CASE REPORT
DOI: 10.17944/interdiscip.1352024
Interdisciplinary Medical Journal 2023;14(49):145-147.

Intestinal-type adenocarcinoma of the sinonasal mucosa: a rare case report

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Abstract

Intestinal-type adenocarcinoma is the most commonly seen adenocarcinoma of the sinonasal region following adenoid cystic carcinoma. They are aggressive tumors that are generally diagnosed in the advanced stage. Histopathological subtype is an important prognostic indicator. In this report, we have presented a case we diagnosed as intestinal-type adenocarcinoma with right nasal cavity localization.

Keywords: Sinonasal, Adenocarcinoma, Intestinal-type

INTRODUCTION

Intestinal type adenocarcinoma (ITAC) is the most commonly seen adenocarcinoma of the sinonasal region following adenoid cystic carcinoma (1). They are classified as papillary, colonic, solid, mucinous and mixed type according to histological features (1, 2). It is more frequently found in males due to occupational exposure and between ages 50-64 years (1, 3). They are most commonly detected in the ethmoid sinuses followed by nasal cavity and maxillary antrum (1). They are generally diagnosed in the advanced stage, they are clinically aggressive tumors and its histopathological subtype is an important prognostic indicator (1, 4, 5). They may have a morphological appearance of normal intestinal mucosa, villous adenoma, colorectal adenocarcinoma, mucinous adenocarcinoma and signet-ring cell carcinoma (4, 6).

In this report, we have presented a case we diagnosed as intestinal-type sadenocarcinoma with right nasal cavity localization.

CASE

The 74-year-old male patient admitted to the policlinic of Otolaryngology Department due to the complaint of difficulty in breathing. Rhinoscopy revealed polypoid tissue that fills the right nasal cavity in the septal midline. No pathological finding was detected in the examination of other systems. The computed tomography encountered a soft tissue density lesion that extended from right maxillary sinus to right nasal passage through secondary ostium and to nasal pharyngeal region. The patient was operated with prediagnosis of antrochoanal polyp? inflammatory polyp? and inverted papilloma? The pathology material consisted of gray-white colored tissue fragments with a volume of 9cc. Microscopic examination showed a tumor with sparse papillary and remarkable tubuloglandular architecture (Figure 1). Tumor cells included hyperchromatic nuclei and sparse goblet cells (Figure 2). No lymphovascular and perineural invasion was present. Tumor cells demonstrated CDX2-positive and CK20-positive immunoreactivity (Figure 3). No staining was observed for CEA and CK7. No lesion detected in the colon by colonoscopy. The patient was diagnosed with intestinal-type adenocarcinoma according to these findings.

Cite this article: Kuzuluk DG, Uysal S, Özler GS.Intestinal-type adenocarcinoma of the sinonasal mucosa: a rare case report. Interdiscip Med J. 2023;14(49):145-147. https://doi.org/10.17944/interdiscip.1352024

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Received: November 24, 2022
Accepted: July 10, 20232

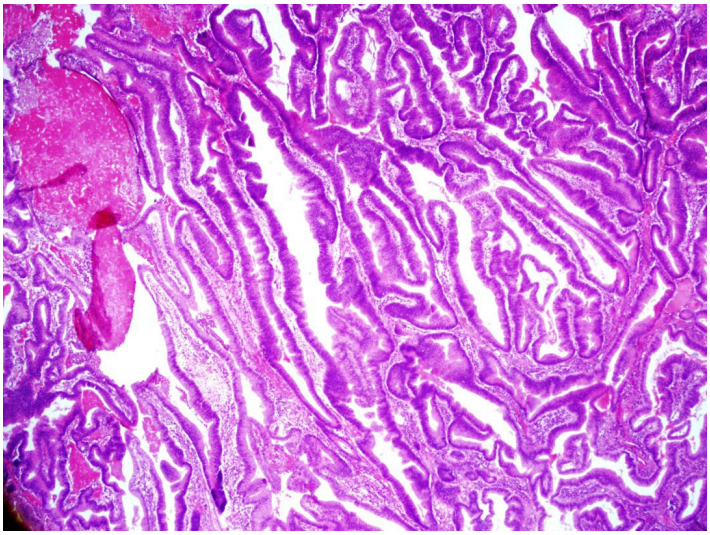

Figure 1. Tubuloglandular architecture (H+E, x100)

