Scientific Research Publishing

Open Journal of Medical Imaging, 2025, 15(4), 220-224
https://www.scirp.org/journal/ojmi
ISSN Online: 2164-2796
ISSN Print: 2164-2788

A Case Report of an Adenoid Cystic Carcinoma of the Nasosinus with Orbito-Encephalic Invasion

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How to cite this paper: Rajaonarison, L.A., Haingomalala, Z., Herinirina, N.F., Andriambelo, R.H., Tsilizy, N.J., Andriamampionona, T.F., and Rakotoarisoa, A.H.N. (2025) A Case Report of an Adenoid Cystic Carcinoma of the Nasosinus with Orbito-Encephalic Invasion. *Open Journal of Medical Imaging*, **15**, 220-224.
https://doi.org/10.4236/ojmi.2025.154020

Received: September 25, 2025
Accepted: October 28, 2025
Published: October 31, 2025

Abstract

Background: Adenoid cystic carcinomas are malignant epithelial tumors that arise from the major and accessory salivary exocrine glands. They represent 0.15% of malignant tumors of the head and neck. Their localization in the nasal cavity is poorly understood and is rare. **Case Report:** We report the case of a 26-year-old patient who presented with a bilateral and progressive decrease in visual acuity, an increasing right lateronasal swelling with nasal syndrome and intracranial hypertension syndrome. Clinical examination found an extensive mass in the middle part of the right side of the face, with signs of local superinfection. We noted signs of bilateral compression of the optic nerves and bilateral oculomotor paralysis by involvement of the 3rd and 6th cranial nerves. Orbitofacial and cranial computed tomography performed with intravenous contrast confirmed the presence of an extensive tumor which was classified as T4bN2c. The anatomopathological exam of intranasal biopsy of the tumor suggests adenoid cystic carcinoma. Palliative care was put in place after a multidisciplinary decision due to the tumor's extensive invasion and unresectable nature. **Conclusion:** Adenoid cystic carcinomas remain tumors with a poor prognosis. Non-specific symptoms and slow evolution of this disease are a source of delayed diagnosis. Orbitofacial and cranial imaging could highlight the lesion and its extension. The anatomopathological exam could deter-

mine the histological nature of the tumor. The multidisciplinary decision fixes the patient's treatment.

Keywords

Adenoid Cystic Carcinoma, Computed Tomography, Nasal Cavity, Sinus

1. Introduction

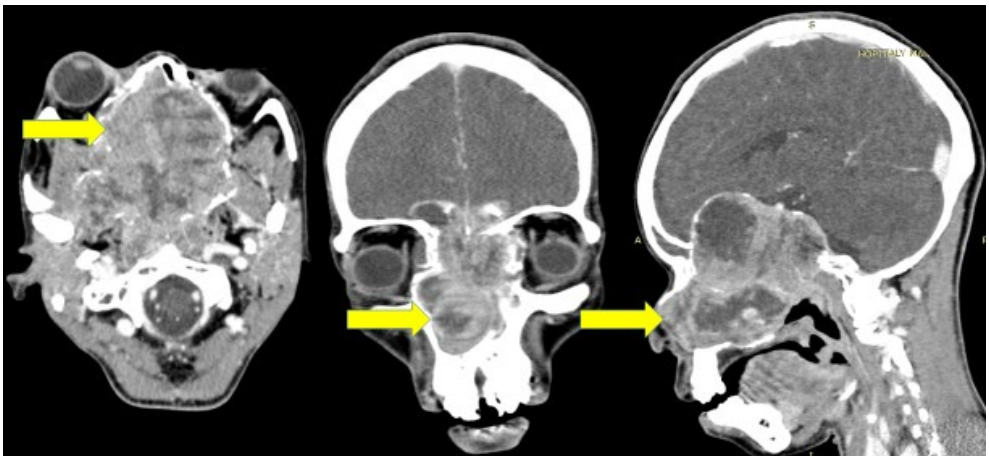
Adenoid cystic carcinomas are malignant epithelial tumors that develop from exocrine glands, primarily the major and minor salivary glands [1]. They represent approximately 0.15% of malignant tumors of the head and neck, and their localization to the nasal fossae is uncommon and not well characterized [2]. This cancer type is known for its slow and insidious progression [3]. Orbitofacial and cerebral imaging play a crucial role in evaluating the locoregional extension of the tumor [4]. We report a case of an invasive intranasal form due to the patient's unusually young age and the severity of the presentation.

