Solitary metastatic breast carcinoma in a trigeminal nerve mimicking a trigeminal neurinoma

Case report

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An unusual case of a metastatic adenocarcinoma located entirely within the trigeminal nerve is reported. The patient, with a history of breast cancer, presented with a pure trigeminal mononeuropathy. The neurological and neuroradiological findings in this patient were quite similar to those of a patient with trigeminal neurinoma. Surgery revealed that the tumor was located within the trigeminal nerve and its appearance was similar to that of a neurinoma. However, histopathological studies proved the tumor to be an adenocarcinoma that was related to the breast cancer treated earlier. A solitary metastatic tumor arising solely in a trigeminal nerve is quite rare; this is the first report of such a case metastasized from breast cancer.

KEY WORDS • metastatic breast cancer • solitary metastasis • trigeminal nerve • trigeminal neurinoma

Involvement of cranial nerves by metastatic tumors is uncommon, and isolated metastasis to a single cranial nerve is especially rare. We report a patient with a trigeminal mononeuropathy caused by a breast cancer metastasis entirely into the trigeminal nerve, mimicking a trigeminal neurinoma. Trigeminal mononeuropathy caused by neoplastic lesions is associated mainly with a trigeminal neurinoma, a cerebellopontine angle meningioma, or an invasive tumor of the neck and nasopharynx. Intracranial metastatic tumors manifesting with trigeminal mononeuropathy are quite rare, with only 23 such cases documented in the literature. In the majority of these cases, the mononeuropathy is not caused by a direct metastasis to the affected nerve but is secondary to diffuse metastases to the leptomeninges, dura mater, or cavernous sinus. Cases of trigeminal mononeuropathy caused by isolated metastasis to the nerve itself from a remote primary tumor are extremely rare, and to our knowledge, only one such case of lymphoma has been reported previously. This is the second reported case in which an isolated metastatic tumor arises solely in a trigeminal nerve.

Case Report

This 66-year-old woman presented to our institute on February 14, 1994, with a 5-month history of paresthesia on the left side of her face. She had had breast cancer, which had been totally removed 5 years prior to her admission. There were no findings of tumor recurrence at the original site. A postoperative follow-up study of tumor markers (α-fetoprotein and carbohydrate antigen 125) showed that they were within normal ranges, except for carcinoembryonic antigen, which was slightly elevated.

Examination. A neurological examination revealed a hypesthesia on the second and the third divisions of the left trigeminal nerve, without a trigeminal hyperactive dysfunction or a masseter muscle weakness. Examination of cerebrospinal fluid, including cytology, was negative. A plain craniogram showed no abnormality. Computerized tomography (CT) scanning with contrast enhancement disclosed a homogeneously enhanced mass at the left petrous apex that extended into the Meckel’s cave and the cerebellopontine angle. Magnetic resonance (MR) imaging revealed that the tumor had a dumbbell-shaped appearance and was homogeneously enhanced by an infusion of gadolinium (Fig. 1). Cerebral angiography demonstrated poor vascularity of the mass. Preoperative radiological diagnosis was compatible with trigeminal neurinoma.

Operation. During surgery, a left transpetrosal combined supra-infratentorial approach was undertaken to explore the whole mass. The tumor was located on the trigeminal ganglion and the ventral half of its cisternal segment. The main part of the mass was well demarcated,
yellowish, and soft with poor vascularity. No apparent adhesion or invasion was observed between the tumor and the surrounding tissues including dura, bone, and leptomeninges. The mass involving Meckel’s cave and the cerebellopontine angle was completely removed from the intact part of the nerve, and biopsy specimens of the dura and the leptomeninges around the tumor were obtained for further pathological examination. The intraoperative macroscopic appearance also suggested a neurinoma.

**Laboratory Studies.** Histopathological examination demonstrated the tumor to be a carcinoma. Carcinoma cells invading the trigeminal nerve fibers were clearly detected in the resected tumor specimen (Fig. 2). However, no microscopic tumor invasion was observed in the biopsy specimens from the dura and leptomeninges. In immunohistochemical examinations, the tumor cells stained positively for epithelial membrane antigen and keratin as markers for epithelial origin but were negative for S-100 protein, neurofilament, and glial fibrillary acidic protein (Fig. 3). Ultrastructural analysis verified that the carcinoma cells had intracytoplasmic lumens with microvilli, which are characteristic of adenocarcinoma (Fig. 4). Taking this evidence together, the tumor was diagnosed as a metastatic adenocarcinoma arising solely within the trigeminal nerve and originating from the breast cancer resected 5 years before.
Postoperative Course. Postoperative neurological study showed no change except for transient left trochlear nerve palsy. Computerized tomography scanning and MR imaging after surgery revealed no evident residual tumor.

