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Article

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Surgical treatment of rare metastatic tumor in Meckel's Cave: a case report

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Abstract: *Objectives*: Metastases in Meckel's cave are a rare tumor entity, but they should be considered in patients with a known primary malignancy and who complain of trigeminal neuralgia. *Methods*: The case of a patient with a left trigeminal neuralgia caused by a metastatic tumor involving the Meckel's cave and Gasserian ganglion is reported. Preoperative symptoms included headache, trigeminal hyperesthesia and facial pain. *Results*: The patient underwent microsurgical resection of the tumor, obtaining a histological diagnosis and a resolution of the pain. Trigeminal motor function was preserved. *Conclusion*: The best treatment for tumors of Meckel's cave, including metastases, is complete microsurgical removal. Although good results have been reported with radiosurgery, this treatment should be reserved only for nonresectable and residual tumors.
Key words: metastases; Meckel's cave; trigeminal neuralgia; microsurgery

Introduction

Tumors of Meckel's cave involving the Gasserian ganglion are usually benign and are either meningioma or trigeminal neurinomas (1,8). They represent less than 0.5% of all intracranial tumors (4). Patients can complain of a variety of symptoms, but most of them present with dysfunction of the fifth cranial nerve (14).

Metastases in Meckel's cave are a rare tumor entity and should be considered as important differential diagnosis, especially if the patient suffers from a known primary malignancy. We report the case of a patient with left atypical trigeminal neuralgia caused by a metastatic tumor that involved the Meckel's cave. He was previously diagnosed with a pulmonary neoplasm.

Case report

A 50 years old man presented with a six months history of burning and stabbing pain around the left infraorbital ridge and cheek. The pain was limited in the territory of the distribution of the trigeminal nerve. During the last month, the frequency and severity of episodes increased, without occasional spontaneous remissions as before. All the symptoms were unilateral. The patient had undergone several therapeutic attempts to control his pain. Previous treatment included dental extractions, diphenylhydantoin and carbamazepine administration. He had normal findings on physical examination, presenting no objective trigeminal motor loss, but a mild hyperesthesia in the left side of his face.

Cerebral computerized tomography (Figure 1) showed a 3x2,5 cm. contrast enhancing lesion in Meckel's cave on the left, without erosion of the apex of the petrous bone, but with little extension in the posterior fossa through the tentorial incisure. Axial T1-weighted MRI with gadolinium enhancement (Figure 2) revealed a relatively well-defined mass in Meckel's cave, with less peritumoral edema. The tumor uniformly enhanced gadolinium and extended to the left cavernous sinus without infiltrated it, and down to the cerebello-pontine angle.

During his hospitalization, the patient was diagnosed, due to a preoperative chest radiography with a right lung lesion. We performed a contrast chest computed tomography which revealed a large right lung and a hilar mass, with smooth, lobulated margins, having around 10 cm. in the largest diameter (Figure 3).

The patient underwent a left temporal craniotomy and a subtemporal intradural approach to the tumor. The craniotomy was enlarged to the base of the temporal fossa to obtain a flat viewing angle across the floor of the middle fossa. We performed a microsurgical complete resection of the supratentorial portion of the tumor, except for the portion that adhered to the inferior margin of the tentorial incisures (Figure 4). The tumoral mass intimately adhered to the dura mater that overlying the Meckel's cave and Gasserian ganglion. It was debulked and its capsule was dissected from the dura (Figure 5). The dura mater was incised, but no extradural extension was noted. Without drilling the petrous apex we did not properly exposed the posterior fossa portion.

The histological diagnosis was metastatic carcinoma with neuroendocrine differentiation (Figure 6). Postoperatively, the patient did well, but continued to have a partial fifth nerve dysfunction involving the second division. He presented episodic double vision, for the next two month, due to a mild sixth nerve paresis.

