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Sinonasal Non-Intestinal-Type Adenocarcinoma: A Case Report

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Abstract

Non-intestinal-type adenocarcinoma (non-ITAC) is defined as an adenocarcinoma that can arise anywhere in the sinonasal tract does not show features of salivary gland neoplasia, and does not have an intestinal phenotype. We report a case of a 79-year-old patient treated for colorectal cancer with lung metastasis by surgical excision and chemotherapy, who presented recurrent episodes of epistaxis evolving over more than 4 years. Her clinical examination showed a mass on the left nasal cavity. The computed tomography (CT) scan showed a locally aggressive left ethmoido-nasal mass. Magnetic resonance imaging (MRI) showed a process of the nasal fossae and the left posterior ethmoidal cells. The patient was treated surgically with a middle meatotomy with anterior ethmoidectomy of the left nasal cavity, the surgical specimen was sent for a histological examination which confirmed the diagnosis of sinonasal non-ITAC. In Conclusion, non-ITACs are a rare type of tumor and are treated by complete surgical excision. Extensive disease or higher histologic grade may require adjunctive radiotherapy. The prognosis is determined by histologic grading: low-grade tumors are relatively indolent, whereas high-grade tumors confer a greater risk of recurrence and death.

Keywords

Endoscopic Surgery, High Grade Adenocarcinoma, Immunohitochemistry, Low-Grade Adenocarcinoma, Radiotherapy, Sinonasal Malignancies, Sinonasal Non-Intestinal-Type Adenocarcinoma

1. Introduction

Primary adenocarcinomas of the sinonasal tract (SNACs) are a heterogeneous group of malignancies representing 10% of all primary malignant neoplasms of the nasal cavity and paranasal sinuses (Leivo et al., 2016) and are classified by

The World Health Organization as salivary and nonsalivary types (Barnes et al., 2005; Franchi et al., 2005). Nonsalivary types are separated into intestinal-type SNAC (ITAC) and non-ITAC, and both have low- and high-grade categories (Bhaijee et al., 2011). Non-ITAC is defined as an adenocarcinoma that can arise anywhere in the sinonasal tract, and does not show features of salivary gland neoplasia, and does not have an intestinal phenotype (Jain et al., 2017). Clinical presentation of this tumor primarily involves nasal obstruction and epistaxis, as well as infrequent pain. immunohistochemical studies showing the respiratory-type immunostaining profile ($CK20^{-}$, $CK7^{+}$, $CDX2^{-}$, and $villin^{-}$) (Bignami et al., 2018).

The literature focusing on n-ITAC is inadequate, as most studies include all the subtypes of adenocarcinoma (Chen et al., 2015) or focus on low-grade tumors (Stelow et al., 2011) or on the histologic profile (Choi et al., 2003).

Here, we present a case of non intestinal sinonasal adenocarcinoma to describe and discuss the clinicopathological and immunohitochemical features of this rare entity.

2. Case Report

2.1. Patient Information

We report the case of a 79-year-old man patient treated in March 2018 for colorectal cancer with lung metastasis by surgical excision and chemotherapy, who presented since 2018 recurrent episodes of epistaxis and which was never explored until March 2022.

2.2. Clinical Findings

The clinical examination revealed an eupneic patient and the rhinological examination revealed a nasal flow absent on the left and preserved on the right, with no nasal pyramid deformity.

Nasoendoscopy showed on the right side an inflamed nasal mucosa and free cavum and middle meat, and on the left side a mass originating from the middle meat, with the presence of a reddish tissular process underneath, reaching down to the lower turbinate and back towards the choana.

Oropharngic and otalgic examinations were normal with absence of palpable adenopathies.

2.3. Diagnostic Assessment

A computed tomography CT scan of nasosinus, thorax, abdomen, and pelvis was performed, showing a locally aggressive left ethmoido-nasal tissue process responsible for lysis of the anterior skull base without intracranial extension, with pulmonary nodules and left adrenal nodule (**Figure 1**).

A magnetic resonance imaging (MRI) of the facial mass showed a process of the nasal fossae and the left posterior ethmoidal cells of benign morphology, making evoke a reversed papilloma (**Figure 2**).

