



Case Report

Human papillomavirus-related multiphenotypic sinonasal carcinoma: First case report associated with an intermediate-risk HPV type and literatures review



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ABSTRACT

HPV-related Multiphenotypic sinonasal carcinoma (previously known as HPV-related carcinoma with adenoid cystic carcinoma-like features) is very rare tumor of which only 55 cases are reported. All cases to date are related to high-risk types of HPV including 16, 31, 33, 35 and 56. We report of a case of a nasal mass in a 50-year-old female. Biopsy demonstrated typical features of an HPV-related multiphenotypic sinonasal carcinoma, with both basaloid and ductal type cells. Tumor cells were arranged in lobulated sheets, with focal cribriform and microcystic growth patterns. The majority of tumor cells expressed myoepithelial markers (S-100 protein, smooth muscle actin and p63) and the ductal cells were immunopositive for CD117. Polymerase chain reaction and sequencing identified HPV type 26. HPV 26 becomes the first intermediate risk type that may be associated with HPV-related multiphenotypic sinonasal carcinoma.

1. Introduction

Malignant tumors of the sinonasal tract are rare neoplasms with about 6 cases per million per year [1]. Almost 80% of tumors occurring in this area are epithelial neoplasms, including squamous cell carcinoma, adenocarcinoma and sinonasal undifferentiated carcinoma. Less common are olfactory neuroblastoma, melanoma, sarcoma, and lymphoma [1, 2]. Recently, other tumors originating in the sinonasal tract have been recognized, including: NUT midline carcinoma, *SMARCB1* (INI-1) deficient sinonasal carcinoma, biphenotypic sinonasal sarcoma, renal cell-like adenocarcinoma, and human papillomavirus (HPV)-related multiphenotypic sinonasal carcinoma [3], formerly known as HPV-related carcinoma with adenoid cystic carcinoma-like features [4]. The last tumor is related to infections by specific high-risk HPV types, namely 16, 33, 35 and 56 [3–7]. We report a case of HPV-related multiphenotypic sinonasal carcinoma that was positive for HPV type 26, which is the first intermediate-risk type to be associated with this type of cancer.

2. Materials and methods

2.1. Case report

A 50-year-old female patient presented with recurrent epistaxis and right nasal congestion for six months. Nasal endoscopy revealed a smooth mass in right nasal cavity, originating from posterior floor, which bled easily on contact. There was also a left deviation of the nasal septum and hypertrophy of both inferior turbinates. No lymphadenopathy was noted. Immune status was not documented. A biopsy showed an infiltrating tumor forming nests and sheets of malignant cells with focal necrosis. The initial diagnosis was a malignant round cell tumor with epithelioid and neuroendocrine features. Endoscopic sinus surgery was performed, revealing a pedunculated mass originating from the right pterygoid plate, posterior to the sphenopalatine foramen. The mass was totally removed. The patient received postoperative radiation therapy for 6 cycles over one month and computed tomography at that time showed no residual tumor. The patient is now 85 months post-surgery, and regular nasal endoscopy and computed tomography have continued to show no evidence of recurrent tumor.

