

## Case Report

# A cutaneous representation of parotid adenocarcinoma not otherwise specified

B. M. Rudagi, Jay Goyal, Akshaya Subramanian\*

Department of Oral and Maxillofacial Surgery, ACPM Dental College & Hospital, Dhule, Maharashtra, India

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**\*Correspondence:**

Dr. Akshaya Subramanian,

E-mail: akshaya1396@gmail.com

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### ABSTRACT

Cutaneous manifestations of adenocarcinoma is a very rare entity. The patient reported to our institute with a complain of swelling on right parotid region that would bleed profusely merely on palpation. The hematological values revealed very low Hemoglobin, RBC and PCV counts indicating severe hemolytic anemia. The patient had palpable lymph nodes of level I and IIA which were enlarged. The patient underwent excision of the lesion along with modified radical neck dissection and reconstruction of the skin defect using pectoralis major myocutaneous flap.

**Keywords:** Parotid adenocarcinoma, Parotidectomy, Pectoralis major myocutaneous flap

## INTRODUCTION

Salivary adenocarcinoma, not otherwise specified, refers to salivary gland malignancies that do not satisfy the diagnostic requirements of other malignancies.<sup>1</sup> It is an aggressive, high-grade malignancy, with a predisposition for the parotid gland. It is characterized by cytologic and architectural diversity and an invasive growth pattern. Adenocarcinoma, not otherwise specified (ANOS) is not common among malignant salivary tumors, accounting for 4.3% to 17.8% of all malignant parotid gland tumors.<sup>1</sup>

We performed a search of the PubMed database over the past 10 years (2011-2021) and found around 620 case reports on adenocarcinoma. This search yielded only one such other case with a cutaneous representation. As per a retrospective study performed by Loxha et al, the tumor recurrence and metastasis rate in adenocarcinoma is 6.3%.<sup>2</sup>

Here we report a rare case of primary high grade ANOS arising from the parotid gland which was treated by surgery followed by radiotherapy.

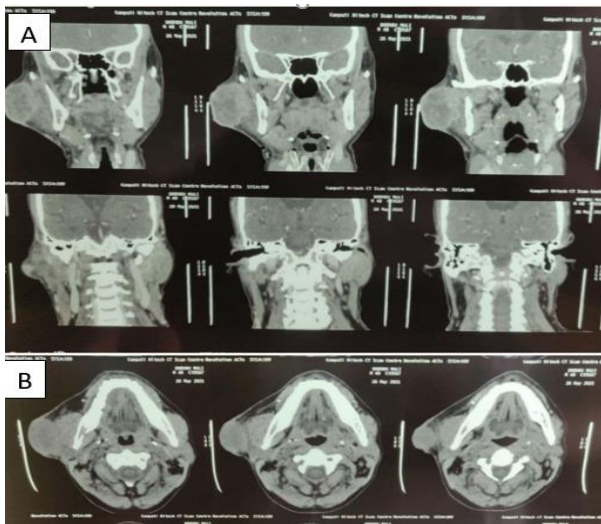
## CASE REPORT

A 48-year-old man reported to ACPM Dental College, Dhule with a chief complaint of swelling, over the right parotid region along with pain and bleeding on palpation since two months. (Figure 1) On examination, the size of the swelling was approximately 60×40×55 mm in size, fixed, painful, and circumscribed in nature, the overlying skin was ulcerated and would bleed profusely on palpation. The lesion was pea sized initially which increased gradually over two months to the current size. Computed tomography (CT) of the Paranasal Sinuses (PNS) was performed in order to gain more knowledge of the lesion. It showed enlargement of the superficial lobe of parotid gland and a soft tissue attenuation that measured 51×36×51 mm in size in the right masticator and parotid space. (Figure 2) The lymph nodes at the right submandibular area were enlarged. There were no signs of bony involvement. The provisional diagnosis was of a primary tumour of the salivary gland and staged at pT4 N2b M0 as per 8th edition of American Joint Committee Cancer Staging (2018). Mucin producing salivary duct carcinoma, mucoepidermoid carcinoma, hybrid carcinoma, polymorphous low-grade

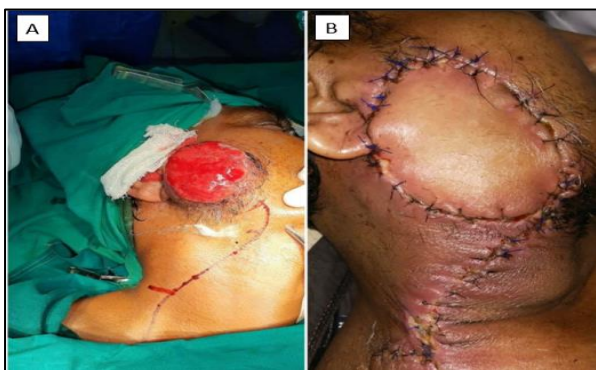
adenocarcinoma, undifferentiated carcinoma and metastatic carcinoma were all included in the differential diagnosis.



