Primary ductal adenocarcinoma of the lacrimal gland: case report

Adenocarcinoma ductal primário da glândula lacrimal: relato de caso

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ABSTRACT

A 78-year-old man had double vision, painless palpable mass under the right superolateral orbital rim, downward displacement and restricted adduction of the right eye. His visual acuity was 20/50 OD and 20/20 OS. Hertel exophthalmometry was 21 mm OD and 17 mm OS. Computed tomographic scans showed an infiltrative orbital mass with ill-defined, irregular margins, involving the lacrimal gland and the lateral rectus muscle. The patient underwent an anterior transcunatanseal transeptotomy with incisional biopsy and surgical debulking. Intraoperatively, an ill-defined mass with infiltration of the lacrimal gland and the lateral rectus muscle was observed. Macroscopically, the mass measured 24 x 15 x 10 mm. The tumor components comprised duct-type structures with papillary and cribriform patterns, surrounded by prominent basement membrane. The tumor cells were positive for cytokeratin-7, matrix metalloproteinase (MMP)-2, MMP-9, MMP-13 and proto-oncogene Her-2/neu, but negative for cytokeratin-5, cytokeratin-20, p63, prostate-specific antigen, S-100 protein and thyroid transcription factor. These histopathologic findings were compatible with poorly differentiated ductal adenocarcinoma of the lacrimal gland, T3N0M0. Twenty-four months after orbital exenteration, microscopically, the tumor elements were characterized by large polygonal cells with vesicular nuclei, prominent nucleoli and amphophilic cytoplasm. The tumor components comprised duct-type structures with papillary and cribriform patterns, surrounded by prominent basement membrane. The tumor cells were positive for cytokeratin-7, matrix metalloproteinase (MMP)-2, MMP-9, MMP-13 and proto-oncogene Her-2/neu, but negative for cytokeratin-5, cytokeratin-20, p63, prostate-specific antigen, S-100 protein and thyroid transcription factor. These histopathologic findings were compatible with poorly differentiated ductal adenocarcinoma of the lacrimal gland, T3N0M0. Twenty-four months after orbital exenteration, microscopically, the tumor elements were characterized by large polygonal cells with vesicular nuclei, prominent nucleoli and amphophilic cytoplasm. The tumor components comprised duct-type structures with papillary and cribriform patterns, surrounded by prominent basement membrane. The tumor cells were positive for cytokeratin-7, matrix metalloproteinase (MMP)-2, MMP-9, MMP-13 and proto-oncogene Her-2/neu, but negative for cytokeratin-5, cytokeratin-20, p63, prostate-specific antigen, S-100 protein and thyroid transcription factor. These histopathologic findings were compatible with poorly differentiated ductal adenocarcinoma of the lacrimal gland, T3N0M0. Twenty-four months after orbital exenteration, microscopically, the tumor elements were characterized by large polygonal cells with vesicular nuclei, prominent nucleoli and amphophilic cytoplasm. The tumor components comprised duct-type structures with papillary and cribriform patterns, surrounded by prominent basement membrane. The tumor cells were positive for cytokeratin-7, matrix metalloproteinase (MMP)-2, MMP-9, MMP-13 and proto-oncogene Her-2/neu, but negative for cytokeratin-5, cytokeratin-20, p63, prostate-specific antigen, S-100 protein and thyroid transcription factor. These histopathologic findings were compatible with poorly differentiated ductal adenocarcinoma of the lacrimal gland, T3N0M0.
(Figure 1 A and B). Microscopically, the tumor elements were characterized by large polygonal cells with vesicular nuclei, prominent nucleoli and amphophilic cytoplasm (Figure 1B). No mucin was identified using periodic acid-Schiff stain (Figure 1C). The tumor cells displaying a high proliferation rate were positive for cytokeratin-7, matrix metalloproteinase (MMP)-2, MMP-9, MMP-13 and proto-oncogene Her-2/neu, but negative for cytokeratin-5, cytokeratin-20, p63, prostate-specific antigen, S-100 protein and thyroid transcription factor (Figure 1D, 1E, 1F). The histopathologic findings of this infiltrative mass were compatible with poorly differentiated ductal adenocarcinoma of the lacrimal gland, T3N0M0 according to the 2010 American Joint Committee on Cancer staging system. Twenty-four months after orbital exenteration, the patient was diagnosed with ipsilateral parotid gland and cervical lymph node metastases and died of disease.

DISCUSSION

Primary ductal adenocarcinoma of the lacrimal gland is a high-grade neoplasm arising from the ductal epithelium. This subtype of lacrimal gland adenocarcinoma was firstly described by Katz et al. in 1996(8). Since then, other cases have demonstrated an aggressive clinical behavior and poor prognosis(3-5,7). Of 7 patients with primary ductal adenocarcinoma, 2 had lymph node involvement (cervical lymph nodes(4) and mediastinal lymph nodes(3)), 4 had distant metastases (brain(5,7), common bile duct(5), liver(5), lung(5), pancreas(5), parotid(5), thyroid(5)).

Figure 1. Histologic sections of a lacrimal gland specimen from a patient with primary ductal adenocarcinoma. A) Low magnification showed tumor cells infiltrating the adjacent capsule and orbital soft tissues (Stain, hematoxylin-eosin; original magnification, 25x). B) High magnification revealed large polygonal cells with vesicular nuclei, prominent nucleoli and amphophilic cytoplasm (Stain, hematoxylin-eosin; original magnification, 200x). C) No mucin was identified (Stain, periodic acid-Schiff; original magnification, 200x).
D, E, F) Tumor cells were positive for matrix metalloproteinase (MMP)-2, MMP-9 and MMP-13 (Original magnification, 200x).
gland\(^4\), pelvic bone\(^3\), skin\(^3\) and vertebral column\(^3\) and 2 died of disease\(^3,5\).

In the current article, the tumor cells were positive for MMP-2, MMP-9 and MMP-13. The expression of matrix metalloproteinases is unknown in primary ductal adenocarcinoma of the lacrimal gland. MMP-2 was previously associated with worse prognosis in several types of salivary gland carcinoma\(^9\). In addition, MMP-9 and MMP-13 were specifically related to poor prognosis in duct salivary carcinoma, which is regarded as histologically and immunohistochemically similar to primary ductal adenocarcinoma of the lacrimal gland\(^10\).

Ophthalmologists and pathologists must remember that the lacrimal gland can develop aggressive tumors, such as primary ductal adenocarcinoma. Because there are few reports of lacrimal gland ductal adenocarcinoma, its clinical behavior, prognosis and treatment are still unknown. Therefore, early recognition of this new entity may support to delineate its management.

REFERENCES