Case Report

Rare Sinonasal Non-intestinal Type Adenocarcinoma of Right Nasal Septum

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Abstract

Sinonasal tract is a complex anatomic structure with many possible diagnoses. Common diagnoses that usually encountered from the biopsied samples are inflammatory polyps and papillomas as well as squamous cell carcinoma. Another entity which most poorly understood is low grade glandular or tubular proliferations which includes reactive lesions, respiratory epithelial adenomatoid hamartoma, seromucinous hamartoma, and low grade sinonasal adenocarcinoma. We describe a case of a young male with right nasal septum mass which found incidentally upon flexible nasopharyngolaryngoscopic examination, who was referred earlier for possible obstructive sleep apnea. After excision of the tumour was done, it was confirmed to be low grade sinonasal adenocarcinoma non-intestinal type. This case highlights the rarity of the entity of low grade sinonasal adenocarcinoma non-intestinal type involving the right nasal septum; and how its subtle finding can be mistaken to be a clinically benign condition.

Keywords: Sinonasal tract; Low grade sinonasal adenocarcinoma non-intestinal type; Nasal septum

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Introduction

Lesions in the sinonasal tract can be reactive, benign or malignant neoplasms. Most of them consist of inflammatory polyps, inverted papilloma and squamous cell carcinoma¹. Other less common diagnoses include salivary type tumour, olfactory neuroblastomas, respiratory epithelial adenomatoid hamartoma, glandular hamartoma and low grade sinonasal adenocarcinoma². Generally, tumours of the nasal and sinonasal regions accounts for 0.4% of all human neoplasm. Amongst these, 13% of them are adenocarcinoma type³. Adenocarcinomas of the sinonasal tract are a group of neoplasms with differing microscopic appearance and different clinical behaviors. Some authors suggested its origin from surface

epithelium while others from seromucous gland⁴.

Case Report

A 17 year old Malay gentleman referred to rule out obstructive sleep apnea during his admission to medical ward for acute bronchospasm secondary to pneumonia. He complaint of having snoring for the past 3 years prior to presentation, associated with daytime somnolence and apneic spell. Apart from that, he also had rhinorrhoea, associated with early morning sneezing and nasal itchiness which occurred for at least 4 days per week. He denies of post nasal drip, facial pain, epistaxis, hyposmia or cacosmia, ear fullness or reduces hearing. On examination, he was alert and conscious. There was presence of allergic salute line. His body mass index was 44.2 kgm⁻², neck circumference

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of 44 cm, bilateral tonsillar enlargement grade 2, Friedman tongue position grade 3 and bilateral inferior turbinate hypertrophy upon anterior rhinoscopy. Flexible nasopharyngolaryngoscopy revealed a benign looking mass arising from right nasal septum at the level of right middle turbinate (Figure 1). It was painful upon probing, however there was no contact bleeding. The adenoid was moderately enlarged and upon Muller's manoeuvre there was presence of significant collapse at retropalatal and retrolingual region of about 50%. He scored 7 out of 24 points on Epworth Sleepiness Scale and 5 out of 8 points on STOPBANG scoring. He was diagnosed as moderate persistent allergic rhinitis with possible obstructive sleep apnea and right nasal septum papilloma.

He was started on tablet loratadine 10 mg once daily, fluticasone furoate nasal spray 2 puffs daily and subjected for endoscopic excision of the right nasal septum papilloma. Subsequently he underwent endoscopic excision of the nasal mass under local anaesthesia and the wound healing well post operatively. The histopathological examination (HPE) showed multiple fragments of polypoidal tissue covered with benign respiratory epithelium and focally by squamous metaplasia. The tissue composed of malignant cell infiltration, predominantly in crowded back to back glands with little intervening stroma. Some of them have tubular features with complex growth including some with papillary architecture (Figure 2). Some glands are cystically dilated and containing abscess. The glands were lined by single layer of uniform mucinous cuboidal to columnar epithelium. The cells have basally located nuclei with eosinophilic cytoplasm. No mitosis was seen. The surrounding stroma shows mild lymphoplasmacytic cells infiltration (Figure 3). The tumour cells were positive for intraluminal mucin and showed diastase resistant positive reaction to PAS and PAS-D, as well as CK7. However the tumour was negative for CK20 immunostaining. These findings are consistent with low-grade sinonasal non-intestinal type adenocarcinoma. A polysomnography done showed apnea hypopnea index of 11.9 with lowest oxygen desaturation of 84%; which categorised as mild obstructive sleep apnea. A computed tomography (CT) of vertex till pelvis done showed no evidence of distant metastasis with clear paranasal sinuses, with only edematous nasal cavity and bilateral inferior turbinate hypertrophy.