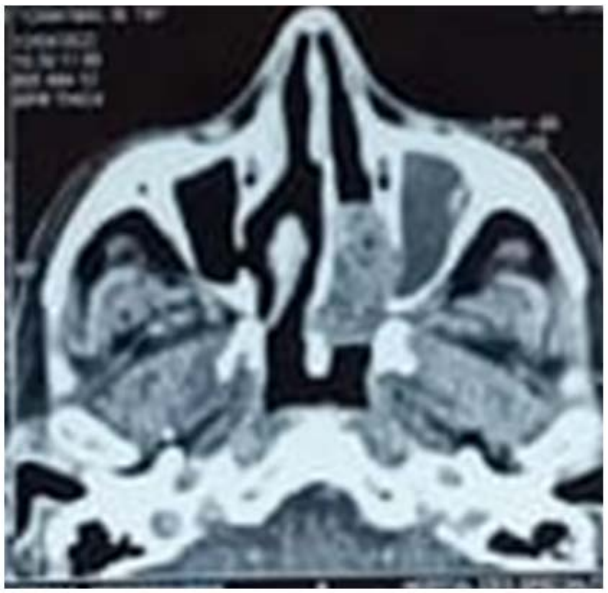

Figure 1. Computed tomographic scan, showing a mass in the left nasal cavity.

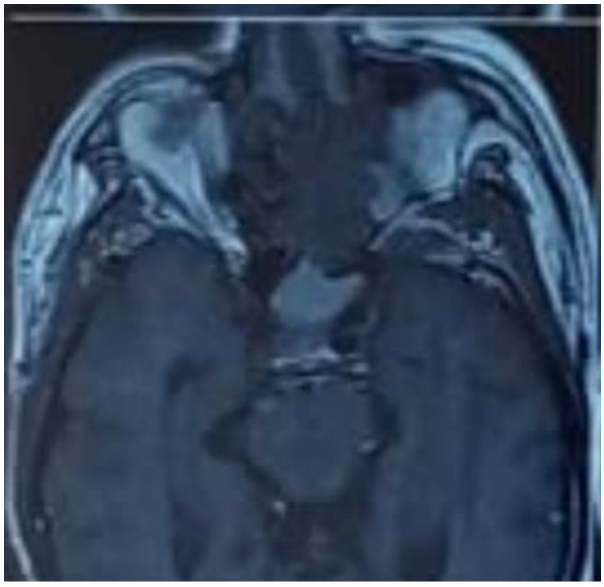

Figure 2. MR images of a tumour involving the right nasal cavity the left nasal cavity.

2.4. Therapeuptic Intervention and Follow-Up

The patient was treated surgically with a middle meatotomy with anterior ethmoidectomy of the left nasal cavity with histopathology reported it as sinonasal non-intestinal-type adenocarcinoma (Figure 3) ($S100^+$, $CK7^+$, $CK20^-$, $CDX2^-$). Then the patient was addressed to the oncology unit, and was treated with adjuvant radiotherapy and chimiotherapy and he died of shock septic after nine months.

3. Discussion

Non-ITACs, of presumed seromucous gland origin, are nonsalivary-type SNACs

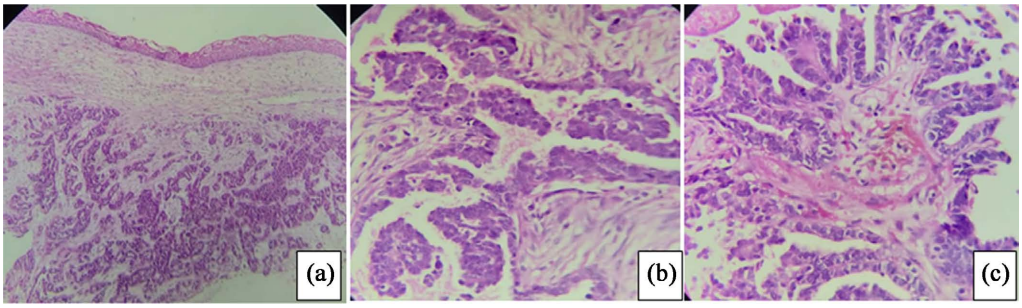

Figure 3. (a) Histology section of tumor cells separated by fibrous septae admixed with large areas of necrosis within the tumor; (b) section of tumor cells arranged in back to back arranged glandular, papillary, cord-like and solid pattern; (c) section of moderately pleomorphic tumor cells, scanty cytoplasm, coarse clumped chromatin, occasional prominent nucleoli and good number of mitotic (Hematoxylin and Eosin).

