Case report

Leptomeningeal dissemination from an ethmoidal sinus adenocarcinoma in cerebrospinal fluid cytology

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Learning Point for Clinicians

Leptomeningeal dissemination of an ethmoidal sinus adenocarcinoma is a complication that may happen when the tumor involves the central nervous system. Cerebrospinal fluid cytology is useful for detecting malignant cells from primary and metastatic tumors that involve the central nervous system.

Case presentation

A 68-year-old man was admitted to hospital because of the presence of neurological symptoms (mainly frontal headache and confusion) and signs of meningeal irritation. According to his medical records, he was an ex-smoker (2 years ago) and he suffered from perennial rhinitis. The patient did not report labor precedents in relation to the exposure to wood, leather or others.

After physical examination, a lumbar puncture, computed tomography (CT) scans, and later, a trans-nasal endoscopic biopsy were performed. The patient’s condition worsened progressively and he finally died 10 days after his admittance to hospital. The autopsy could be performed.

The Papanicolaou-stained cytospin smears of the cerebrospinal fluid (CSF) showed tumor cells with enlarged nuclei and abundant vacuolated cytoplasm (Figure 1A). Numerous mononuclear cells and basophilic amorphous material were in the background. The vacuolated cells were also positive for the PAS stain.

CT scans revealed a sinonasal tumor of an apparent ethmoidal origin (Figure 1B).

The trans-nasal biopsy of the sinonasal tumor showed an intestinal-type adenocarcinoma (mucinous growth pattern) with single signet-ring-type cells within a mucoid background under a benign respiratory epithelium (see the inset in Figure 1A). A mucicarmin stain exhibited strong cytoplasmic positivity, and the tumor cells stained positive for cytokeratin 7, cytokeratin 20 and CDX2.

Based on the clinical setting and this pattern of immunostaining, a diagnosis of primary sinonasal adenocarcinoma was favored over one of metastatic adenocarcinoma from the gastrointestinal tract or from any other site. Autopsy confirmed the ethmoid sinus as the primary site of the tumor, showing an infiltrating growing pattern with abundant mucosubstance.

Discussion

This case illustrates the value of CSF cytology in conjunction with CT imaging for the diagnosis of...
leptomeningeal dissemination of ESA, a complication that has been reported.1,2 Adenocarcinomas of various types comprise 10–20% of all primary paranasal sinus cancers, and they are thought to be originated from seromucous glands of the nasal cavity and paranasal sinuses, as well as from the surface epithelium. After adenoid cystic carcinoma, intestinal-type sinonasal adenocarcinoma is the second most common type of adenocarcinoma of the sinonasal tract. Some tumors are able of exhibiting predominantly mucinous features containing signet-ring-type cells, which appear suspended in a pool of mucin signet-ring-type cells.

Because the distinction among this type of tumor and metastatic adenocarcinoma of intestinal origin and other primary adenocarcinomas of the sinonasal tract is important, immunohistochemical studies are mandatory. In a similar way, intestinal-type sinonasal adenocarcinoma stains for cytokeratin 20, CDX-2 and MUC2, but in contrast to colonic adenocarcinoma, it also stains for cytokeratin 7.3 Nevertheless, it has been mentioned by other authors that the pattern cytokeratin 7–cytokeratin 20, in a series of 14 cases, was not useful in the distinction between primary and metastatic intestinal adenocarcinoma.4 Neither the mucin profile (MUC2, MUC5AC, MUC5B and MUC6) for signet-ring cell carcinoma of intestinal-type adenocarcinoma has been useful to differentiate between primary and metastatic adenocarcinoma.5

In conclusion, sinonasal malignancies, particularly those of ethmoidal origin, are uncommon head and neck cancers, adenocarcinoma being one of the most frequent histological types. They appear, in most of cases, at an advanced stage of the disease. Leptomeningeal carcinomatosis is a fulminant clinical course condition, mortal in most patients, which can occur in metastatic disseminated cancers.

The majority of malignant cells observed in CSF cytology correspond to a metastatic dissemination, adenocarcinoma being the most frequent type of solid tumor. This case report highlights the role of CSF cytology in an accurate and promptly diagnosis, as this procedure helps avoid unnecessary or invasive procedures and discomfort to the patient, in spite of the poor prognosis that this tumor has.

Conflict of interest: None declared.

References

Figure 1. (A) CSF smear showing malignant vacuolated cells (Papanicolaou stain, ×400). The inset at the top right corner correspond to the biopsy of sinonasal tumor showing ‘signet-ring’ adenocarcinoma cells subjacent to benign respiratory epithelium (H&E stain, ×200). (B) CT scan demonstrating a tumor (white arrows) that involves the ethmoid sinuses. Destruction of the orbital medial wall is also observed (left arrow).