Figure 1: Right nasal septum mass (arrow).

Figure 2: The tissue is lined by benign respiratory epithelium. There is malignant cells infiltration in crowded back to back glands with little intervening stroma seen within the subepithelium (blue arrow). Some are arranged in papillary architecture (yellow arrow). (H&E x100)

Figure 3: The glands are lined by single layer of uniform mucinous cuboidal to columnar epithelium having basally located nuclei with eosinophilic cytoplasm. No mitosis seen. The surrounding stroma shows mild lymphocytic cells infiltration. (H&E 400x)

Upon subsequent follow-up, there was no obvious mass or lesion seen at previous operated site. He was offered for adenotonsillectomy however he refused for the operation. He was given follow-up for the next 6 months.

Discussion

Adenocarcinomas involving the sinonasal tract are rare. In 2005, World Health Organization classified them as salivary and non-salivary type. Salivary type consist of specific type of carcinoma such as adenoid cystic carcinoma and mucoepidermoid carcinoma while the non-salivary type is divided into intestinal type adenocarcinoma (ITAC) and non-intestinal type adenocarcinoma (NITAC). The NITAC can be diagnosed as low-grade or high-grade⁵. ITAC of the sinonasal tract are composed of cells mimicking normal, adenomatous or carcinomatous intestinal mucosa². This case highlighted the low-grade sinonasal adenocarcinoma (LGSNA), non-intestinal type as diagnosed in our patient.

Many terms have been used to describe LGSNAs; which includes low-grade adenocarcinoma of sinonasal tract, seromucous adenocarcinoma, terminal tubulus adenocarcinoma and sinonasal tubule-papillary adenocarcinoma⁶. It is a rare neoplasm with no known association with carcinogen. This is true in our case, where he had no risk factors such as exposure to environmental carcinogens. In fact, the tumour was found as an incidental finding during nasoendoscopic examination. It has slight male preponderance and in most cases, it affect the adults with a mean age of 58 years and ranging from 9-89 years of age^{7,8} as in our case. Frequently involved site is the nasal cavity, followed with ethmoid and maxillary sinuses¹.

Usually in this type of case, patients mostly present with unilateral nasal obstruction and epistaxis. However in our patient, he denied of neither nasal

obstruction nor epistaxis. Pathological features of LGSNA includes predominantly tubule-cystic architecture which comprised of small round tubules lined by a single row of cuboidal to columnar cells and the tumour cells exhibit uniform cytology characterised by pale eosinophilic to clear neoplasm, mild to moderate nuclear atypia without prominent nucleoli as well as rare mitotic figures¹. In our case, there was no mitosis seen on HPE. On immunohistochemical studies, LGSNA express CAM 5.2, AE1, CK7, CK19 and S100 and negative on CK20 and MUC2. In our patient, the immunohistochemical staining is positive on CK7 and negative for CK20.

LSGNA are relatively indolent tumour and most of the patients have an excellent prognosis⁸. In our case, after few months follow-up post operatively, there was no tumour recurrence noted and there was no distant metastasis seen.

Conclusion:

Primary adenocarcinomas of the sinonasal tract are a diverse group of malignancies that can be classified as salivary and non-salivary types. LGSNA non-intestinal is a rare non-salivary type sinonasal tumour with good prognosis. Any mass in nasal cavity should be sent for histopathological examination, though clinically it appeared as benign condition.

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