**Figure 1: (A) Frontal profile. (B) Right lateral profile.**



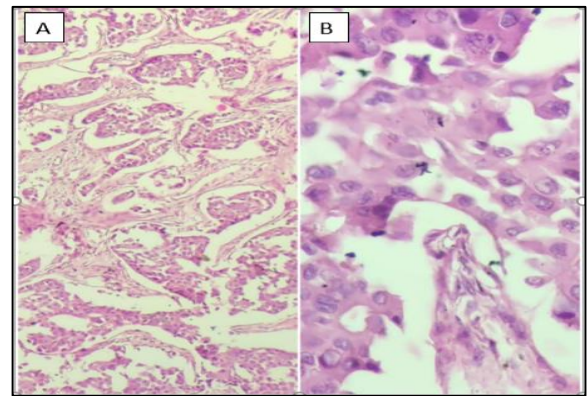
**Figure 2: CT PNS. (A) Coronal section. (B) Axial section.**



**Figure 3: (A) Pre-operative section. (B) Reconstruction with PMMC flap.**

Chest CT and Sonography of the abdomen and pelvis region were performed, which did not support the diagnosis of a primary lung or Gastrointestinal tumor. The patient went on to undergo radical surgery, which

included wide local excision, with right superficial parotidectomy, right modified radical neck dissection type 3, and reconstruction with a pectoralis major myocutaneous flap. (Figure 3) The results of the final histopathological examination was NOS type of Adenocarcinoma with evidence of aggressive features that included an infiltrative pattern in desmoplastic stroma, (Figure 4) with perineural and lymphovascular spread. The lymphnodes were encapsulated and completely replaced with tumor cells.



**Figure 4: Histopathologic specimen. (A) Low magnification. (B) High magnification.**

## DISCUSSION

Evans and Batsakis coined the term Adenocarcinoma Not Otherwise Specified (ANOS) which denotes a specific category of salivary gland tumor amongst a host of heterogeneous salivary gland tumors.<sup>3</sup> The incidence rate of ANOS in Oral and Maxillofacial area is less. The most common site of occurrence is the parotid gland followed by palate.<sup>4</sup>

Kevin et al had conducted a retrospective review of National Cancer Database from the year 1992-2012 in order to gain a better understanding of the Parotid Adenocarcinoma Not Otherwise Specified (PANOS). He found out that 10% of all the parotid cancers was PANOS. The commonly affected population were men with a high grade histology, which is consistent with this case report.<sup>5</sup>

Patients undergoing medical radiation or exposure to direct sunlight and UV light therapeutic treatment causing head and neck exposure have increased predisposition to adenocarcinoma. Alcohol, tobacco and smoke exposure have not been consistently associated with it's occurrence.<sup>6</sup> Similarly, the patient in this report had exposure to sunlight due to his occupation as a farmer, which could be a possible etiology for his disease.

Clinically, the carcinoma is poorly circumscribed in nature and has solid tan cut surface with hemorrhage and necrosis.<sup>7</sup> Clinically our patient showed a massive lesion

in the right parotid region with hemorrhagic characteristics.

The histopathological features typical of ANOS are Invasive with glandular or ductal differentiation. Glandular spaces with cyst formation, papillary formation, solid sheets, necrosis, hyalinized "shadow" nodules, Small clusters of cuboidal, round or ovoid cells with distinct borders and abundant cytoplasm, may have clear cell or oncocytic features are some of the commonly encountered patterns.<sup>7</sup> They may be classified as Low, intermediate or high grade based on cytomorphic features. The histopathologic report of the patient showed invasive epithelial tumor cells with high grade nuclear pleomorphism. There was evidence of lymphatic and perineural invasion.

Complete surgical excision is required for T1 and T2 salivary gland carcinomas and post operative radiation for cases with positive neck nodes, as per the guidelines of the National Comprehensive Cancer Network (NCCN) 2011: Head and Neck Cancers.<sup>8,9</sup>

Due to the highly invasive and aggressive nature of the ANOS, there are high chances of recurrence and distant metastasis. Brandon et al, reported a rare case of metastasis of Parotid Adenocarcinoma to breast.<sup>10</sup>

The patient underwent wide local excision of the lesion with clear tumour margins. Due to cutaneous involvement of the lesion, the skin had to be reconstructed by harvesting the Pectoralis Major Myocutaneous Muscle Flap (PMMC) for esthetic purposes. The histopathology report was suggestive of lymphatic spread that was treated by performing modified radical neck dissection. After considering several factors such as the recurrence rate, distant metastasis and following the guidelines provided by NCCN, the patient was subjected to Post operative Radiotherapy. Immunohistochemical staining showed positive for SMA, P63 and S100. A three year follow up of the patient revealed no recurrences or signs of distant metastasis and showed an overall good prognosis. The role of chemotherapy was not deemed to be important in this case, for the survival of the patient.

The overall survival rate of the patients were 5-10 years. Due to limited availability of literature, there is insufficient knowledge on the prognosis of the disease. Recurrence and secondary metastasis of the disease requires prolonged periods of follow up at due intervals.

## CONCLUSION

The lack of a standard systemic classification for treatment of salivary gland malignancies is due to it's rare occurrence. Our case throws light on the highly aggressive and invasive clinical nature of ANOS. The treatment plan adopted by us emphasizes the importance

of radiotherapy as an adjuvant therapy to wide local excision and neck dissection in order to avoid recurrences or distant metastasis. The clinicopathological features and the treatment modality described in this report forms a knowledgeable drop that will further push the boundaries in order to battle such a deadly disease.

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