

Case Report

Meckel's cave and bilateral internal auditory canal metastasis of adenocarcinoma of unknown primary

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Abstract

Adenocarcinoma of unknown primary presenting as metastasis to Meckel's cave and bilateral internal auditory canals is rare. We report a case of a 65-year-old chronic smoker presenting with otological symptoms and trigeminal neuralgia who, on neuroimaging, revealed metastatic deposits to bilateral internal auditory meatus (IAM) and also Meckel's cave.

Introduction

Lesions of the cerebellopontine (CP) angles and internal auditory canal are mostly benign, with acoustic neuromas accounting for 80-90%, the others being meningioma, primary cholesteatoma, glomus jugulare tumors, facial neuromas, trigeminal neuromas and arachnoid cysts¹. Malignant lesions make up less than 1% of all lesions¹, of which metastasis to the CP angle and internal auditory meatus (IAM) are rare^{2,3,4}. Similarly lesions in Meckel's cave tend to be schwannomas and meningiomas, with metastasis being an unusual entity^{7,9}. We present a rare case of metastasis to bilateral internal auditory meatus and also Meckel's cave, of adenocarcinoma of unknown primary.

Case report

A 65-year-old lady, under the care of medical team for exacerbation of chronic obstructive pulmonary disease (COPD) was referred to Ears, Nose and Throat (ENT) team for assessment of 4-5 weeks of right sided earache, bilateral reduced hearing (worse in the right ear than left), right facial paraesthesia and maxillary pain,

retromastoid pain and headache. She also complained of dizziness, which she attributed to her loss of appetite and subsequent weight loss, but denied any true rotatory vertigo. She had no nasal symptoms, odynophagia, dysphagia or neck swellings. Her symptoms had persisted despite antibiotics for a possible ear and sinus infection by her general practitioner (GP). She was a chronic smoker of 25 pack years. Neuro-otological examination was normal except for right middle ear effusion. Nasoendoscopy revealed a normal post nasal space, larynx and hypopharynx. Pure tone audiometry (PTA) showed moderate mixed hearing loss on the right, with a sensorineural hearing loss on the left side. As she was a chronic smoker and her symptoms were not explained solely by unilateral otitis media with effusion, she had several radiological imaging. An initial MRI Head reported normal intracranial appearances with right middle ear fluid and also picked up suspicious T1, T2 isointense lesions in bilateral intracanalicular regions. A subsequent MRI IAM post-gadolinium demonstrated enhancement within the three soft tissue nodules within both the IAMs measuring 8x5 mm each and right Meckel's cave measuring 15x7 mm.

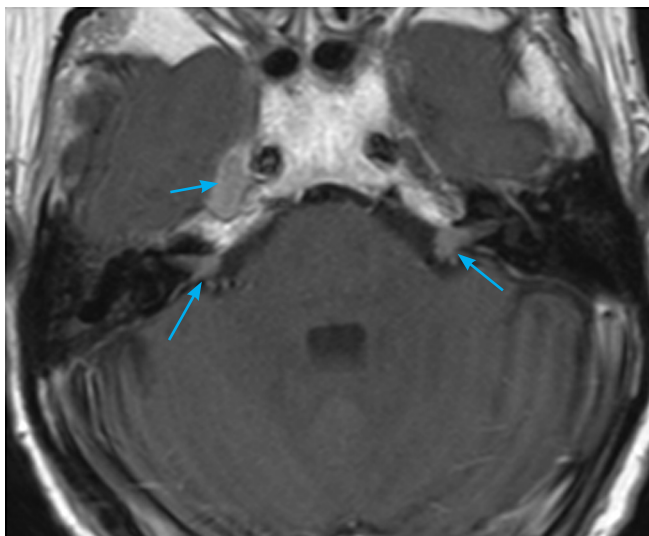


Fig 1. Transaxial T1-weighted post-gadolinium sequence demonstrating enhancement within the three soft tissue nodules within Meckel's cave on the right and within both IAMs (arrows)