Discussion

Common intracranial sites of metastatic cancer include brain parenchyma, dura, and leptomeninges; metastases in cranial nerves are unusual. According to the reports of 3359 autopsy cases at the Montefiore Institute, only 24 cases (0.7%) were found to have had metastasis in the cranial nerves. Although a clear mechanism was not documented, the sixth and the seventh cranial nerves were reported to be the most frequently involved, with the fifth cranial nerve being less frequently involved. Furthermore, details as to whether the metastasis was exclusive to the cranial nerves were not given in this report. Rubinstein reported six cases in which the first clinical symptom of intracranial metastasis was trigeminal mononeuropathy. Horton and Delaney also reported 16 such patients. However, all these 22 patients had diffuse tumor metastases in the brain or meningeal infiltration, which were detected during postmortem examination. Only one case of trigeminal mononeuropathy caused by a metastatic lymphoma located solely in the trigeminal nerve has been reported. This is the first reported case of a solitary metastasis from a breast cancer to the trigeminal nerve causing trigeminal mononeuropathy.

Because the metastatic route in this case is considered to be via blood flow, the mechanism and the rarity of the solitary metastasis into the trigeminal nerve should be discussed with special reference to the vasculature. Considering the blood supply to the trigeminal nerve root and ganglion, the root receives blood from the trigeminal arteries of superolateral pontine artery and peduncular cerebellar artery, and the ganglion is furnished by the branches of both the internal and the external carotid artery. All these arteries also supply neighboring structures such as the dura covering the ganglion and the brain parenchyma. Therefore, the chance of isolated metastasis only within the nerve may be quite low. Another hypothesis for its rarity may be due to a microanatomical feature: the trigeminal nerve is tightly covered with the dura at the ganglion and with the arachnoid and leptomeninges at the cisternal segment. If a metastasis occurs within the nerve, tumor cells will soon infiltrate and spread to the surrounding tissues because of their invasive nature. Also, it is difficult to diagnose the mass neurologically or radiologically at the early stage before it disseminates diffusely around the nearby structures and other symptoms become manifest.

Radiologically, the tumor in the present case showed a dumbbell shape, mimicking a trigeminal neurinoma. Neurinoma also occurs frequently in this location and has a dumbbell shape; it extends from the middle to the posterior cranial fossa and is reported to be found in approximately 20% of all trigeminal neurinomas. A Meckel’s cave meningioma must also be considered in the differential diagnosis, although its incidence is less frequent than that of the trigeminal neurinoma. Approximately one-eighth of Meckel’s cave meningiomas are reported to show a dumbbell shape. Neurologically, the present case manifested as a trigeminal mononeuropathy, consisting of facial hypesthesia. The most common symptom in the patient with a trigeminal neurinoma is de-
creased facial sensation, followed by trigeminal neuralgia, and trigeminal motor weakness.\textsuperscript{12,13,16} Therefore, from a neurological standpoint, it is difficult to distinguish the tumor of the present case from a trigeminal neurinoma.

Surgery via a transpetrosal combined supra–infratentorial approach was useful to expose and resect the whole tumor extending in both middle and posterior fossae.\textsuperscript{1,16} Intraoperatively, the findings in this tumor were quite similar to those of a trigeminal neurinoma. It might be because the mass was explored before it infiltrated into the surrounding tissues.

The histological studies were striking in demonstrating the tumor infiltration into the nerve fibers. De Pena, \textit{et al.},\textsuperscript{5} were the first to report a case with a focal metastasis to the trigeminal nerve. Although radiological features of their case are described in detail in that article, histological evidence was not given to prove the isolation of the tumor within the nerve. The present study is the first report of an isolated metastasis within the trigeminal nerve that was verified surgically and histopathologically. Finally, we would like to emphasize that metastasis to a cranial nerve should be borne in mind as a differential diagnosis when a patient with a history of malignancy displays a mononeuropathy.

\textbf{References}


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