



## Case Report

# Absence of contrast enhancement in a petroclival meningioma: Case report and systematic literature review

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## ABSTRACT

**Background:** Meningioma is one of the most common intracranial tumors with well-established radiologic features such as contrast enhancement, dural tail, and hyperostosis on computed tomography and magnetic resonance imaging. Contrast enhancement is usually homogeneous or heterogeneous based on tumor vascularity and underlying histopathology. Even in this context, faint or nonenhancing meningioma is exceedingly rare.

**Case Description:** A 57-year-old male presented with progressive right hearing loss, disequilibrium, occasional difficulty swallowing, and facial numbness. Imaging demonstrated an extensive hypodense, nonenhancing right cerebellopontine angle mass extending from the interpeduncular, and ambient cisterns to the foramen magnum. The pathological analysis demonstrated a microcystic meningioma WHO Grade I. There are few reported case reports or series of minimal or nonenhancing meningiomas, and a systematic review was performed for these cases. Seven peer-reviewed articles with 14 verifiable cases were identified and reviewed for radiologic features, tumor location, and tumor classification. The majority of minimal or nonenhancing meningiomas were microcystic, and most of them located at the convexity. This is the second case reported of a nonenhancing meningioma at the cerebellopontine angle and petroclival region

**Conclusion:** Meningioma should be considered a differential diagnosis for nonenhancing lesion at the cerebellopontine and petroclival regions.

**Keywords:** Cerebellopontine angle, Glial fibrillary astrocytic protein, Meningioma, Microcystic meningioma, Petroclival

## INTRODUCTION

Meningioma is one of the most common benign intracranial tumors. The use of neuroimaging has increased the diagnosis of meningioma, and there are classic radiologic features associated with the tumor, including contrast enhancement, dural tail, and hyperostosis of the adjacent bone. On computed tomography (CT), meningiomas are mostly isodense to hyperdense, and about 20% have calcifications.<sup>[3,8]</sup> The tumor has variable features on magnetic resonance imaging (MRI) from isointense to hypointense on T1-weighted sequence, and isointense to

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hyperintense on T2-weighted sequence.<sup>[19]</sup> Meningiomas enhance avidly on contrast-weighted images except for areas of necrosis or cyst.<sup>[8]</sup> We are presenting a rare case of a patient with a nonenhancing meningioma in the cranial base that had neither radiological feature of cystic changes nor necrosis. Nonenhancing meningiomas are extremely rare, and there are few reported cases in the literature. This case demonstrates that microcystic meningioma is included in the differential diagnosis in cases of nonenhancing tumors at the cerebellopontine angle. The implication of the enhancement on pathological features and clinical outcomes is not known. A systematic review was performed to analyze minimal or nonenhancing meningioma with regard to tumor location, histological type, and prognosis.