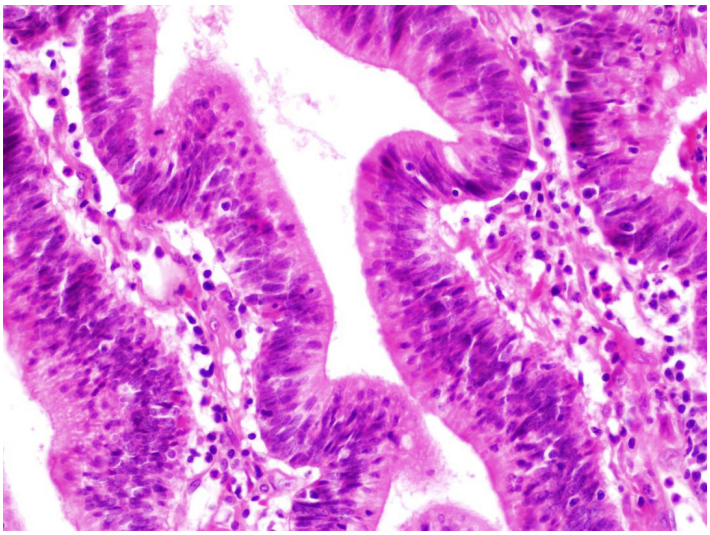

Figure 2. Tumor cells included hyperchromatic nuclei (H+E, x400)

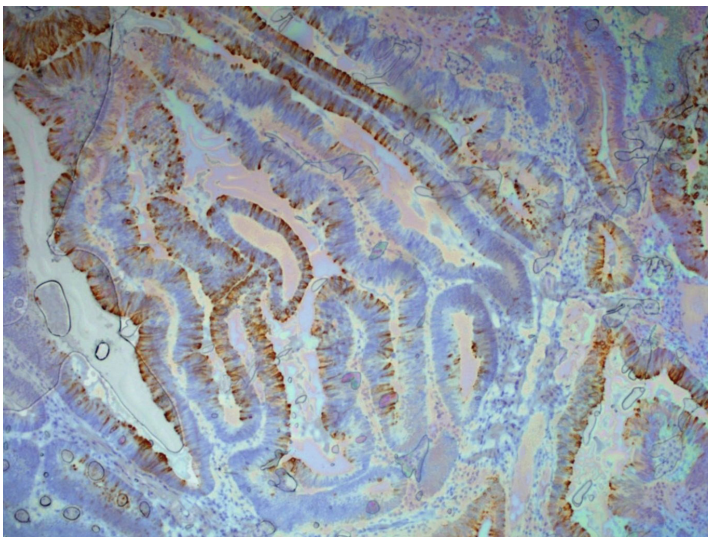

Figure 3. Cytoplasmic immunostaining with CK20 (CK20, x200)

DISCUSSION

The adenocarcinomas of the nasal cavity and paranasal sinuses (SNAC) are very rarely seen tumors and they constitute 10-20% of the primary sinonasal carcinomas and 3-5% of all head and neck malignancies (2). They are divided into two as salivary and non-salivary types based on the originating cell while non-salivary type is classified as intestinal type and non-intestinal type adenocarcinoma according to histopathological features (2, 6). ITAC is the second most common type of sinonasal adenocarcinoma after adenoid cystic carcinoma (1). Occupational exposure (leather dust, nicel, wood dust) is blamed etiologically, however, sporadic cases also have been reported (2, 4, 6). The time elapsed from exposure to tumor development is 18-50 years, however, there is no sufficient data on the relationship between inhaled dust amount and tumor development period. The other factors considered to be etiologically responsible are Epstein-Barr viral infection and cigarette smoking (7). The studies carried out in Europe have determined a strong relationship between occupational exposure and development of these tumors (2, 3, 8) whereas a weaker relationship has been detected in the USA-based studies (2, 9). Occupational exposure has been reported more frequently in males as an etiological factor in the Europe-based studies whereas it has been reported in 2014 Survival, Epidemiology and End Results (SEER) study that these tumors were found with frequency in both genders (2, 10). Our case was also a male patient with advanced age and no risk factor was present.

