Sinonasal Intestinal-Type Adenocarcinoma
Involvement of the Paranasal Sinuses

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Sinonasal Intestinal-Type Adenocarcinoma
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Summary: We present a patient with a biopsy-proved sinonasal intestinal-type adenocarcinoma who presented with moderate confusion. He was found to have bifrontal hemorrrhages, which to our knowledge has not been previously described in the literature for this entity. Intestinal-type adenocarcinoma should be in the differential diagnosis of aggressive lesions in the base of the skull with intracranial spread from the paranasal sinuses.

Adenocarcinomas account for 10–20% of all primary malignant neoplasms of the nasal cavity and paranasal sinuses (1). Many of these are of salivary gland origin, but others are less familiar and have histologic patterns similar to those of adenocarcinoma of the colon. These latter ones have been named intestinal-type adenocarcinoma (ITAC) and are responsible for less than 4% of the total malignancies of this region (2). These tumors occur primarily in men aged 55–60 years. The tumors are common in workers in the hardwood and shoe industries. Exposure to wood dust increases the risk of adenocarcinoma by 900 times. Although these neoplasms result in almost 4% of the primary neoplasms of the sinonasal tract, little has been written about them in the radiology literature. We present a case of an ITAC in the sinonasal cavity with intracranial spread, which occurred with bifrontal hemorrhages. To our knowledge, this finding has not been previously described.

Case Report

A 68-year-old man presented to an outside hospital, where he was found to have moderate confusion and evidence of bifrontal hemorrrhages, as demonstrated on a CT scan. He was then transferred to our Veterans Affairs hospital. The patient’s relatives related that he was becoming increasingly confused, disheveled, and inattentive to his personal hygiene. In addition, the patient complained of a 11-kg unintentional weight loss over the last 3 months. He denied any history of trauma. At our institution, neurologic examination showed marked cognitive deficits, but his motor and sensory functions were intact. MR imaging was performed, and the images showed a bifrontal lesion, mostly of high signal intensity on T1-weighted images (Figs 1–3). The lesion extended from the ethmoid sinuses to the skull base and into the parenchyma of the frontal lobes. Erosion of the cribriform plate was highly suspected. The tumor had heterogeneous signal intensity on T2-weighted images, although areas of high signal intensity on T1-weighted images demonstrated corresponding low signal intensity on T2-weighted images. The paramidline portions of the tumor enhanced after the administration of contrast material. In addition, substantial edema surrounded the lesion. A preoperative CT scan (Fig 4) was obtained at our institution. This scan demonstrated a bifrontal lesion with attenuation consistent with that of hemorrhage. The ear nose and throat (ENT) team was consulted and performed a transnasal biopsy, which revealed ethmoidal adenocarcinoma. The patient then underwent bifrontal craniotomy with bilateral orbital osteotomy by means of a skull-base approach involving a neurosurgeon in collaboration with the ENT team. Tumor removal was achieved, and histopathologic analysis revealed a primary ITAC. The patient’s immediate postoperative course was uneventful, and plans were made for postoperative radiation treatment. Subsequently, the patient’s postoperative course was complicated with meningitis, and despite antibiotic therapy, the patient died 3 months after surgery.

Discussion

ITAC of the sinonasal tract may occur sporadically or as an occupational hazard. Exposure to softwood dusts in the logging and milling industries and leather dust in the shoemaking industry has been implicated as risk factors for the development of these neoplasms (3). About 20% of these tumors have historically arisen in individuals with exposure to industrial wood dust (4). The time between the first occupational exposure to wood dust and the development of adenocarcinoma of the sinonasal tract averages 40 years (5).

Findings from recent studies have suggested clinical differences between ITAC arising in individuals with occupational dust exposure and ITAC arising sporadically (3). Tumors related to occupational exposure affect men in 85–95% of cases, and the tumors show a strong tendency to arise in the ethmoid sinuses (6, 7). Sporadic tumors frequently arise in women and involve the maxillary antrum in 20–50% of cases (3). Patients with sporadic ITAC tend to have survival times shorter than those of patients with tumors related to occupational exposure. The reason for this difference is related to the initial stage of the tumor at the time of its discovery. Tumors arising in the maxillary sinus (typically sporadic cases) do not become symptomatic until they are at an advanced stage, unlike those of the nasal cavity and ethmoid tumors, which become symptomatic before they invade local structures (3, 8). Interestingly, our patient had no history of occupational exposure, and although the
lesion seemed to arise in the ethmoid sinus, his symp-
toms were not related to the sinuses, but rather, to the
intracranial spread. Therefore, the tumor was quite
advanced at presentation.

Reported sites for the origin of ITACs have been as
follows: ethmoid sinuses, 40%; nasal cavity, 28%;
maxillary antrum, 23%; and indeterminate, 9% (3).
Typical presenting symptoms include nasal obstruc-
tion, epistaxis, rhinorrhea, mass in the cheek, and
exophthalmos. Less common are symptoms related to
facial nerve involvement (3). Symptoms are usually
present for less than a year, but they may last as long
as 5 years.

The gross appearance of these tumors is similar to
that of colonic adenocarcinoma. The most common
variant of ITAC resembles typical gland-forming co-
lonic adenocarcinoma, and in many instances, the
biopsy specimens are completely indistinguishable
from those of a primary colonic neoplasm. Areas of
mucosal ulceration are typical and may have associ-
ated hemorrhage, as in our case. In poorly differen-
tiated tumors, gland formation is less obvious, and the
tumors acquire more-prominent solid components.

The grading of ITAC has been the subject of sev-
eral studies. Barnes (3) recognized five variants of
ITACs: papillary, colonic, solid, mucinous, and
mixed. Papillary and colonic tumors have an appear-
ance similar to that of colonic adenomas. They often
lack clear-cut features of malignancy, although some
of these could be clearly invasive. The literature sug-
ests that the papillary ITAC may have the best prog-
nosis, as it typically behaves as a smoldering, locally
destructive lesion with a limited tendency for regional
or distant metastases (7, 9).

Initial immunohistochemical results helped con-
firm the intestinal differentiation of these tumors
by documenting the presence of intestinal-type hor-
mones. Interestingly, although carcinoembryonic
antigen is strongly expressed in virtually all colonic
adenocarcinomas, in one study, only two of 12
ITACs showed strong staining for this antigen (10).
In further distinction from colonic neoplasms, nine
of 12 ITACs showed numerous chromogranin-
positive cells, whereas only three of 12 colonic
adenocarcinomas showed rare chromogranin pos-
itivity (10).

Pathologists unaccustomed to ITACs may suspec-
ta metastasis from the gastrointestinal tract or even the
breast. Metastatic tumors to the nasal cavity and
paranasal sinuses do exist and include tumors of the
following (in order of decreasing frequency): kidney,
lung, breast, testis, gastrointestinal tract, uterus, thy-
roid, adrenal glands, and pancreas. Also included in
this list are melanomas (more common than pancre-
atic tumors). For this reason, examination of the
gastrointestinal tract is important in all patients with
a sinonasal ITAC. In our patient, the results of an
upper GI series, barium enema study, and CT of the
abdomen were negative. Other tests that may be help-
ful in the differential diagnosis include staining for
carcinoembryonic antigen, with which strong positiv-
Intestinal-type adenocarcinoma should be considered in the differential diagnosis of skull base lesions with spread from the paranasal sinuses. These lesions may present with intracranial hemorrhages as in this case. These tumors occur primarily in workers in the hardwood or shoe industry.

**References**


