Spontaneous Cerebrospinal Fluid Rhinorrhea as the Primary Manifestation of Maxillary Carcinoma
—Case Report—

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Abstract

A 66-year-old man presented with a maxillary carcinoma manifesting as unrelenting spontaneous cerebrospinal fluid (CSF) rhinorrhea. Anterior craniofacial resection of the tumor was performed with multilayered repair of the dura mater. Maxillary carcinoma usually manifests as nasal blockage, epistaxis, or a mass lesion. This case highlights the necessity for a high index of suspicion for malignant tumor and the need for meticulous repair of the dura mater to seal off the CSF leakage.

Key words: cerebrospinal fluid rhinorrhea, carcinoma, maxilla

Introduction

Tumors in the maxillary sinus usually present as a mass that may involve the surrounding soft tissue and bony structures. The primary clinical presentation usually involves nasal obstruction, epistaxis, intraoral growth, and cheek swelling, whereas spontaneous cerebrospinal fluid (CSF) rhinorrhea is extremely rare. We treated a patient with poorly differentiated squamous cell carcinoma of the maxillary sinus manifesting as spontaneous CSF rhinorrhea.

Case Report

A 66-year-old man presented with profuse, persistent watery discharge from the right nostril persisting for 6 months. He had a history of chronic smoking (15 cigarettes per day for 30 years) and tobacco chewing for 20 years. Computed tomography (CT) of the paranasal sinuses revealed a large homogeneously enhanced soft tissue mass in the right maxillary sinus, extending into right nasal cavity and eroding the medial wall of the right maxillary sinus. The tumor extended as far as the cribriform plate and had possibly caused erosion (Fig. 1). Biochemical examination of the nasal discharge was consistent with CSF. Endonasal biopsy of the tumor showed squamous cell carcinoma cells.

Anterior craniofacial resection was performed in July 2001. The tumor was approached through a lateral rhinotomy incision. The tumor involved the right maxilla, extended into right nasal cavity, and displaced the nasal septum to the left. Superiorly, the tumor extended up to the cribriform plate, causing multiple dehiscence of the floor of the anterior cranial fossa at the cribriform plate. A bicoronal scalp flap was raised and a bifrontal craniotomy was performed. The dura mater had densely adhered to the cribriform plate, but there was no intracranial...
Spontaneous CSF Rhinorrhea in Maxillary Carcinoma

Spontaneous CSF rhinorrhea was first reported in 1826 in a boy with progressively enlarging head. Autopsy revealed a large internal hydrocephalus with a fistula between the cranial cavity and the nose. CSF rhinorrhea is common following trauma or surgery involving the cranium. Less common causes include infection of the paranasal sinuses, osteomyelitis of the adjacent bone, and congenital anomalies of the brain and its membranes such as meningoceles or meningoencephaloceles. The CSF may be under normal or high pressure in such cases. Spontaneous high-pressure CSF rhinorrhea develops following sustained and chronic obstruction of the ventricular system, usually within the posterior fossa, and is always associated with a slow-growing or relatively static lesion.

Destructive lesions along the skull base may cause CSF rhinorrhea. Pituitary tumors are most frequently associated with spontaneous CSF rhinorrhea due to erosion of the sella turcica floor. Other tumors like meningiomas, meningoencephaloceles, craniopharyngiomas, cerebellopontine angle tumors, colloid cyst, osteomas, ethmoid-sphenoidal mucocoeles, malignant lymphoma, and nasopharyngeal tumors may also cause CSF rhinorrhea. A malignant tumor in paranasal sinuses usually presents with nasal blockade and epistaxis. Spontaneous CSF rhinorrhea in such a case is extremely rare. However, a high index of clinical suspicion of a malignant tumor in the paranasal sinuses is essential in patients presenting with unrelenting spontaneous CSF rhinorrhea.

The case of CSF rhinorrhea raises interesting issues regarding the pathogenesis and treatment strategy. The CSF rhinorrhea may have been a direct consequence of destruction of the anterior cranial fossa base in the region of the cribriform plate or a...
result of erosion of the membranous barriers separating the CSF pathways and the nasal cavity, in contrast to traumatic CSF rhinorrhea, which results from disruption of the skull base. The cribiform plate is an anatomically fragile portion of the anterior cranial fossa base vulnerable to the normal rhythmic variations in the CSF pressure. Anatomical studies have identified embryological variations in the cribiform plate and the olfactory nerves traversing the plate.\(^1,9\) The cribiform plate is a relatively ‘weak site’ allowing transmission of the CSF from the intracranial region to the nasal cavity, presumably related to constitutional or developmental factors including a persistent craniopharyngeal canal, a fistula in the retained embryonic lumen of the olfactory bulb, incompletely occluded holes around the olfactory nerves in the cribiform plate, or meningeal dysplasia in the region of the olfactory nerve.\(^1,3,9,15,16\) Any incident increasing intracranial pressure suddenly, such as sneezing or coughing, could force open an otherwise obscure anatomical defect and allow CSF leakage.\(^13\) Erosion of the cribiform plate and CSF rhinorrhea are indicators of malignant potential and aggressive nature of the tumor. The variation in the CSF flow dynamics at the tumor-arachnoid interface may accentuate tumor growth and expansion.\(^19\) The fairly large size attained by the tumor in a relatively short period, as in our case, may have been related to the persistent spontaneous CSF rhinorrhea, which was normal pressure CSF rhinorrhea as there was no obstruction to the ventricular or subarachnoid pathways.

The surgical management of spontaneous CSF rhinorrhea in our case was directed primarily towards extirpation of the malignant tumor and repair of the CSF fistula. The basal dura is thin and fragile in nature. Communication of the intracranial contents with the paranasal sinuses is a predisposing factor for meningitis in the postoperative period. Therefore, repair of the CSF fistula is mandatory in addition to the removal of the tumor. A multilayered reconstruction of the anterior cranial fossa was performed using pericranial and temporalis muscle-fascia flap after combined craniofacial resection of the tumor. A multilayered reconstruction of the anterior cranial fossa was mandatory in addition to the removal of the tumor. Multilayered reconstruction of the anterior cranial fossa using autogenous materials is mandatory in addition to complete resection of the tumor.

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