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Case Report

Resection of Salivary Duct Carcinoma ex Pleomorphic Adenoma of Minor Salivary Gland of Parapharyngeal Space without Mandibular Osteotomy – A Case Report

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Abstract

Parapharyngeal space (PPS) tumors offer significant surgical challenge owing to their rarity, difficult anatomy and presence of vascular structures nearby. Although most of the tumors in the prestyloid compartment of PPS are benign, malignant neoplasms have been reported infrequently. Pleomorphic adenoma, although most common benign salivary gland tumour, is rarely found to arise from the minor salivary glands of PPS. Salivary duct carcinoma ex pleomorphic adenoma arising from the minor salivary glands of the PPS has never been reported in English literature. Surgical excision remains the treatment of choice for these PPS tumors. *Trans* cervical approach is the most commonly adopted approach for these rare tumors. Although mandibular osteotomy improves access, is associated with significant long term morbidity. We are reporting a case of salivary duct carcinoma ex pleomorphic adenoma arising de novo from the minor salivary glands of the parapharyngeal space which was resected via transcervical approach avoiding a mandibular osteotomy.

Key words: Parapharyngeal space tumour; salivary duct carcinoma; Pleomorphic adenoma

Introduction

Parapharyngeal space (PPS) tumors are rare accounting for 0.5% of neoplasms of head and neck [1, 2]. Majority of these tumors are benign with pleomorphic adenomas being the most common histological subtype. Although pleomorphic adenomas in the PPS typically originate from deep lobe of the parotid, rarely they can develop de novo from displaced or aberrant salivary gland tissue within a lymph node [3,4]. Transoral, *Trans* cervical and *Trans* parotid approach are the 3 most common approaches to these PPS tumors depending on the size and the histology [5, 6]. Although transcervical approach is associated with lower morbidity it cannot be performed in all patients, especially for large tumors or suspected malignancies. Mandibular osteotomy significantly improves the exposure though it also increases the overall morbidity of the procedure. Salivary duct carcinoma (SDC) is a highly aggressive histological subtype of salivary gland malignancy. Although majority of these tumors arise de novo in major salivary glands, 10-27% have been found to originate in pre existing pleomorphic adenoma [7,8]. Salivary duct carcinoma ex pleomorphic adenoma in PPS has never been documented in English literature. We are reporting one such case which was resected via a transcervical approach while avoiding a mandibular osteotomy.

Case Report

Forty five year old gentleman presented with history of swelling in front of left ear of 3 months duration. On examination, he had 6 X 5 cm, firm swelling with smooth surface, below, behind and in front of left ear pushing the left ear lobe upwards (Figure 1a). Intra oral examination revealed medially displaced left tonsil and uvula. Facial nerve examination didn't reveal any deficits. Patient was investigated with the working diagnosis of parotid tumour. MRI of the left parotid area revealed 9 x 7 cm, dumbbell shaped mass in the pre styloid compartment of left PPS situated lateral to external carotid artery and inferior to superficial temporal artery (Figure 1b and 1c). Relationship with deep lobe of the parotid and facial nerve were not well defined because of the large size of the mass. FNAC from the mass was inconclusive.

Patient was planned for excision of the mass with the working diagnosis of benign tumour arising from the deep lobe of the parotid. Transverse incision was placed 2 cm below the base of the mandible and sub platysmal skin flaps were elevated. Parotid was separated from the underlying mass through blunt and sharp dissection. As the working space was limited, midline lip split incision was placed and cheek flap was elevated (Figure 1d). Styomandibular ligament was cut to lift the mandible superiorly to improve access to the mass. Tumour was separated from the styloid process superiorly and resected en bloc (Figure 2a and 2b). Intra

