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Sinonasal Minor Salivary Gland Carcinoma- Our Experience

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Abstract
Aim and objective*: The study aimed to evaluate the clinical profile and management of patients with minor salivary gland carcinoma of nose and paranasal sinuses.* ***Materials and Method:*** *A retrospective study was conducted in the department of otorhinolaryngology over a period of 5 years from year 2016 to 2020. Ten patients of minor salivary gland carcinomas of nose and PNS were found. The distribution of patients according to age, gender, primary site, histologic subtypes and surgical management along with their clinical profile were studied.* ***Results:*** *Patient's age ranged from 38-60 years with a mean of 51.5 years. There was a male preponderance seen in 80% cases. Out of these, 50% cases were ACC, 30% Adenocarcinoma(AC) and 20% MEC.* ***Conclusion:*** *Minor salivary gland malignancies of nose and PNS is an uncommon group. An elderly or middle aged male with unilateral obstruction should be seen with suspicion of malignancy and should be worked up to rule it out.*

Keywords: *Adenoidcystic carcinoma, Mucoepidermoid carcinoma, Adenocarcinoma, Minor salivary gland.*

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I. Introduction

Malignant tumours of the sinonasal tract account for 3% of all head and neck malignancy.[1] Minor salivary gland tumours account for only 9 to 23% of all salivary gland tumours and are malignant in 80% cases.[2] Minor salivary gland carcinomas are uncommon in sinonasal region following that of squamous cell carcinoma (40%).[3,4] Unlike the oral cavity where both Adenoidcystic carcinoma (ACC) and Mucoepidermoid carcionoma (MEC) are frequent, it has been reported that over 90% of salivary gland tumours in the sinonasal tract are ACC,[5] which is generally the more aggressive of the two.[6] Carcinomas of the sinonasal tract occur primarily in men aged 55 to 60 years.[7] Smoking and occupational exposure to nickel, wood dust, leather dust, chromium, aflatoxin, mustard gas, thorotrast, radiation and genetic causes are known to be the primary predisposing factor.[8] Surgery and radiotherapy remains the mainstay of treatment. We present here a series of ten cases of minor salivary gland carcinomas of nose and paranasal Sinus (PNS).

II. Material And Methods

A retrospective study was conducted in the department of otorhinolaryngology over a period of 5 years from year 2016 to 2020. Ten patients of minor salivary gland carcinomas of nose and PNS were found. The distribution of patient according to age, gender, primary site and histologic subtypes along with their clinical profile was studied. Tumour location and extent were determined using complete head and neck clinical examination and radiological imaging with contrast enhanced computed tomography (CT) and magnetic resonance imaging (MRI) of nose and PNS. Diagnostic nasal endoscopy was performed in each case. All patients underwent a proper histological diagnosis before undergoing surgical intervention. Open surgery was done in all cases following which patients were sent for chemoradiation.

III. Result

Patient's age ranged from 38-60 years with a mean of 51.5 years. There was a male preponderance seen in 80% cases. Out of these, 50% cases were ACC, 30% Adenocarcinoma(AC) and 20% MEC. Clinical profile of these patients is shown in Table 1. No palpable cervical lymphadenopathy was noted in any cases. Most common symptom was nasal obstruction, secondary to nasal mass seen in anterior rhinoscopy obstructing nasal cavity (Fig. 1).

Out of five cases with ACC, 40% patients reported a history of nasal allergies and chronic cigarette smoking. In CT scan, most commonly involved sinus was maxillary sinus while frontal sinus was least commonly affected. Lamina papyracea was found eroded in patient presenting with excessive lacrimation and proptosis though orbital periosteum was intact (Fig. 2).

Out of 3 patients with AC, none had history of past nasal surgery, smoking, tobacco use, alcohol or wood dust exposure. Septum was found eroded in one case along with lamina papyracea (Fig. 3). Maxillary sinus was most commonly involved and sphenoid sinus was least commonly affected. On immunohistochemistry, CK20, CDX-2, and Ki-67 stains were found positive which made a diagnosis of Intestinal type adenocarcinoma (ITAC) colonic subtype in one case, while other two were non-intestinal type.

In the two patients diagnosed with MEC, histopathology showed low grade disease of the nasal cavity showing cystic areas with mucus, intermediate and squamous cells. Well differentiated mucinous cells predominated and mitoses were sparse.

Surgery: All cases underwent medial maxillectomy. Under general anaesthesia, modified Weber-Ferguson incision with lateral extension was given. After raising skin flaps, maxillary antrum entered using high speed drill and its medial wall removed. Tumour was excised and removed in toto. Surgical specimen was sent for histopathology. All patients are under regular follow-up (Fig. 4 & 5 showing preoperative and 3-week postoperative photograph of two patients). No recurrence on diagnostic nasal endoscopy or mortality noted till present date.

IV. Discussion

The sinonasal tract is affected by a diverse range of neoplasms, which may be epithelial, mesenchymal, or neuroectodermal in origin. Sinonasal adenomatous tumours may arise from submucosal muco-serous glands or surface mucosa.[9,10] Prevalence of each salivary gland carcinoma is diverse, as majority studies support most common to ACC [6,10–12], whereas others advocate ACC same as AC.[13] Common age group for occurrence of sinonasal minor salivary gland malignancies is 49-62 years (similar to our study); with similar prevalence in male and female, which is discordant with our study.[10,12] We found male preponderance and minimum age was 38 years though maximum patients were in age group 50-60 years.