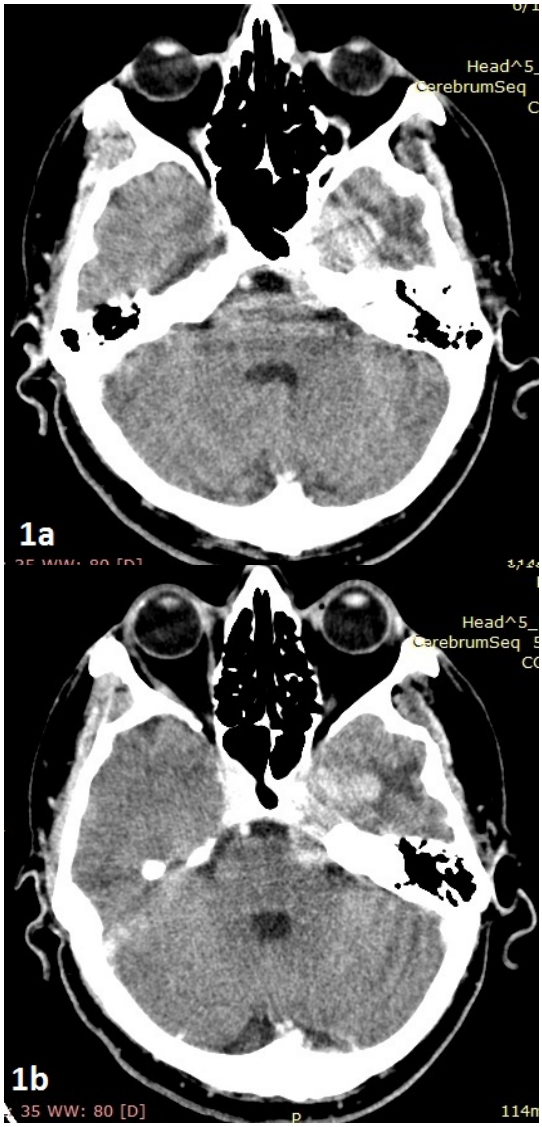

Figure 1 (a, b) - Preoperative axial contrast CT scan showed a relatively not well circumscribed tumor, with little peritumoral edema, which enhanced the contrast and extended down in the left cerebello-pontine angle

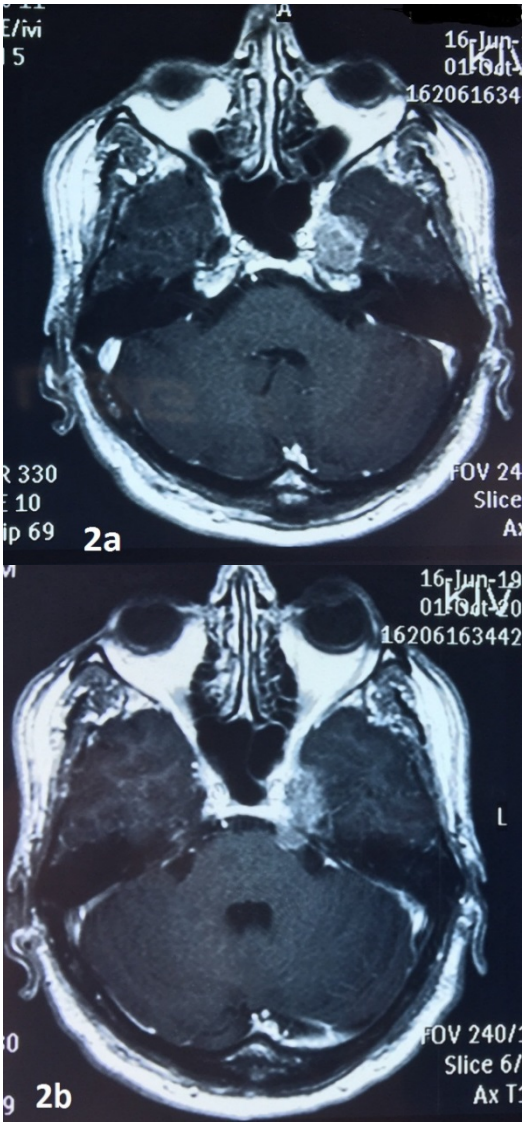

Figure 2 (a, b) – Axial T1-weighted images with gadolinium enhancement showed a 2,5x3 cm diameter tumor, with relatively uniform enhancement of gadolinium contrast, located in the left Meckel's cave, extending to the left cavernous sinus without infiltrated it, and down to the cerebello-pontine angle