## CASE PRESENTATION

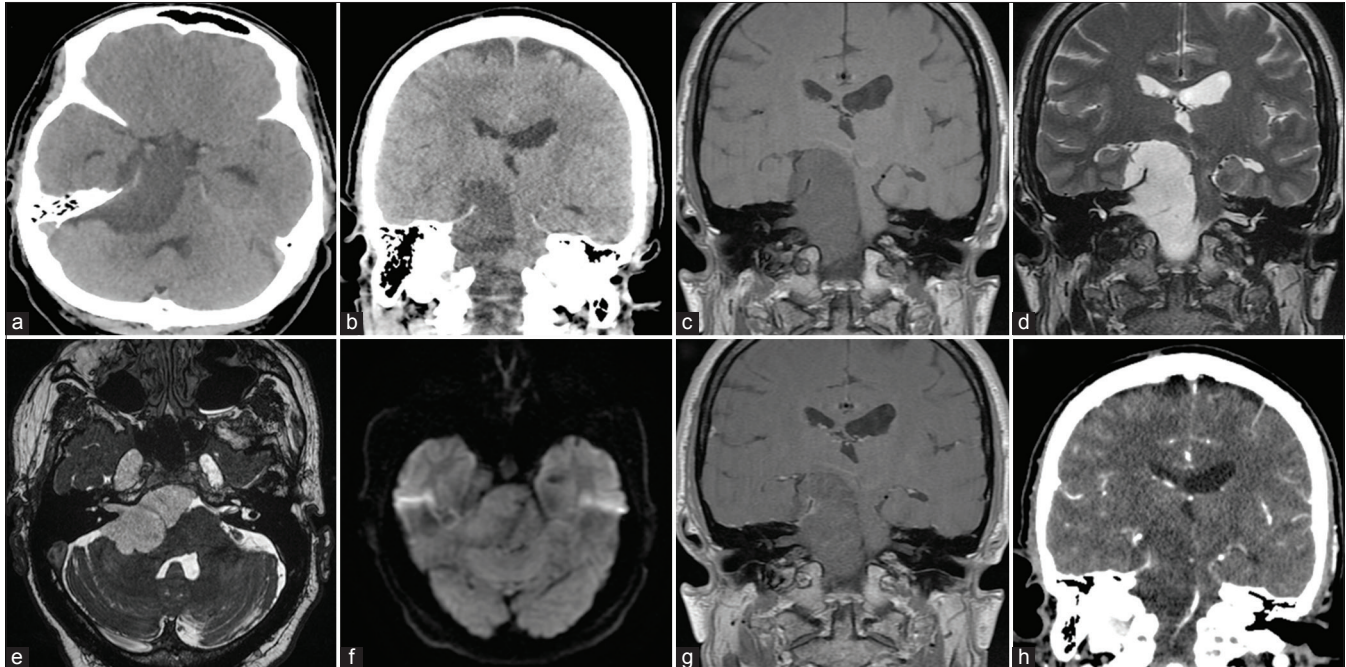
### History and physical examination

A 57-year-old right-handed male presented with progressive right hearing loss, disequilibrium, occasional difficulty swallowing, and right facial numbness. CT and MRI showed a right cerebellopontine angle tumor extending from the interpeduncular cistern to the foramen magnum. An

audiogram revealed moderate right sensorineural hearing loss. Neurological examination demonstrated right decreased hearing, right facial numbness, and a positive Romberg sign.

### Imaging

Noncontrast CT of the head revealed a hypodense right cerebellopontine angle mass extending from the interpeduncular and ambient cisterns to the foramen magnum [Figure 1a and b]. The tumor also infiltrated the right Meckel's cave. There was compression of the brainstem with effacement of the fourth ventricle but no hydrocephalus [Figure 1]. There was no abnormality in the adjacent bone to suggest hyperostosis. MRI showed a  $5.2 \times 3.8 \times 5.5$  cm (anteroposterior  $\times$  lateral  $\times$  craniocaudal) T1-weighted hypointense, and T2-weighted hyperintense mass [Figure 1c and d]. The three-dimension fast imaging employing steady-state acquisition (FIESTA) sequence revealed the tumor also extended into the right internal acoustic canal [Figure 1e]. The mass displayed intermediate diffusion signal when compared to adjacent CSF [Figure 1f]. Gadoterate meglumine contrast injection did not reveal an apparent enhancement. There was a faint enhancement around the tentorium but no significant enhancement within the tumor [Figure 1g]. Computed tomography angiography



**Figure 1:** Preoperative images. Axial (a) and coronal (b) noncontrast CT image showing the hypodense cerebellopontine mass extending in the supratentorial space, interpeduncular and ambiens cisterns, and the right Meckel's cave. (c and d) T1- and T2-weighted coronal MRI showing the extent of the tumor. (e) 3D-FIESTA sequence indicating the extension of the tumor into the internal acoustic canal. (f) Axial diffusion-weighted imaging demonstrating intermediate diffusion signal when compared with CSF. (g) Coronal view of T1-weighted with gadolinium demonstrating faint tentorial enhancement and no enhancement within the tumor. (h) Coronal view of a contrasted CT demonstrating the tentorial enhancement.

revealed displacement of the basilar artery and the posterior cerebral arteries [Figure 1h].

