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Acinic Cell Carcinoma of the Parotid Gland: A Rare Case

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Abstract

Acinic cell carcinoma of parotid gland is defined by parotid neoplasm with at least some serous acinous differentiation. It is one of the rare malignancies of parotid (3–11% of adult parotid neoplasm) with very unpredictable clinical behavior in nature. Close differentials are warthins tumor, oncocytoma, clear cell and mucoepidermoid carcinomas. Surgery is the standard of care in this case.

A 22-year-old male patient developed a swelling over the left parotid region in 2018, which gradually increased in size without any difficulty faced to the patient. Fine needle aspiration cytology of the swelling revealed pleomorphic adenoma. The patient underwent surgery in 2018 under general anesthesia at another facility, and no post-operative documents were available with the patient. Over time, the swelling recurred in the same region. The patient sought medical attention at IGIMS, ENT department, where fine needle aspiration cytology of the swelling revealed pleomorphic adenoma once again. Magnetic Resonance Imaging showed a heterogeneous mass involving both the superficial and deep lobes of parotid. Subsequently, the patient underwent left parotidectomy for the same condition. The post-operative biopsy and immunohistochemistry suggestive of acinic cell carcinoma with cystic and solid patterns. After confirming the absence of metastasis with PET scan, radiation therapy was recommended due to the recurrence involving the deep lobe.

Acinic cell carcinoma is initially considered benign but later reclassified as malignant due to its metastatic and recurrence capability. It commonly affects parotids (85% cases) and constitutes 6-16% of major salivary gland malignancies. Its clinical course is uncertain, with better prognosis in pediatric cases. Family history, radiation exposure, and certain genetic transformations are risk factors. Radiological assessments are inadequate for characterization many times. Surgical resection is the main treatment modality. Chemotherapy is ineffective, but adjuvant radiation can be considered. Recurrences are common, but prognosis is generally good. Correct diagnosis is crucial for effective treatment and recurrence prevention.

Keywords: Acinic cell carcinoma • Parotid gland • Pleomorphic adenoma

Introduction

Acinic Cell Carcinoma (ACC) is uncommon salivary gland tumor with incidence of 1 in 100000 populations. ACC is commonly seen in 5^{th}–6^{th} decade of life and has very unpredictable clinical behavior. This type of carcinoma is often slow-growing and in more than 85% of cases found in the parotid gland, which is the largest salivary gland located near the ear.

Symptoms of acinic cell carcinoma may include a painless lump in the face or neck, difficulty swallowing, facial weakness, or persistent pain in the area. Treatment typically involves surgery to remove the tumor, and in some cases, radiation therapy may be recommended. Tumor involving deep lobe of parotid will require post-operative radiotherapy because of adverse pathologic features such as limitation of surgical margin in resection of tumor.

Mostly patients present with well circumscribed painless mass in parotid region which is often solid in nature. Facial nerve involvement could be there. They have a high tendency for distant hematogenous spread, although positive neck nodes could be found in up to 43% of cases.

Case Presentation

A 22-year-old male patient developed swelling over the left parotid region in 2018, gradually increasing in size. It was not associated with any difficulty in mastication, pain, deviation of face, or discharge from swelling. There is no any history of anorexia or weight loss. Personal, medical and family history were not significant. Fine Needle

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Aspiration and Cytology (FNAC) done from the swelling site shown pleomorphic adenoma. Patient underwent surgery in 2018 outside, however no post-operative documents were available. The swelling recurred gradually over time over the same region. Patient visited IGIMS, ENT department (2023) and FNAC was done from the swelling site which showed pleomorphic adenoma yet again. Magnetic Resonance Imaging revealed 5.4 × 3.9 × 4.1 cm (AP × TR × CC) sized heterogeneous mass involving the superficial as well as deep lobes suggestive of recurrence with neurovascular bundles seen posterior to lesion (Figure 1).

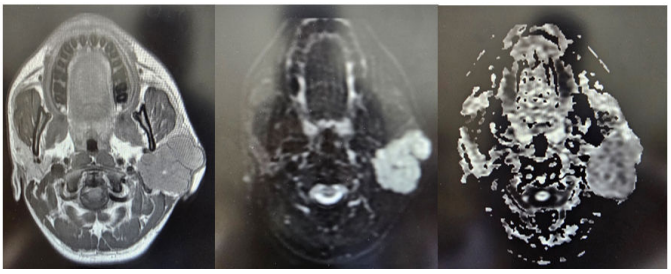

Figure 1. Pretreatment MRI sequences showing left parotid mass.

Patient underwent left total parotidectomy for the same under general anesthesia. Post-operative biopsy report revealed features suggestive of Acinic cell carcinoma with cystic and solid patterns. The tumor measured 6 × 4 × 3 cm. Immunohistochemistry analysis Figure showed cytoplasmic and membranous positivity for DOG-1 in tumor cells, while CD 117 and HER2neu were negative. Ki67 demonstrated 4%positivity in tumor cells (Figure 2).