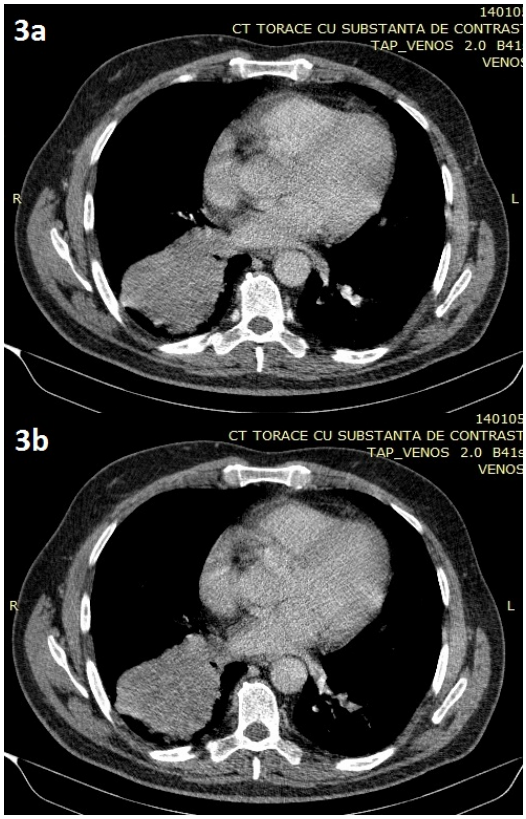

Figure 3 (a, b) - Nonenhanced CT scan of the chest showed a large right lung and a hilar mass, with smooth, lobulated margins, having around 10 cm. in the largest diameter, and extending right near the pleura

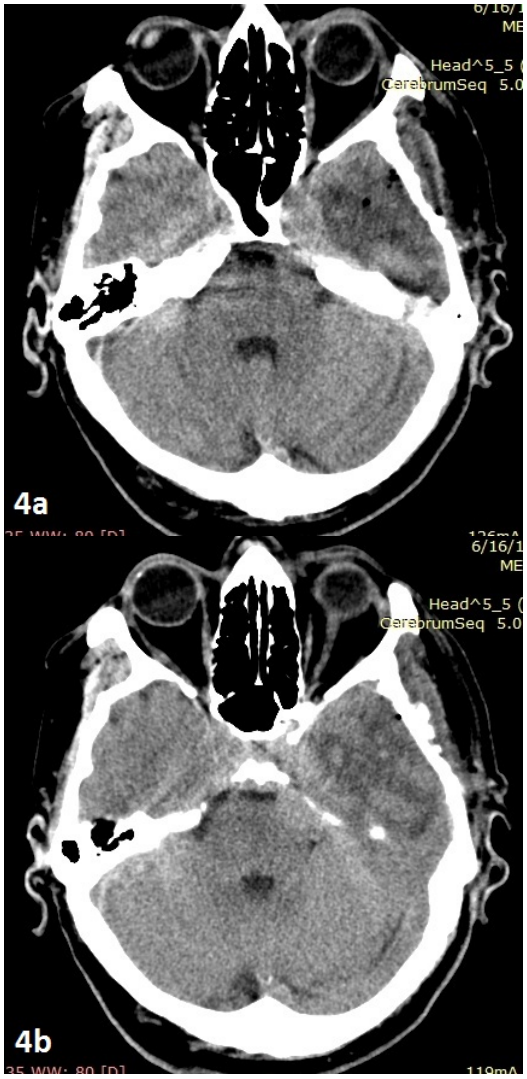

Figure 4 (a, b) – Control native CT scan performed 6 days after surgery. The rest of the tumor attached to the inferior margin of the tentorial incisura was noticed

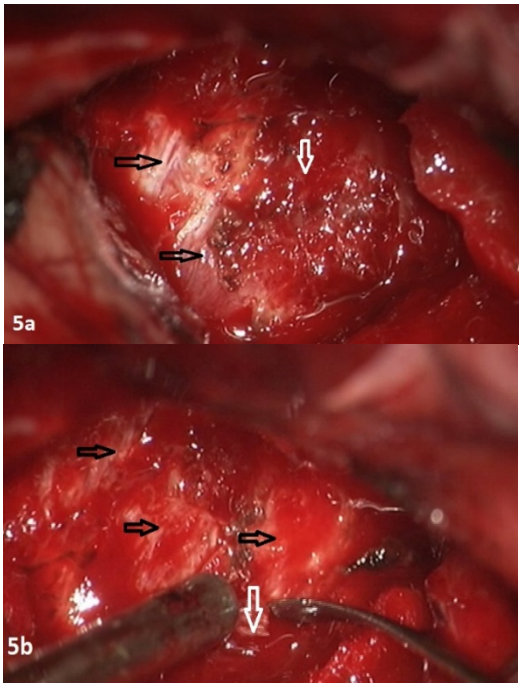

Figure 5 (a, b) - Intraoperative images obtained under microscope during a subtemporal intradural approach. (a) – black arrows: tentorium; white arrow- microscopic aspect of the tumor overlying Meckel's cave; (b) – black arrows: tentorium after removing the tumor over the gasserian ganglion; white arrow – the rest of the tumor extending inferiorly into the CPA

