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Trismus as the first manifestation of cholangiocarcinoma

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Abstract
The initial presentation of a cholangiocarcinoma (CC) as trismus due to metastasis to the parotid gland is extremely rare and no previous reports have been found in the literature. A 29-year-old woman presented trismus that initiated 2 months before admission, just after superior left third molar extraction. Physical examination revealed severe trismus and a mass in the parotid gland. Computed tomography (CT) showed a heterogeneous mass in the left parotid gland. The next day she presented jaundice, bilirubinuria, and fever, followed by massive bleeding, intravascular disseminated coagulation, and respiratory insufficiency. Two days later she died. At autopsy, the parotid tumor was identified as a metastasis from a CC of the extrabiliary tract. To our knowledge, this is the first reported case of a metastatic CC that initially presented as trismus due to a mass in the parotid gland. This case represents a rare but important diagnosis that otolaryngologists and oral surgeons should add to the catalog of uncommon causes of trismus.

Key words: *Parotid metastasis, cholangiocarcinoma, trismus, salivary gland neoplasms.*

Introduction
Cholangiocarcinomas (CCs) are malignancies of the biliary duct system. CCs tend to grow slowly and to infiltrate the walls of the ducts, dissecting along tissue planes. Local extension occurs into the liver, porta hepatis, and regional lymph nodes of the celiac and pancreaticoduodenal chains. Distant metastases to the skin, breast, bone, and cervical lymph node have also been reported (1,2) and usually occur several months after abdominal symptoms.
We report a unique case of a metastatic CC initially presenting as trismus due to a mass in the parotid region.

Case Report
A 29-year-old woman presented trismus just after upper left third molar extraction. The dental surgeon prescribed two courses of antibiotics and steroids during two months with no improvement. At the time the patient was referred to us, she presented severe trismus (5 mm of mouth opening) and a mass in the left parotid region. Computed tomography (CT) showed a 5 cm diameter heterogeneous mass in the left parotid gland that extended to the parapharyngeal space, but was clearly separated from other structures by a capsule (Figure 1). The next day she presented jaundice, clay-colored stools, bilirubinuria, fever, enterorrhagia, and upper right quadrant abdominal pain. Abdominal CT scan detected the presence of a biliary tract mass (Figure 2). Intensive care treatment was initiated but she developed massive bleeding, intravascular disseminated coagulation, shock, and respiratory insufficiency. Two days later she died.

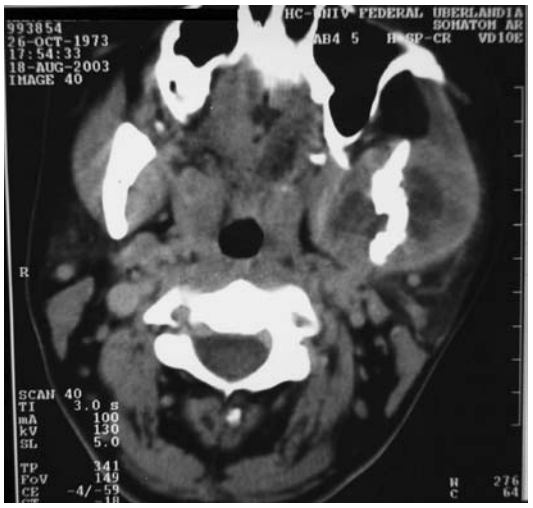

Fig. 1. Computed tomography of the neck showing a 5 cm diameter heterogeneous mass in the left parotid that extended to parapharyngeal space.

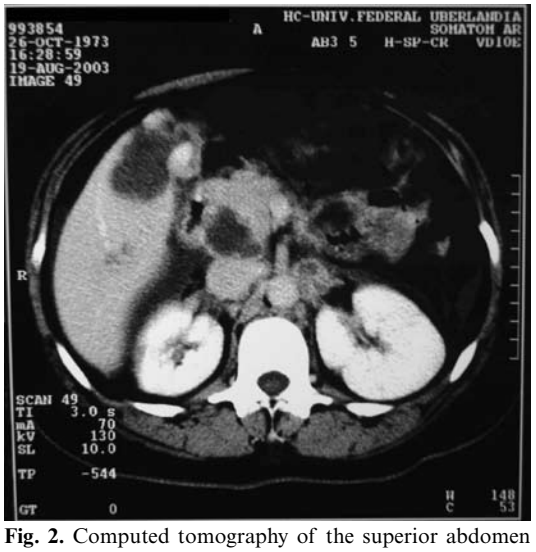

Fig. 2. Computed tomography of the superior abdomen showing a biliary tract mass.

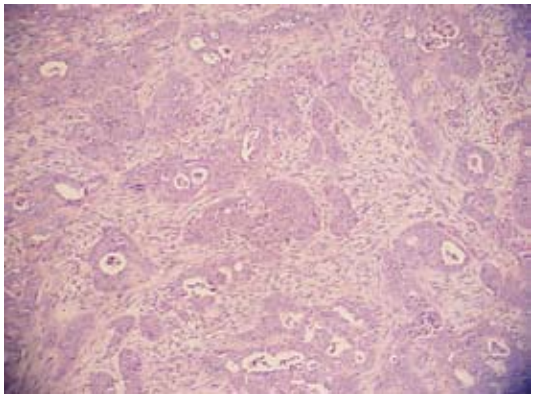

Fig. 3. Histologic examination of the parotid tumor showing infiltrating nests of mucous-producing cells with malignant features (H&E).