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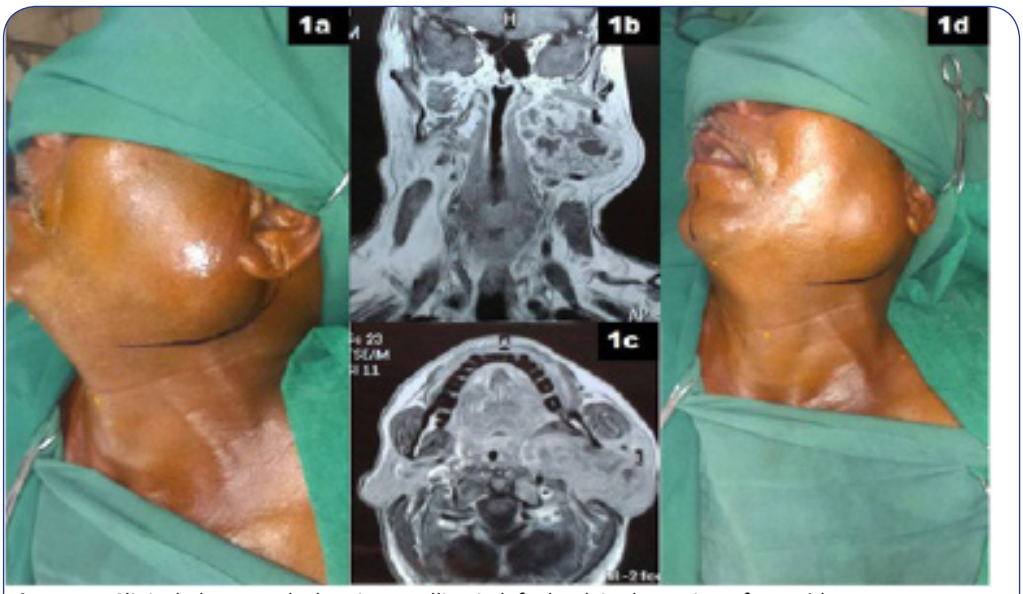

Figure 1a: Clinical photograph showing swelling in left cheek in the region of parotid.
Figure 1b: Coronal cuts on MRI showing the tumour extending superiorly upto medial pterygoid muscle with superficial temporal artery on the supero lateral aspect of the tumour.
Figure 1c: Axial cuts on MRI showing the tumour situated lateral to external carotid artery
Figure 1d: Transcervical sub mandibular incision to gain access to the tumour.

and post operative recovery was uneventful.

Histopathological examination revealed tumour composed of epithelial as well as chondrmyxoid stromal components along with a small focus of infiltrating malignant tumour composed of large, polygonal cells having eosinophilic cytoplasm and large, vesicular oval nucleus suggestive of salivary duct carcinoma ex pleomorphic adenoma (Figure 2c and 2d). All the resection margins were free. In view of aggressive nature of the histological subtype, patient was offered options of either adjuvant radiotherapy or close observation. Patient chose former and received external beam radiotherapy of 50 Gy over 5.5 weeks. At 3 months follow up, patient was disease free and healthy.

Discussion

Pleomorphic adenoma is the most common benign salivary gland tumour characterised histologically by the presence of epithelial and stromal components [1]. Although parotid gland is the most common site of origin, 5–10% of cases may originate from the minor salivary glands. The most common site of pleomorphic adenoma of the minor salivary glands is the palate followed by lip [2]. Pleomorphic adenoma arising de novo from the minor salivary glands in the parapharyngeal space are considered very rare.

SDC are aggressive malignancies which occur predominantly in the parotid glands of elderly men. Histologically, SDC is characterized by presence of intra ductal and infiltrating components. The

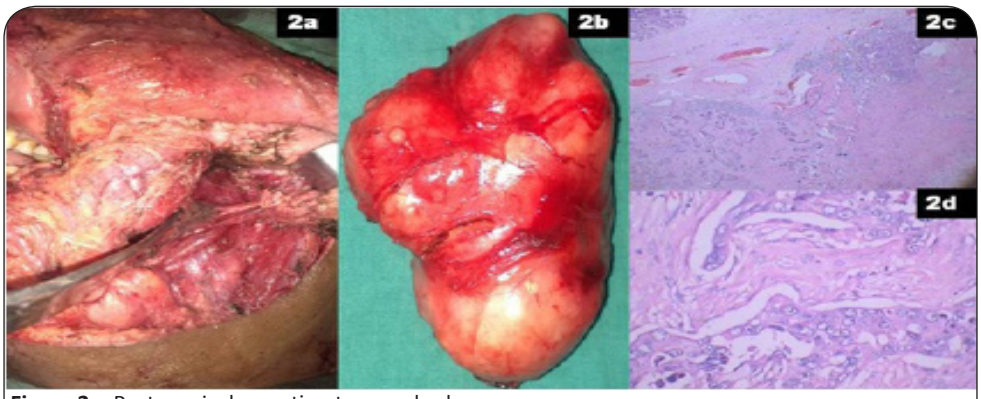

Figure 2a: Post surgical resection tumour bed.
Figure 2b: Gross examination of the resected tumour.
Figure 2c and 2db: Microscopic examination showing tumour composed of epithelial and stromal components along with malignant tumour showing comedonecrosis.