Discussion

The most common tumors in Meckel'cave are neurinomas and meningiomas (3, 4). Besides this two entities, other types of pathological lesions have been reported in the region of the gasserian ganglion: metastatic carcinomas, sarcomas, arteriovenous malformations and amyloidomas (2, 4). We presented the case of a patient harboring a metastatic tumor in the Meckel's cave, who had been previously diagnosed with a lung primary malignancy.

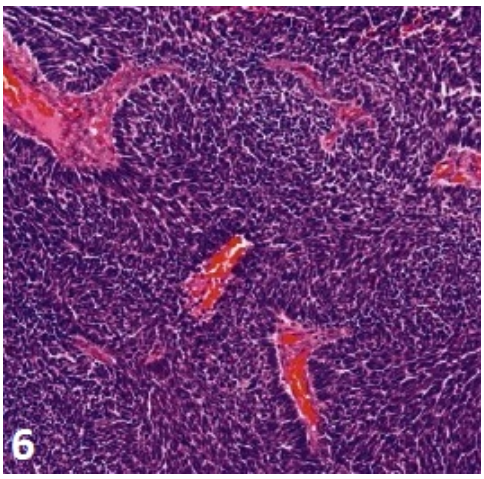

Figure 6 - Hystopathology of the tumour (Hematoxylin-Eosin): metastatic carcinoma with neuroendocrine differentiation; typical histopathology includes nests of small primitive appearing cells with hyperchromatic nuclei, high nuclear to cytoplasmic ratio, mitotic figures and necrosis

The incidence of brain metastases is difficult to determine with precision. In earlier neurosurgical series the overall incidence of cerebral metastases was 20%- 30% for all patients with systemic cancer (16). Estimates based on more recent series and autopsy studies show a much higher incidence of brain metastases, with incidences varying between 8.3 and 11.1 per 100.000 individuals (11). Approximately 40% of intracranial neoplasms are metastatic (7). These estimates place brain metastases first in frequency among all intracranial tumors, before intracranial glioma and meningioma (26).

The histological type of the primary tumor appears to be the major factor of the frequency and pattern of intracranial spread. Multiple, large autopsies series suggest that, in order of

decreasing frequency, lung, breast, melanoma, renal and colon cancer are the most common primary tumors that metastasize to brain (7, 11, 26). Primary lung tumors account for 30% to 60% of all brain metastasis cases (8). Breast cancer ranks second, contributing 10% to 30% (12) of all brain metastases among women (19), and renal cancer has a frequency of 11% (24). When the ability of a primary tumor to spread to the brain is considered, melanoma ranks first followed by lung and breast cancers (7, 15).

The first sign of a tumor of Meckel's cave is usually that of trigeminal nerve dysfunction. Facial hypalgesia along the distribution of the trigeminal nerve is the most common symptom. This is usually constant and burning in nature. A diminish corneal reflex and motor involvement of the fifth cranial nerve usually follows. Not all tumors of the gasserian ganglion presented with pain. According to Jefferson (10), those tumors located in the posterior fossa, compressing the sensory root, are thought to be typically painless, with posterior fossa and pyramidal signs. In our case, the tumor had little extension down in the cerebello-pontine angle, most of it compressing the gasserian ganglion in the Meckel's cave. That's why our patient presented with stubbing facial pain and numbness, especially involving the second division of the trigeminal nerve, and no pyramidal signs were noticed.

Computerized tomography (CT) and magnetic resonance imaging (MRI) permit the detection of small tumors in the region of Meckel's cave. MRI study gives the best information concerning the localization and extent of the tumor in addition to revealing the displacement of neighboring structures and involvement of the cavernous sinus and vessels (20). Sometimes, the limits of the tumor were very indistinct on CT scan, and only the presence of bone erosion suggests a mass lesion (2). In low-density tumors, such as lipoma, epidermoid cyst or chordoma, magnetic resonance imaging was especially useful. In our case, axial MRI T1-weighted images with gadolinium enhancement showed a tumor, which enhances the contrast relatively uniform, located in the left Meckel's cave, extending to the left cavernous sinus without infiltrated it, and down to the cerebello-pontine angle. Differential diagnosis must include meningiomas, neurinomas, epidermoid cysts, metastasis, chordomas, chondrosarcomas and maxillary sinus tumors (23).