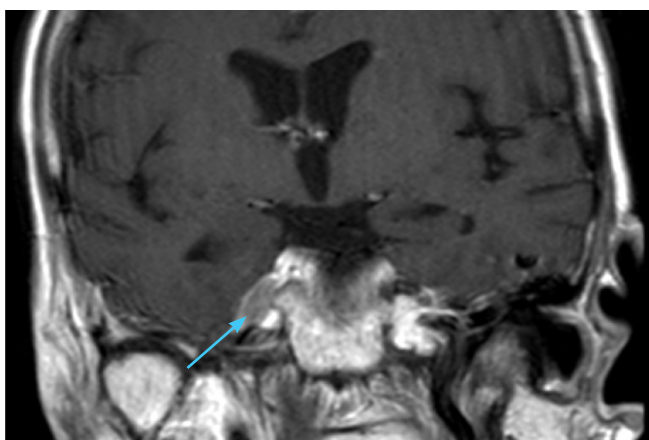


Fig 2 Coronal T1-weighted post-gadolinium sequence demonstrating enhancement within the soft tissue nodules within Meckel's cave on the right (arrow).

With multiple schwannomas less likely to be the case clinically, there was high index of suspicion for multiple cranial nerve deposits with the primary still under investigation. A concurrent computed tomography (CT) neck and chest showed emphysematous lung with small volume paratracheal nodes, subcarinal node and right hilar node but no discrete focal lung lesion. An additional note of likely metastatic patchy lucency and sclerosis was made in L1 vertebral body the nature of which was planned to be ascertained with an MRI spine. She had an endobronchial ultrasound (EBUS) fine needle aspiration (FNA) of 7 mediastinal lymph nodes showing CK7 and TTF1 positive metastatic adenocarcinoma, strongly suggesting a lung primary. A pipelle biopsy of a

distended endometrial cavity detected on CT abdomen and pelvis excluded malignancy. Cerebrospinal fluid (CSF) obtained from a lumbar puncture showed no malignant cells. Over a period of weeks whilst still under investigation, she developed a right facial palsy. Although a definite lung lesion was not found, it was considered a likely primary causing the metastasis to bilateral IAM and right Meckel's cave. Palliative treatment mainly for pain relief in the form of whole brain radiotherapy was started with chemotherapy being deferred due to her poor general condition. She passed away after two cycles of radiotherapy and before MRI spine could be carried out.

Discussion

Malignancy presenting as one or more metastatic intracranial lesion can range from 5 to 12%⁹ however either IAM or Meckel's cave is a very unlikely site.^{4,5} If at all they occur, the primary could be lung, colon, breast and kidney for both while prostate primary could metastasize in IAM and thyroid, ovary, lymphoma and melanoma in Meckel's cave^{2,5,7,8} There are only a few unilateral and bilateral IAM metastasis¹⁻⁵ and unilateral Meckel's cave metastasis cases being reported separately in the literature mostly with a background of known primary^{7,8,9}. However, there is no reported case of simultaneous involvement of bilateral IAMs and Meckel's cave metastasis.

Detecting metastasis as a first sign of the disease can be challenging as it shares similar clinical and radiological characteristics of a relatively common internal auditory canal (IAC) and Meckel's cave lesion^{2,7,8}. Rapid progression of hearing loss associated with facial weakness and vertigo can hint towards metastasis as opposed to benign lesions in the IAM, which mostly presents with progressive or sudden hearing loss but with facial nerve palsy only in a few instances^{2,3,4,5,8}. Another characteristic feature is the severe unremitting pain localised to the mastoid and retromastoid area requiring narcotics^{3,5}. Metastasis to the Meckel's cave can mimic trigeminal neuralgia which can be a presentation of trigeminal schwannoma or meningiomas also^{7,8}. Soni et al.⁹ analyzed clinical features of 21 patients with malignant tumor of the Meckel's cave and compared them with those of meningioma and schwannoma of trigeminal nerve. Pain and paresthesia were more common in malignant lesion with mean age of presentation being 52 as compared to 42 and 37 consecutively for meningioma

and schwannoma of trigeminal nerve. Our patient presented with facial pain and paresthesia along with mastoid and retromastoid pain requiring pregabalin and morphine for pain relief. These symptoms are similar to those described for metastatic lesions of IAM and Meckel's cave.