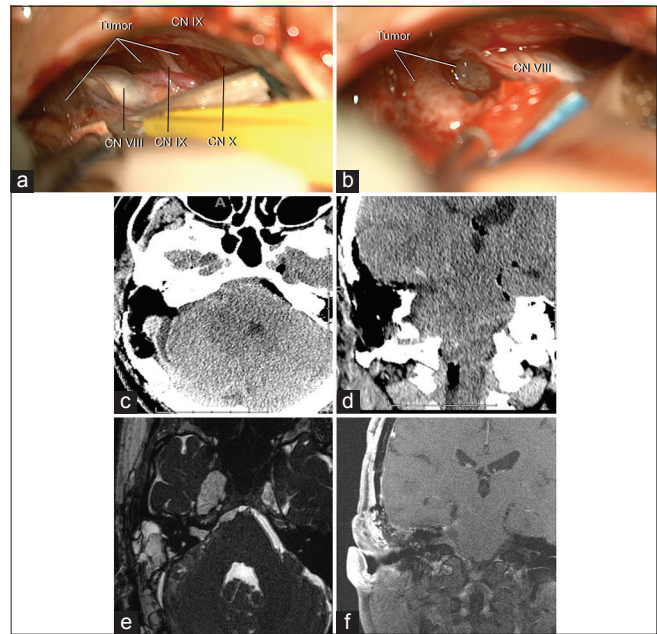
### Operative procedure

A posterior petrosal retrolabyrinthine approach with retrosigmoid and temporal craniotomies was used for the surgical approach in a staged operation (approach and resection in different days). After orotracheal intubation, the patient was placed in a left lateral decubitus position. Bone landmarks and neuronavigation were used for surgical incision planning. Facial nerve monitoring was placed. Incision and opening were made in a layered fashion, and the retrosigmoid and temporal craniotomies were performed as described in Graffeo *et al.* (2018). With the aid of a surgical microscope, the presigmoid retrolabyrinthine part of the approach was performed. There was no dural opening. The bone flap was reattached, and the incision was closed accordingly. The patient was extubated and was observed on the neurosurgical floor.

Two days later, he returned to the operating room for resection of the tumor and was positioned as described above. Neuromonitoring, including brainstem auditory evoked potential, was utilized for ipsilateral facial, glossopharyngeal, and accessory nerves. The incision and the bone flap were reopened. The retrosigmoid dura was opened in a curvilinear fashion, and the CSF was drained from the cisterna magna to relax the posterior fossa. The tumor was identified as a greyish mass with moderate vascularity and soft consistency, and resection was performed with an ultrasonic aspirator. In the posterior fossa, the vestibulocochlear, facial, and lower cranial nerves were identified posterior to the tumor, whereas the abducens nerve was found within the tumor. The trigeminal nerve was displaced superiorly against the tentorium. The infratentorial portion of the tumor was completely resected through the retrosigmoid part of the approach. The presigmoid and the temporal dura mater were opened, and the tentorium was divided after ligation of the superior petrosal sinus.<sup>[6]</sup> The trochlear nerve was identified entirely within the tumor and preserved, and the oculomotor nerve was displaced medially by the tumor. Tumor resection was completed except for a small portion in the Meckel's cave. The dura mater was closed primarily in the temporal and retrosigmoid areas, and the mastoid was reconstructed with fascia lata and fat graft. A lumbar drain was placed to minimize CSF leak postoperatively.

### Postoperative course

He had a right partial fourth, and sixth nerve palsies and mild facial weakness (House-Brackman 2) after surgery. A postoperative CT scan is shown in [Figure 2]. The lumbar drain was removed 3 days later, and he was discharged home. The facial weakness resolved during the hospital stay, and the



**Figure 2:** Intraoperative pictures and postoperative imaging after tumor resection. (a and b) intraoperative pictures demonstrating a grayish pink tumor of moderate vascularization anteromedial to cranial nerves VII, VIII, XI, and X. The cranial nerves IV and VI were embedded within the tumor and preserved. CN, cranial nerves. Axial (c) and coronal (d) non contrast postoperative CT images showing the posterior petrosal approach. Axial FIESTA (e) and coronal (f) T1-weighted with contrast postoperative MRI images showing the extent of tumor resection, a small portion of meningioma was left in Meckel's cave.

partial fourth and sixth nerve palsies improved within 6 weeks after the operation.

