the presence of supernumerary normal structure in an abnormal location. HSGT usually comes in attention when there is a large amount of tissue presenting as a mass, development of tumour within the tissue or presence of a duct leading to drainage of clear or mucoid fluid. HSGT can be found in medullary portion of a lymphnode or an extranodal site. Extranodal heterotropia can be high or low type. In high type of heterotropia, salivary gland tissue is usually found in mandible, ear, palatine tonsil, mylohyoid muscle, pituitary gland or cerebellopontine angle, resulting from defective embryonic migration of salivary gland. Lower type of heterotropia is associated with branchial pouch anomaly. Pathological changes including benign and malignant neoplasm can develop in HSGT. Intranodal Salivary Gland Tissue (ISGT) is found either in the form of mature acini, intercalated ducts, intralobular ducts or immature acini or immature small ducts with a potential for proliferation and differentiation [3]. Warthin's tumour is the most common tumour found in HSGT. Aboobakker et al., reported a case of large painless swelling of lateral neck which turned out to be an extraparotid Warthin's tumour [4]. They also mentioned that the extraparotid Warthin's tumour can arise in the cervical lymph nodes. Danial et al., reported 15 cases of Heterotopic salivary tumours where Warthin's tumour was the most common benign tumour followed by pleomorphic adenoma [5]. Among the malignant tumours of HSGT, mucoepidermoid carcinoma was the most common one followed by acinic cell carcinoma and adenocarcinoma. Manganaris et al., reported four cases where benign salivary gland neoplasms were developed in the HSGT [6]. Three of them were Warthin's tumour and one was pleomorphic adenoma. Minic AJ reported a case of AcCC in a 37-year-old women, that arose from HSGT present in the intra-parotid lymph node and also stressed upon the awareness of possible malignant alteration in HSGT [7]. Our first case of AcCC arose from an extranodal high type of HSGT and is an extremely rare occurrence.

Cytologically, cell rich smear is seen with microacinar groups composed of tumour cells, having abundant finely vacuolated or occasionally dense oncocyte like cytoplasm with rounded nuclei, bland nuclear chromatin and mild to moderate anisokaryosis with many stripped nuclei in background. Cytological diagnosis is difficult due to low cellular yield in papillary cystic variant, resemblance with benign salivary gland tissue or tumours having oncocytic or clear cell component. Histologically, AcCC shows solid, microcystic, follicular and papillary cystic pattern. Tumour cells have typical basophilic granular cytoplasm, monotonous bland nuclei and rare mitotic figures. Other rare cell types are intercalated duct, clear cell, vacuolated cell and non specific glandular type.

Patient of AcCC should be followed-up for a prolonged period with attention to the contralateral gland [8]. Clinical and radiological examination should be done to find synchronous contralateral tumour because detection of contralateral tumour is important for its management and prognosis. Synchronous contralateral tumour was detected in our second case which was an unique presentation and could easily be missed without having clinical suspicion.

AcCC has a lower incidence of regional lymph node and distant metastasis and usually treated with complete surgical excision followed by radiotherapy, if resection margin comes positive. The tumour is not diagnosed radiologically because of its radiological resemblance with benign tumours but radiological imaging are used for tumour size evaluation, location, extant, relation to facial nerve and lymphnode metastasis [2]. It shows a favourable prognosis after complete resection. Poor prognostic features are blood vessel involvement, invasion to adjacent tissue, undifferentiated histological pattern, facial nerve involvement and lymphnode metastasis [9]. Our patients didn't show any sign of recurrence or metastasis since one year of follow-up.

CONCLUSION

AcCC is a rare, slow growing malignant tumour of parotid gland, can rarely arise in submandibular or minor salivary gland or extremely rarely in HSGT. Diagnosis of AcCC is important on FNAC because cytopathologists play an important role in its preliminary diagnosis and it is easily missed clinically, radiologically and cytologically.

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