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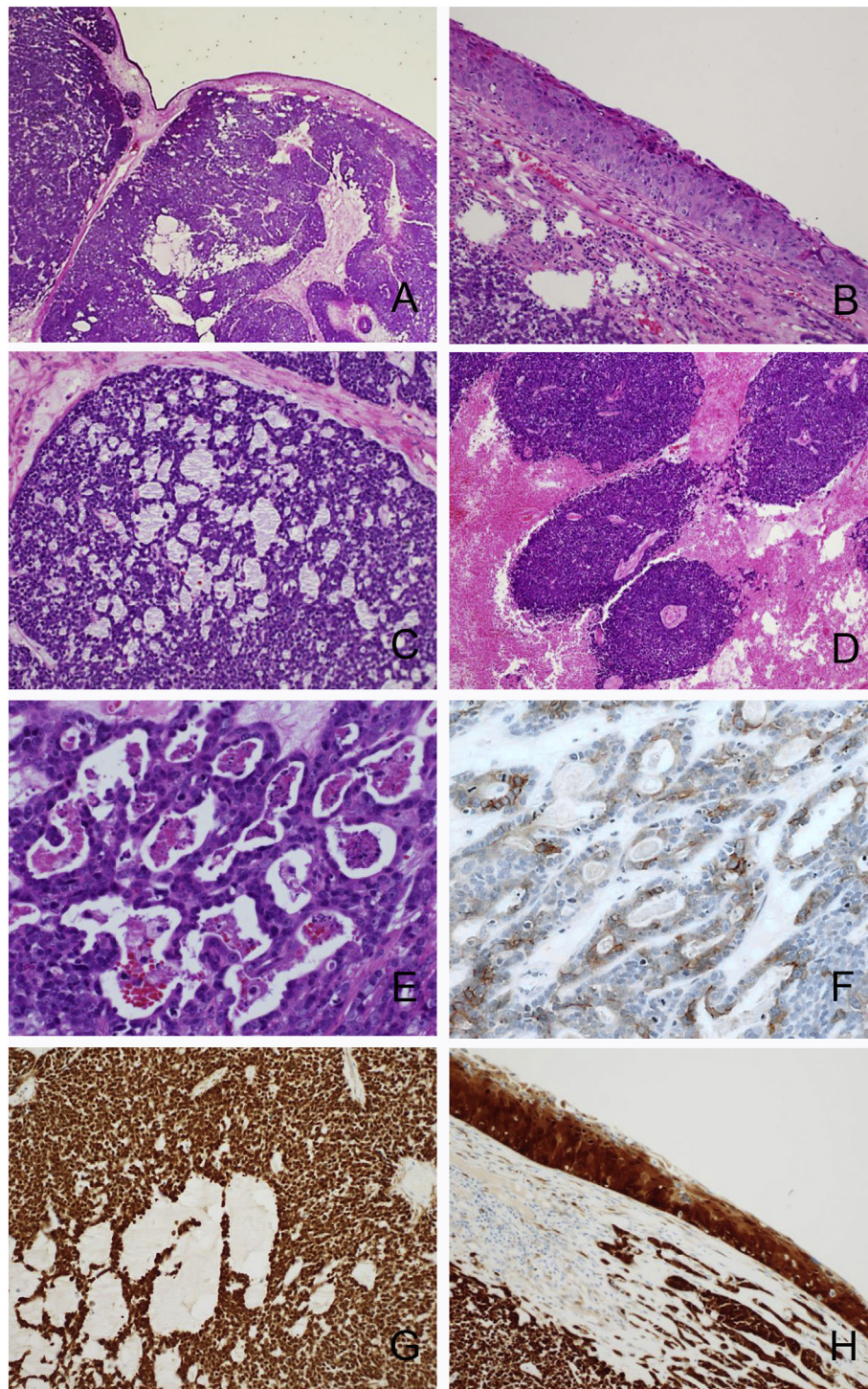


Fig. 1. Microscopic appearance of the tumor. (A) Lobulated tumor mass composed of basaloid cells; (B) the surface epithelium exhibit carcinoma-in-situ; (C) tumor cells arranged in a microcystic pattern with lumina filled with pale blue material resembling mucopolysaccharide; (D) focal tumor necrosis; (E) true ducts lined by cuboidal tumor cells; (F) ductal cells expressing CD117; (G) basaloid tumor cells and (H) surface epithelium showing diffuse, strong expression of p16. (A-E: hematoxylin and eosin, original magnifications A x40, Bx100, C x100, D x100, E x200; F-H: immunoperoxidase, original magnifications F x200, G x100, H x100).

2.2. Pathology findings

The resected tumor was a broad-based polypoid lesion, covered by squamous epithelium with dysplastic change involving the whole thickness of the epithelium (i.e., squamous carcinoma-in-situ). The underlying stroma was infiltrated by lobulated sheets of malignant basaloid cells with focal necrosis. The tumor cells possessed mildly pleomorphic, round to oval, hyperchromatic nuclei with indistinct

nucleoli and scant eosinophilic cytoplasm. There was focal cribriform and microcystic arrangement, with the lumina filled by pale eosinophilic to pale blue material resembling the mucopolysaccharide material seen in adenoid cystic carcinoma. This material stained positively with PAS-diastase and mucicarmine. True duct formation was present with a lining of cuboidal neoplastic cells possessing oval, vesicular nuclei with prominent nucleoli and a moderate amount of eosinophilic cytoplasm. The mitotic count was 15/10 high power fields. No evidence

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