relative proportion of these two components varies among the patients. The intra ductal component is typically characterized by comedonecrosis. The reported incidence of nodal metastasis for SDC is 56–73% [7-9]. Such higher incidence of nodal involvement as well as frequent incidence of facial nerve paralysis point towards the aggressive nature and hence poor prognosis associated with these rare tumors. Although vast majority of the SDC arise de novo, upto 20% may arise from pre existing pleomorphic adenoma [7,8]. The present patient didn't give long standing history of swelling in the parotid area. This is probably related to the fact that pleomorphic adenoma was arising from parapharyngeal space and hence it was not apparent to the patient.

MRI is the imaging modality of choice for imaging the PPS [10]. T1-weighted images are ideal for demonstrating normal anatomy and any tumour-fat interfaces. Radiologically parapharyngeal space is divided into prestyloid and post styloid compartments by styloid process, tensor veli palatini muscle and its fascia. Prestyloid tumors displace the parapharyngeal space fat antero medially whereas post styloid space tumors displace it antero laterally. Differentiation between these 2 compartments is essential as it narrows down the differential diagnosis. Masses commonly encountered in the prestyloid PPS include pleomorphic adenoma or medial extension of a deep lobe parotid tumour whereas those encountered in post styloid PPS include carotid body tumors, or paragangliomas *or* schwannomas. Deep lobe parotid tumors are differentiated from ectopic salivary gland tumors by the preservation of a fat plane between the tumour and the parotid. MRI features suggestive of high grade malignant tumors of the salivary glands include low-to-intermediate signal intensities on T2 weighted images, ill-defined margin and infiltration into the adjacent tissue. The relative sensitivity and specificity of each of these features for the diagnosis is quite low, although when combined together, overall accuracy increases significantly. In the present case the fat plane was not demonstrated between the deep lobe of parotid and the tumour owing to the large size of the mass. As a result the working diagnosis at the time of surgery was tumour arising from the deep lobe of the parotid.

Although histopathology remains the gold standard, FNAC is usually performed pre operatively to aid in better counselling of the patients [11]. FNAC is non diagnostic in 25–60% of cases and is the result of lack of cellular material, excessive bleeding or technical problem in targeting the lesion adequately. Open *Trans* oral or *Trans* parotid biopsies are contraindicated not only because of the chances of tumour seeding but also because of the chances of injury to vital structures.

Surgical excision remains the mainstay of treatment for PPS tumors. *Trans* cervical, *Trans* parotid and *Trans* oral are the 3 most common surgical approaches adopted for the pre styloid compartment of PPS. Majority of these tumors can be resected via cervical, submandibular approach [12,13]. Mandibular osteotomy improves surgical access and provides greater working space for better vascular control. Mandibular osteotomy is favored for malignancies, highly vascular tumors, recurrent tumors and very large tumors particularly those reaching upto the skull base [14]. Although mandibular osteotomy improves surgical access, it increases overall morbidity of the procedure. Hughes et al. [15] in their series of 172 cases of PPS tumors, *trans* cervical and trans-parotid approaches were used in majority of the tumors with Mandibular osteotomy required in only 2% cases. In the present patient, tumor was resected via *trans* cervical, submandibular approach without mandibular osteotomy in spite of the large size with tumor reaching upto medial pterygoid. Transection of the stylomandibular ligament was the key in improving the access and avoiding a mandibular osteotomy.

The purpose of reporting this case was because it is the first reported case of salivary duct carcinoma ex pleomorphic adenoma originating denovo from minor salivary gland tumors of the parapharyngeal space and complete resection (R0) with negative margins achieved via *trans* cervical approach while avoiding a mandibular osteotomy.

Conclusion

Parapharyngeal salivary duct carcinoma ex pleomorphic adenomas are rare neoplasm which can be safely resected via *trans* cervical approach. Precise knowledge of the anatomy as well as clear definition of the relationship of the tumour with surrounding structures on MRI is the key to R0 resection. Division of the stylomandibular ligament during the *trans* cervical approach improves the surgical access and hence avoids a mandibular osteotomy.

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