Multiple site involvement is seen in ACC, however nasal cavity is commonly involved (29%), similar to our study where 80% ACC were arising from nasal cavity while one from maxillary sinus.[10] A malignant salivary gland neoplasm that does not fit any of the classic tumors (MEC, ACC, acinic cell or mixed) is usually classified as AC.[12] ITAC is composed of growth patterns that resemble carcinomas or adenomas of intestinal origin, or may mimic normal histology of the intestinal mucosa.[14] Most common site for AC is nasal cavity whereas in our study it was lateral wall of nose.[12] Most common site for ITAC is ethmoid sinus (40%) followed by nasal cavity (25%) and maxillary antrum (23%), whereas in our study only one case of ITAC was found which arose from maxillary sinus.[7] It is a locally aggressive malignancy with frequent spread to the orbit, the skull base, and the intracranial space.[15,16] Recurrences are frequent, however lymph node involvement and distant metastases are rare.[17] The most common site of MEC is maxillary antrum followed by nasal cavity, whereas in our study nasal cavity was most common site.[18]

Trauma, chronic irritation, vitamin A deficiency, occupational factors, radiation exposure and smoking are the risk factors for MEC and sinonasal carcinoma in general. In patients with MEC, no significant exposure or family history was noted in our study, hence genetic factors may play role in its origin. At present, the most common chromosomal translocation described for sporadic MEC is t(11;19)(q21;p13).[19] The occurrence of ITACs is found to be strongly related to occupational exposure to hard wood dust and leather dust. About 20% of ITACs are reported to be idiopathic, without evidence of exposure to industrial dust, similar to our case.[17] Furthermore, ITACs associated with dust exposure were diagnosed mostly in men (85-95%) and predominantly in the ethmoid sinus.[15] This contrasts with sporadic ITACs that are more frequent in women and often arise in the maxillary sinus, unlike our case which originated in nasal cavity of a male patient.[15]

Patients with sinonasal carcinoma present late with nasal obstruction, epistaxis, diplopia and/or facial swelling, similar to our cases.[10] Lymph node enlargement is more common in MEC and AC compared to ACC.[6] Though none of our cases had cervical lymphadenopathy.

Imaging studies, including CT, should be part of the early clinical management to determine if a neoplasm is present. When a mass is identified, MRI is essential for assessing possible intracranial and

intraorbital extension. Perineural invasion is characteristic of ACC seen in MRI, as was seen in our one case of ACC.[10] Hence, all patients with ACC underwent MRI in our series.

Classic histologic patterns of ACC of salivary glands (cribriform, tubular and solid) are also seen in nasal cavity.[10] ACC does not require immunohistochemistry for accurate diagnosis, though it may show positivity for keratin, CK5/6,s100,p53 and CD 117 but non-specific.[10]

Adenocarcinomas are divided into salivary-type and non-salivary-type, the latter being further subdivided into intestinal and non-intestinal-type adenocarcinoma.[10,16] Based on histological parameters, colonic subtype is the most frequent ITAC (40%).[20] Immunophenotype of ITAC includes staining for CK20, CDX2, villin, and MUC2, and variable positivity for CK7 which was also seen in our patient.[21] The histopathologic picture of MEC includes varying proportions of cystic and solid epithelial elements with different proportions of mucous, epidermoid, intermediate, and clear cells, similar to our cases. Intermediate cells usually predominate. Tumour grading of nasal MEC is based on morphological and cytological features.

Primary treatment consists of complete surgical resection if resectable. Surgical approach may differ, but may require a combined maxillectomy with neck dissection. Frozen sections may be helpful in AC and MEC, but not in ACC due to skip metastases.[10] Overall getting a clear surgical margin in sinonasal malignancies is difficult due to complex anatomy, which emphasize on postoperative radiation, as was followed in all our cases due to non-availability of frozen section.[6] If required, radiation or combined therapy is done, but treatment failure with advanced disease is fairly common. Extent of surgery did not affect prognosis in advanced-stage.[6] Radiation is more of palliative or used as adjuvant and helps in local control of disease.[11] Clinical nodal disease should be treated with appropriate neck dissection and postoperative radiotherapy. Inability to achieve wide margins should not be a contraindication for surgery in minor salivary gland tumors of the sinonasal tract.[6] The most frequently employed chemotherapeutic agents for all sinonasal malignancies are cisplatin, 5-fluorouracil and taxanes.[16]

Poor prognostic factors include high tumour grade, positive tumour margins, skull base invasion, age beyond 60 years, pain, intracranial extension, lymphovascular invasion and positive cervical nodes.[6,10,22] It was also found on a metanalysis that perineural invasion had no significant impact on survival in ACC [22], whereas significant in others.[10] Franchi et al. found that 46% of patients with ITAC had local recurrence after initial treatment and 56% ultimately died of the disease.[23] For MEC, ten year survival rate varies with low grade being 90% and 42% for high grade [24] or as low as 20% five year survival in high grade.[25] Recurrence is common in ACC to near 60%-75%.[10,11]

However, till date, there is no standard treatment for irresectable local recurrences or metastasis in sinonasal salivary gland malignancies. Further study needs to be done to devise a standard protocol for such rare and aggressive diseases.

V. Conclusion

Minor salivary gland malignancies of nose and PNS is an uncommon group. An elderly or middle aged male with unilateral obstruction should be seen with suspicion of malignancy and should be worked up to rule it out. These are aggressive and have poor prognosis except for low grade mucoepidermoid carcinoma. Diagnosis is mainly based on histopathology supported with immunohistochemistry. Aggressive surgery remains the primary treatment of choice followed by radiation. Recurrence is quite common with these except for low grade mucoepidermoid.

Clinical Significance

In this case series, we studied ten cases with nasal mass and who were diagnosed with sinonasal minor salivary gland carcinomas. We studied clinical profile, imaging, and surgical mangement for disease and followed up patients at regular interval. In a setup with limited facilities, we didn't found any mortality till date. We also studied our data in relation to prior studies done for sinonasal malignancies.

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