ITACs of the nasal cavity are the destructive lesions that generally emerge in the nasal midline. These tumors present better prognosis compared with squamous cell carcinomas (7). They constitute 8-25% of all malignant sinonasal tumors. It is supposed to develop from intestinal metaplasia of the ciliated respiratory epithelium and inferior and middle turbinates are the preferred sites in nasal cavities. Epistaxis, unilateral nasal obstruction and rhinorrhea are the most commonly seen complaints (11). Pain, neurological deficits, exophthalmos and visual impairments may be seen in the advanced stage tumors. They have grossly papillary or polypoid features and fungating appearance, and they may have gelatinous viscosity resembling mucocele (12). Our case was found to have a polypoid tissue that filled the nasal cavity.

There are two major classifications for ITACs as Barnes Classification and the Classification of Kleinasser and Schroeder. There are five subtypes as papillary, colonic, solid, mucinous and mixed ITACs according to Barnes Classification while Kleinasser and Schroeder have defined four subtypes as papillary tubular cylinder cell ITACs (grade 1, 2, 3), alveolar goblet type, signet-ring type and transitional type (6, 13). There are differences between histological classes and clinical behaviors (1, 5, 6, 13). The colonic ITAC is the

most frequently seen subtype (1). Goblet cell, paneth cell and argentaffin cell may be observed in all subtypes and well-differentiated ITAC may resemble normal intestinal mucosa. Immunohistochemical staining indicates that ITACs are positive for CK20, CDX-2, villin and MUC2, and variably positive for CK7 (1, 4, 5).

The differential diagnosis of ITAC involves colonic adenocarcinoma metastasis and sinonasal low-grade non-intestinal adenocarcinoma (1, 6). Morphological and immunohistochemical differentiation of primary ITAC from colonic adenocarcinoma metastasis is difficult. The clinical features and colonoscopic findings of the patient will be helpful in differential diagnosis. Thus, if an intestinal-type tumor has been detected in the sinonasal tract, colonoscopy or colorectal radiographic studies should be performed to rule out primary colorectal adenocarcinoma. Colonoscopy revealed no lesion in our case. The differential diagnosis of ITAC from sinonasal non intestinal adenocarcinomas is supported by immunohistochemistry for CK20, CDX-2, villin and SATB-2 that only stain ITACs.

The treatment for ITACs is surgical and type of surgery may vary between lateral rhinotomy, partial maxillectomy and total maxillectomy. ITAC is a high-grade malignancy. The rates of local recurrence, lymph node metastasis and distant metastasis were found 50%, 8% and 13% in a study carried on 213 patients with ITAC, respectively, and 60% patients became exitus due to this disease. The patients who developed this tumor due to exposure to wood dust were found to have a better prognosis compared with sporadic ITAC. The well-differentiated ITACs manifested a better prognosis than solid and mucinous subtypes (1, 6). Our case was colonic type ITAC and well-differentiated, and he has been under follow-up with remission for 16 months.

CONCLUSION

Intestinal-type adenocarcinomas are the rarely seen neoplasms of the sinonasal tract. They are aggressive tumors that are generally diagnosed in the advanced stage. Histopathological subtype is an important prognostic indicator. Colonic adenocarcinoma metastasis should be ruled out by clinical history and colonoscopic examination before diagnosis of primary ITAC.

ACKNOWLEDGEMENT

Conflict of Interest

The authors declare that they have no conflict of interests regarding content of this article..

Financial Support

The Authors report no financial support regarding content of this article.

Ethical Declaration

Informed consent was obtained from the participant and Helsinki Declaration rules were followed to conduct this study.

Authorship Contributions

Concept: DGK, SU, Design: DGK, SU, Supervising: DGK, GSO, Data collection and entry: SU, Analysis and interpretation: DGK, GSO, Literature search: DGK, SU, GSO, Writing: DGK, Critical review: DGK, GSO.

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