2. Case Presentation

A 26-year-old Malagasy female farmer, with no history of alcohol or tobacco use, was admitted to the hospital due to a progressive bilateral decrease in visual acuity. This began with blurred vision in the right eye, followed by involvement of the left eye, culminating in complete loss of vision over a six-month period. She also presented with a progressive right lateronasal swelling, initially associated with bloody discharge that later became greenish and foul-smelling, along with anosmia. She reported persistent, bilateral fronto-parietal headaches that were worse in the morning, positional, and resistant to common analgesics (pain score 7/10 on a visual analog scale), accompanied by nausea that alleviated the headache. The entire clinical picture evolved in a febrile context.

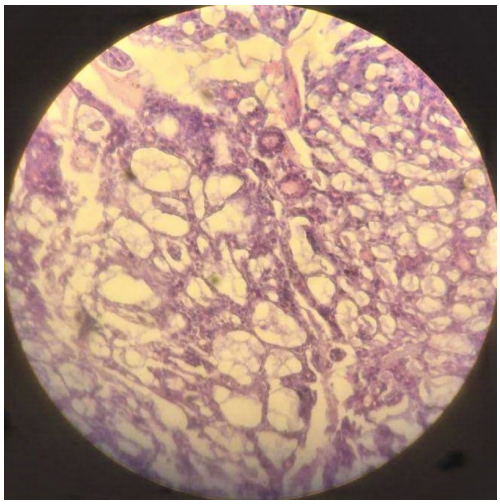
On clinical examination, the patient was asthenic with significant weight loss and a body mass index of 15.62 kg/m^2. Her temperature was 35.9˚C. ENT examination revealed a right lateronasal swelling that depressed the floor of the palatal vault, along with yellow-greenish purulent rhinorrhea. The right maxillary sinus point was tender, and there was a periapical abscess corresponding to the first maxillary molar. Subdigastric lymph nodes were palpable. Ophthalmological examination revealed bilateral exophthalmos, which was more pronounced on the right side. Visual acuity was reduced to no light perception bilaterally. The pupils were mydriatic and nonreactive. Ocular motility assessment showed complete bilateral paralysis of the third cranial nerve (ptosis, limitation of elevation, depression, and adduction, with areflexic mydriasis) and the sixth cranial nerve (limitation of abduction). Funduscopy revealed optic atrophy without edema bilaterally. There were no sensorimotor deficits in the limbs. The remainder of the physical examination was unremarkable.

Contrast-enhanced orbitofacial and cranial computed tomography (see Figure 1) revealed an invasive, heterogeneous mass with well-defined borders that enhanced after intravenous contrast administration. The lesion was consistent with a malignant mass of the nasal cavity, classified as T4bN2c (this indicates a very advanced, locally invasive tumor with extensive lymph node involvement). An endonasal biopsy was performed, and histopathological examination (see Figure 2) confirmed the diagnosis of adenoid cystic carcinoma. EBV serology was not performed.



An invasive mass demonstrating heterogeneous contrast enhancement, with a hypodense center. It measures 100 mm in anteroposterior diameter, 72 mm in height, and 73 mm in transverse diameter. This mass occupies the nasal fossa, bounded inferiorly by a bulging palate. Anteriorly, it lyses the wall of the right ala nasi. Laterally, it occupies both maxillary sinuses, lyses the medial orbital walls, and displaces the extraocular muscles, with compression of the optic nerve at the entrance to the optic canal. Superiorly, it lyses the ethmoid air cells and the cribriform plate of the ethmoid bone, and superiorly and posteriorly, the sphenoid sinuses and their walls. The brain is displaced superiorly.