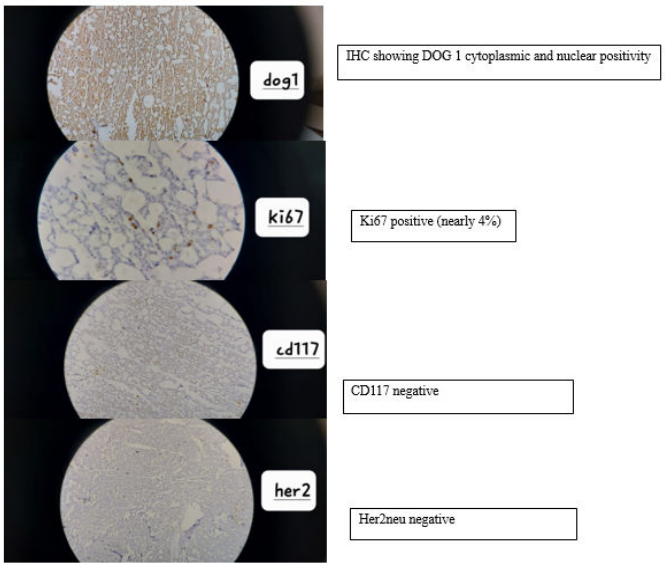

Figure 2. Immunohistochemistry analysis showed cytoplasmic and membranous positivity for DOG-1 in tumor cells, while CD 117 and HER2neu were negative. Ki67 demonstrated 4% positivity in tumor cells.

Sensorineural and cranial nerve examination revealed no abnormalities. Patient is asymptomatic at present. Whole body PET scan was advised for metastatic workup. After confirming by the whole body PET–C, it is non-metastatic disease, radiation therapy was advised in view of recurrent disease involving deep lobe (Figure 3).

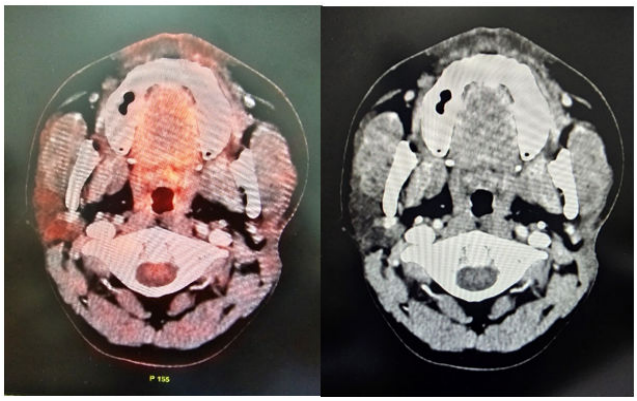

Figure 3. Post left parotidectomy PET CT scan showing no residual mass or uptake.

Discussion

Acinic cell tumor were initially considered as benign tumor [1]. It was in 1953 that Bruxton et al. Re-categorized it as malignant disease because of the capability of metastatic and recurrence. Among salivary glands, most commonly it involves parotids (85% cases) and constitutes 6-16% of all malignancies of major salivary glands. Female to male ratio is roughly 1.4:1, mostly presenting in 5^{th}-6^{th} decades of life [2]. Bilateral disease can be seen in up to 3% of cases [3-5]. They have highly uncertain clinical course with pediatric disease having better prognosis [6]. Family history and previous radiation exposure can be considered as risk factors [7]. Smoking and certain genetic transformation (involving chromosome 6, 7, 8, Y and Rb gene) are related to the development of disease [8-10]. This rare tumor commonly presents as well circumscribed solid mass often not painful and defined near the tail of parotids in up to 50% of cases [11,12]. Facial nerve involvementis controversial (but can trace the nerve up to stylomastoid foramen and above) and it usually does not involve lymph nodes [13-17]. Radiological assessment using ultrasound, CT, and MRI are usually inadequate for proper characterization because of their resemblance to other benign and malignant tumors [18-19]. Cytopathological characterization is usually difficult and final diagnosis is made correlating MRI findings and immunohistochemistry [20]. Adequate surgical resection along with or without neck dissection is the main modality of treatment.

Acinic cell carcinoma is usually resistant to chemotherapy. Adjuvant radiation can be considered in cases of recurrence, positive margin, large size tumor >4 cm, facial nerve, deep lobe and lymph node involvement. Recurrences are common but overall prognosis is good. Five-year disease specific survival is more than 90%. All these factors warrant long term vigilant follow up.

The present case report is about a young male who might had misdiagnosed ACC as benign pleomorphic adenoma outside. Due to the disease nature and probable inadequate surgery, patient landed at our centre after 4-5 years with recurrent symptoms. Beside a painless solid mass in parotid region, patient was apparently well with no facial nerve involvement or lymphadenopathy. The MRI findings of deep parotid lobe involvement with controversial cytopathology revealing pleomorphic adenoma further raised the diagnostic dilemma. Patient was taken for resurgery, and postoperative histopathology and IHC was

confirmatory of acinic cell carcinoma. Deep lobe involvement in case of recurrent tumor warrants post-surgery radiotherapy in this case.

Conclusion

Although rare, but vigilant evaluation and correct diagnosis of acinic cell carcinoma differentiating it from other benign and malignant disease is important for proper treatment and avoiding recurrence.

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