Spread from the primary to the temporal bone and Meckel's cave can occur through three routes: haematogenous dissemination, direct neoplastic extension from adjacent areas or diffuse metastatic leptomeningeal carcinomatosis when tumor cells gain access to the CSF^{1,7}. However, isolated metastasis are more likely to have hematogenous spread which in itself is rare^{3,8}. There was no sign of direct extension or leptomeningeal metastasis on MRI with normal CSF further supporting a hematogenous spread in our case.

There are no distinctive MRI characteristics of IAM metastasis differentiating it from a benign lesion in the IAM⁴ or Meckel's cave⁹. Commonly, vestibular schwannoma shows homogenous and isointense signal compared to gray matter on both T1- and T2-weighted images and exhibits strong post-gadolinium enhancement. The presence of adjacent thick linear and extranodular contrast enhancement on MRI may favour metastasis^{4,6}. High resolution CT scan does not provide additional information in aiding diagnosis². Although our patient had radiological findings mimicking multiple schwannomas, the clinical scenario of a heavy smoker and no features suggestive of NF2 lead to high index of suspicion for metastasis. This was further reinforced by EBUS FNA of the mediastinal lymph nodes which suggested metastatic adenocarcinoma favouring a lung primary.

Combined chemoradiotherapy is the preferred treatment for multiple skullbase metastasis. However, it carries a very poor prognosis of only a few months.²

Conclusion

Metastasis in the IAM and Meckel's cave are very rare. Inner ear symptoms with rapid progression, along with a facial nerve palsy can indicate a metastatic IAM lesion whereas trigeminal nerve neuropathy could be a presentation for metastasis in the Meckel's cave. There are no specific radiological findings hence the diagnosis is mainly based on clinical evaluation. Metastasis should also be considered a differential diagnosis in lesions in the IAM and Meckel's cave even in absence of a previous known malignancy.

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Conflict of interest: None declared.

Reference

1. Schrock A, Laffers W, Bootz F. Solitary metastasis of lung carcinoma to the internal auditory canal. *Am J Otolaryngol*. 2006 May-Jun; 27 (3):214-6.
2. Falcioni M, Piccirillo E, Di Trapani G, Romano G, Russo A. Internal auditory canal metastasis. *Acta Otorhinolaryngol Ital*. 2004 Apr; 24(2):78-82.
3. Yamakami I, Oishi H, Iwade Y, Yamaura A. Isolated metastases of adenocarcinoma in the bilateral internal auditory meatuses mimicking neurofibromatosis type 2—case report. *Neuro Med Chir (Tokyo)* 1999 Oct; 39(11):756-61.
4. Kim C-H, Shin JE, Kim HJ, Lee KY. Bilateral Internal Auditory Canal Metastasis of Non-small Cell Lung Cancer. *Cancer Research and Treatment : Official Journal of Korean Cancer Association*. 2015; 47(1):110-114.
5. Loo SW, Dean AF, Murray P. Internal auditory canal metastasis mimicking a vestibular schwannoma at presentation – a case report and review of the literature. *International Seminars in Surgical Oncology: ISSO*. 2009; 6:8.
6. Krainik A, Cyna-Gorse F, Bouccara D, Cazals-Hatem D, Vilgrain V, Denys A, Rey A, Sterkers O, Menu Y. MRI of unusual lesions in the internal auditory canal. *Neuroradiology*. 2001 Jan; 43(1):52-7.
7. Dana MT, Andrei IC, Victor VC, Claudia FC. Rare etiology of trigeminal nerve neuralgia- metastatic adenocarcinoma of the colon. *Romanian Journal of Oral Rehabilitation*. 2016 April-June; 8(2):40-43.
8. Wang A, Kleinman G, Murali R, Wainwright J, Tandon A. Metastatic Renal Cell Carcinoma Mimicking Trigeminal Schwannoma in a Patient Presenting with Trigeminal Neuralgia. *J Neurol Surg Rep*. 2015 Nov; 76(2):e282-6.
9. Soni CR, Kumar G, Sahota P, Miller DC, Litofsky NS. Metastases to Meckel's cave: Report of two cases and comparative analysis of malignant tumors with meningioma and schwannoma of Meckel's cave. *Clin Neurol Neurosurg*. 2010 Dec; 112(10):927-32.