Figure 1. Contrast-enhanced orbitofacial and cranial computed tomography in axial (A), coronal (B), and sagittal (C) planes.



Tumor proliferation with a cribriform architecture, showing a dual cell population. The majority of cells have a basaloid appearance and surround cavities containing pale, acellular material (black arrow). A minority of cells have more abundant cytoplasm and surround small gland-like structures (blue arrow).

Figure 2. Nasosinusal adenoid cystic carcinoma. Hematoxylin-eosin staining. Magnification ×200.

HIV serology was negative. Assessment for pituitary insufficiency was not conducted. The search for metastatic foci was limited to a chest X-ray and an abdominopelvic ultrasound, both of which were unremarkable.

Following a multidisciplinary consultation, the patient was placed under palliative care.

3. Discussion

Nasosinusal adenoid cystic carcinoma is a rarely described type of cancer [2]. Onset typically occurs around the sixth decade of life, with a female predominance, as reported in the multicenter study of 95 patients by M. Meyers *et al.* in 2016 [5]. Rare occurrences in younger patients may reflect underlying genetic susceptibility or early-life environmental exposures influencing tumorigenesis. The clinical presentation evolves slowly, often leading to diagnosis only when the tumor has reached a considerable size. Intracranial extension typically occurs along cranial nerve pathways.

The clinical manifestation of cancers of the nasal cavity and sinuses can involve a combination of three types of syndromes. The symptoms depend on the tumor's location and extension relative to the anatomical structures of the face and skull base. First, there is the nasal syndrome, where patients may present with nasal obstruction, mucopurulent rhinorrhea, or recurrent epistaxis. Second, the ophthalmological syndrome is characterized by edema of the upper eyelid, exophthalmos, or oculomotor paralysis. Third, the neurological syndrome is marked by the appearance of symptomatic facial neuralgia [6].

Contrast-enhanced facial and cranioencephalic computed tomography allows for the study of the tumor's characteristics and potential invasions to establish the TNM classification. Adenoid cystic carcinoma typically appears as an expansive mass, well or poorly defined, and heterogeneous after contrast injection [4].

Histopathological examination determines the tumor's histological nature and type. Three histological subtypes can be distinguished: cribriform, tubular, and solid. All these tumors are aggressive. T2-weighted MRI sequences can help differentiate between the cribriform type (T2 hyperintensity) and the solid type (T2 hypointensity) [7]. MRI also offers the possibility of assessing perineural tumor invasion. This perineural invasion, along with the solid histological subtype, constitutes features of poor prognosis in adenoid cystic carcinomas [4]. The staging workup for the tumor was limited, restricting our ability to identify potential metastatic sites. A contrast-enhanced thoracoabdominal-pelvic CT scan and/or a PET scan may assist in the detection of metastatic disease.

Therapeutic management is primarily based on surgery aimed at achieving complete tumor resection [5]. Adjuvant radiotherapy is often reported due to the tumor's propensity for recurrence [2]. In our context, given the highly invasive nature of this tumor observed at a stage of locoregional complication, palliative care was decided upon after multidisciplinary consultation. Furthermore, Madagascar has only one radiotherapy center, located 1200 kilometers from Antsiranana.

4. Conclusion

Adenoid cystic carcinomas are slow-growing tumors that are rarely described. Their symptomatology is nonspecific and depends on the involvement of the affected anatomical structures. The prognosis depends, on one hand, on the findings from contrast-enhanced facial and cranioencephalic imaging. Histopathological examination of the tumor determines its histological type. It is important to maintain a high index of suspicion for malignancy in young patients presenting with progressive and atypical sinonasal symptoms. A multidisciplinary team meeting, based on these investigations, will decide the therapeutic approach to be adopted.

Conflicts of Interest

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

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