At autopsy, a primary tumor was found in extrahepatic bile duct and metastases were found in the left parotid, paraaortic, and peripancreatic lymph nodes, liver, colon, and heart. Histopathologic examination showed a similar pattern of adenocarcinoma in both biliary and parotid tumor. The cells were immunoreactive against cytokeratins-34ßE12, CK 8, CK 7, CK 19, CEA, and MOC-31, but were negative for CK 20 and vimentin. Thus, the parotid tumor was diagnosed as a metastasis from a CC of the extrabiliary tract (Figure 3).

Discussion

Metastases to head and neck area from abdominal organs via lymphatic spread (with exception of Virchow metastasis) are uncommon. They usually occur years after abdominal presentation and denote severity (3). The initial manifestation of an abdominal tumor as metastasis to parotid region is extremely rare and few cases were reported in the literature (kidney, prostate, liver, and colon) (4-8). A CC initially presenting as a metastasis to the parotid region has not been previously reported in the literature.

In the United States, each year, approximately 2,500 cases of CC occur. The incidence in most Western countries ranges from 2-6 cases per 100,000 individuals per year. The highest incidences occur in Japan with 5.5 cases and in Israel with 7.3 cases per 100,000 individuals. The male-to-female ratio is 1.5:1. Highest prevalence rates occur in the sixth decade of life (9).

Symptoms may include jaundice, clay-colored stools, bilirubinuria, pruritus, weight loss, and abdominal pain. Jaundice is the most common manifestation of bile duct cancer. The obstruction and subsequent cholestasis tends to occur early if the tumor is located in the common bile duct or common hepatic duct. Jaundice often occurs later in perihilar or intrahepatic tumors and is often a marker of advanced disease. Abdominal pain is relatively common in advanced disease and often is described as a dull ache in the right upper quadrant.

Despite aggressive anticancer therapy and interventional supportive care (i.e., wall stents or percutaneous biliary drainage), median survival rate is low since most patients (90%) are not eligible for curative resection (9).

The occurrence of a CC in a 29-year-old woman that presented trismus due to a mass in the parotid gland, with no jaundice, is extremely rare. The metastasis to the gland extended to masticatory and parapharyngeal spaces, affecting pterygoid, masseter, and buccinator muscles, and causing trismus. Differential diagnosis should include local infectious diseases, primary parotid tumors, parapharyngeal space tumors, and metastatic tumors of the neck (10-12).

In our opinion, the patient management was delayed due to incorrect diagnosis of the trismus. It is understandable because metastatic abdominal tumors to the parotid region are extremely rare and a challenging diagnosis, especially

without any mass at the initial presentation. In spite of this, the patient presented an advanced CC with poor prognosis that only palliative care could be performed.

Conclusions

To our knowledge, this is the only reported case of a CC metastatic to the parotid region that initially presented as trismus. This case represents a rare but important diagnosis that dentists, oral surgeons, and otolaryngologists should add to the catalog of uncommon causes of trismus.

References

1. Hardeman SH, Collins B, Lowe VJ, Solomon H, Stack BC Jr. Cholangiocarcinoma metastatic to the neck: first report of a case. Ear Nose Throat J. 2002 Nov;81(11):776-8.
2. Yiannakis PH, Patrikeos A, Ford HT, Davis CL. Metastatic cholangiocarcinoma with an occult primary. Clin Oncol (R Coll Radiol). 1995;7(6):394.
3. Mason AC, Azari KK, Farkas LM, Duvvuri U, Myers EN. Metastatic adenocarcinoma of the colon presenting as a mass in the mandible. Head Neck. 2005 Aug;27(8):729-32.
4. Melnick SJ, Amazon K, Dembrow V. Metastatic renal cell carcinoma presenting as a parotid tumor: a case report with immunohistochemical findings and a review of the literature. Hum Pathol. 1989 Feb;20(2):195-7.
5. Owens RM, Friedman CD, Becker SP. Renal cell carcinoma with metastasis to the parotid gland: case reports and review of the literature. Head Neck. 1989 Mar-Apr;11(2):174-8.
6. Simpson RH, Skálová A. Metastatic carcinoma of the prostate presenting as parotid tumour. Histopathology. 1997 Jan;30(1):70-4.
7. Romanas MM, Cherian R, McGregor DH, Wu Y, May CL, Baranda JC. Hepatocellular carcinoma diagnosed by fine-needle aspiration of the parotid gland. Diagn Cytopathol. 2004 Jun;30(6):401-5.
8. Velez A, Petrelli N, Herrera L, Lopez C, Mittelman A. Metastasis to the parotid gland from colorectal adenocarcinoma. Dis Colon Rectum. 1985 Mar;28(3):190-2.
9. Khan SA, Davidson BR, Goldin R, Pereira SP, Rosenberg WM, Taylor-Robinson SD, et al. Guidelines for the diagnosis and treatment of cholangiocarcinoma: consensus document. Gut. 2002 Nov;51 Suppl 6:VI1-9.
10. Alba García JR, Armengot Carceller M, Zapater Latorre E, Pérez Valles A, Basterra Alegría A. Malignant fibrohistiocytoma of the parotid region. Report of a case. Med Oral Patol Oral Cir Bucal. 2008 Feb 1;13(2):E148-50.
11. Suárez Roa Mde L, Ruíz Godoy Rivera LM, Meneses García A, Granados-García M, Mosqueda Taylor A. Solitary fibrous tumor of the parotid region. Report of a case and review of the literature. Med Oral. 2004 Jan-Feb;9(1):82-8.
12. Aguirre-Urizar JM. Salivary gland tumours in Mexico. 2003 Mar-Apr;8(2):81; author reply 82-3.

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