that lack intestinal features (Poizat et al., 2009; Weinreb, 2010). Marked morphologic heterogeneity precludes precise definition of non-ITACs, often resulting in diagnostic uncertainty and rendering non-ITAC a diagnostic category of exclusion (Weinreb, 2010). Low- and high-grade non-ITACs are distinguished by the presence or absence of necrosis, mitotic activity, and cytologic atypia (Franchi et al., 2005).

Non-ITACs do not show sex or racial predilection and are more common after 50 years of age (Stelow et al., 2010). They can arise anywhere in the sinonasal tract; the nasal cavity is most commonly affected, followed by the ethmoid and maxillary sinuses (Perez-Ordonez et al., 2009). No risk factors, predisposing conditions, or environmental exposures have been described for non-ITACs (Stelow et al., 2011, 2010; Jo et al., 2009). As in our case, recurrent epistaxis or nasal obstruction may be the presenting symptoms. Non-ITACs are radiographically evaluated by computed tomography or magnetic resonance imaging.

Low-grade non-ITACs vary in both consistency and morphology. Usually, these tumors demonstrate well differentiated glandular patterns, which may be exophytic and papillary or infiltrative and tubulocystic (Yue et al., 2021). Compact acini, back-to-back glands, cystic dilatation, and papillae formation are commonly seen. The tumor cells exhibit uniform morphology, with abundant cytoplasm, mild-to-moderate nuclear atypia, inconspicuous nucleoli, and few mitoses (Perez-Ordonez et al., 2009; Skalova et al., 2003). Despite the apparent bland morphology of low-grade non-ITACs, their complex pattern and locally invasive growth indicate malignancy.

The high-grade histologic features in our case prompted the diagnosis of high-grade adenocarcinoma. By WHO definition, high-grade non-ITACs (Stelow et al., 2011) cannot be better diagnosed as a specific form of differentiated salivary gland neoplasia (Cardesa et al., 2006) do not have an intestinal phenotype, and are high grade with moderate-to-marked cytologic pleomorphism, high mitotic activity, and/or necrosis (Poizat, et al., 2009; Stelow et al., 2011). Other features of high-grade non-ITACs include solid growth patterns with sheets of cells, irregular glandular patterns, and hyperchromatism (Heffner et al., 1982). Unlike

the predominantly papillary or tubular patterns of low-grade lesions, high-grade lesions are often solid or trabecular (Stelow et al., 2010). Non-ITACs are positive for CK7 and S-100 and negative for CK20, MUC2 (Mucin 2), and CDX2 (Jo et al., 2009; Skalova et al., 2003). This immunostaining profile is helpful in distinguishing non-ITAC from ITAC but does not distinguish non-ITACs from hamartomas (Jo et al., 2009). Other negative immunostains include calponin, CD10, p63 (except in areas of squamous metaplasia), chromogranin, and CD57 (Skalova et al., 2003).

Non-ITACs are treated by complete surgical excision (Heffner et al., 1982). Extensive disease or higher histologic grade may require adjunctive radiotherapy. The prognosis of patients with non-ITACs is determined by histologic grading: low grade tumors are relatively indolent, with low risk of local invasion or metastasis, whereas high-grade tumors confer a greater risk of recurrence and death (Stelow, 2010).

Authors' Contribution

Diagnostic and therapeutic management, surgical intervention and writing the article were done by Mohamad Ezadine, Moad El Mekkaoui. Interpreting data, guiding management, reviewing the article by Leila Essakalli, Abdelilah Oujilal, Anas Benbouzid and Razika Bencheikh. All authors read and approved the final version of the manuscript.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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