The surgical approach usually depends on the extent of the tumor. The main part of the tumor is located in the middle fossa but it may have extracranial extension or posterior fossa extension. These modifications are important to assure the best surgical approach for each type of lesion. These tumors can be resected via an extradural approach or an intradural approach. Most of them are extirpated via a middle fossa approach, either intradurally (for large lesions) or extradurally (for small lesions). The degree of difficulty in radical surgical removal depends on the particularities of each case, but, mainly, it depends on anatomical extension of the tumor. In order of their frequencies, the most used approaches are: middle fossa approach, retrosigmoid approach and presigmoid approach (20). The

last two approaches are especially used in the resection of tumors with important extension in the posterior fossa. In cases of small extranevraxial lesions, such as meningiomas and neurinomas, a safe surgery could be performed only by using an extradural approach (20, 23).

The majority of tumors in Meckel's cave location are amenable to be resected via the subtemporal intradural approach. Hyperventilation and intraoperative spinal drainage reduced brain bulk, and temporal bone retraction was minimal. The tumor could extend from Meckel's cave to the superior orbital fissure and down to the cerebello-pontine angle. The mass is usually well circumscribed and total removal can be achieved, leaving only remnants of capsule adherent to the fifth cranial nerve. Under microscope magnification, a dural opening must be made parallel to the tentorial edge in order to avoid cutting across fibers of the trigeminal nerve that lie directly beneath. Most of the tumors in this location are benign and can be totally resected. When flecks of capsule from the tumor is adherent to nerve fibers and brain stem, or is important extension in the CPA, attempts to remove the entire tumor would significantly increase the neurological deficits (5). For the rest of the tumor, fractioned radiation therapy is used.

Using microsurgical techniques, the outcome of tumors involving Meckel's cave and gasserian ganglion is usually favorable. The most frequent symptom after surgery is trigeminal hypesthesia, which is transient in most cases. Facial pain may persist after surgery, but most patients report improvement or total relief during follow-up. Patients who had experienced preoperative atypical trigeminal pain reported a resolution of their symptoms after surgery. Diplopia, CSF leak or even meningitis have been also described as possible complications (20, 21). Most new cranial nerve deficits present resolution within 4-6 months (22). Several contemporary series have demonstrated no deaths or major surgical complications with radical removal of tumors in the Meckel's cave and gasserian ganglion (6, 21).

For small, nonresectable and residual tumors, good results have been reported with radiosurgery (17, 18). Long-term follow-up of patients treated with this method is still needed to evaluate the exact role of the radiosurgery in the late control of these lesions (17, 18).

Lesions in Meckel's cave can have a varied and unusual presentation, as well as an assortment of pathology. Beside neurinomas and meningiomas, metastatic tumors have been reported occupying Meckel'cave and compressing gasserian ganglion. Metastatic involvement of the Meckel's cave occurs either via hematogenous dissemination from distant metastatic or perineural spread from tumors of the head and neck. Very few cases of metastatic involvement of the Meckel's region have been published. On a meta-analysis (2010) of malignant tumors involving this region, Soni RC et al. (23) found 20 cases, including theirs, of malignant involvement of Meckel's cave described in the literature. Most primary neoplasm involved were: small cell lung cancer (2 cases), breast cancer (4 cases), adenocarcinoma of colon (2 cases), lymphoma

(4 cases), renal cell cancer (1 case) and folicullar thyroid cancer (1 case) (9, 13, 25).

Conclusion

Malignant lesions involving the structures of Meckel's cave are uncommon and may present later in life compared to meningioma and schwannoma. We underline the importance to thoroughly investigate the patients early for a possible primary as well as metastases, in those patients found to present a lesion in Meckel's cave. Moreover, it is difficult to distinguish metastatic tumors from benign tumors solely based on clinical or radiographic characteristics. We believe that the best treatment is microsurgical removal of the metastases, followed, when necessary, by fractioned radiation therapy for any rest of the tumor.

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