### Pathology

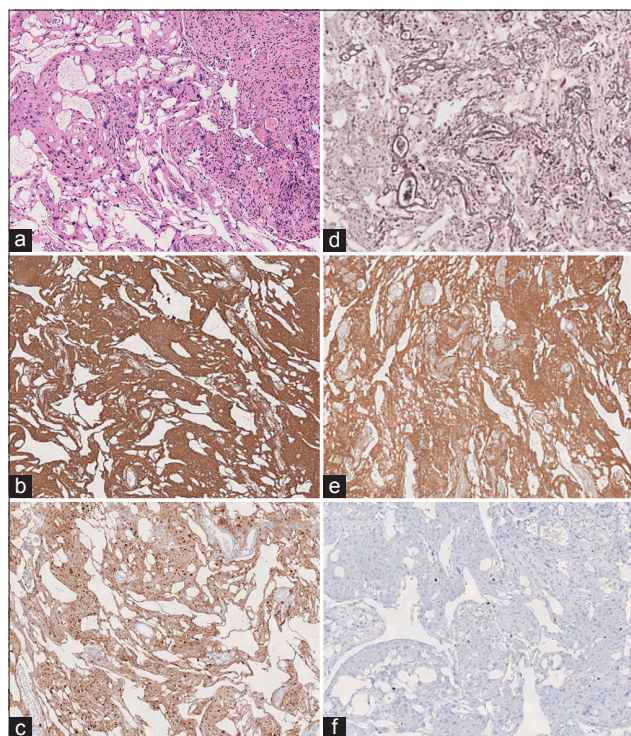
Grossly, the tumor appeared tan to pink. Microscopically, the tissue had lobulated architecture, mixed with prominent loose myxoid microcystic background and minor angiomatous changes. The lobulated component contained meningothelial whorls [Figure 3]. The cells showed an indistinct membrane with uniform nuclei, and the eosinophilic cytoplasm with no inflammatory cells infiltrate. There was no frank anaplasia or significant atypia. There was a clear arachnoid plane between the tumor and the brain parenchyma with no evidence of brain invasion. The tumor stained for Vimentin, glial fibrillary astrocytic protein (GFAP), and Cyclin D1, with low Ki-67 (<2%), and with an epithelioid reticulin pattern [Figure 3]. The tissue was diagnosed as microcystic meningioma, the WHO Grade 1.

### Systematic review

A systematic review of the literature for minimal or nonenhancing meningioma was conducted using the



Preferred Reporting Items for Systematic Reviews and Meta-analysis guidelines. Search items including “nonenhancing



**Figure 3:** (a) Hematoxylin and eosin ( $\times 4$ ) stained fragments of tissue with the proliferation of bland ovoid to epithelioid cells, with pink cytoplasm and indistinct cell borders. The whorled appearance is better appreciated. Nuclei appear to have some size variability, but no significant atypia or anaplasia is identified. The cystic pattern and angiomatous changes are also evident ( $\times 40$ ). (b) Vimentin: Immunohistochemical staining for Vimentin shows a strong, diffuse cytoplasmic staining pattern. (c) Cyclin-D1: Immunohistochemical staining for Cyclin-D1 shows a diffuse staining pattern. (d) Reticulin: Immunohistochemical staining for reticulin shows an epithelioid staining pattern. (e) GFAP: Immunohistochemical staining for GFAP shows a strong, diffuse cytoplasmic staining pattern. (f) KI-67: Immunohistochemical staining for GFAP shows scattered positive staining, corresponding to a low proliferation index estimated to be  $< 2\%$ .

meningioma,” “unenanced meningioma,” and more broadly “enhancing meningioma” were entered into the online databases PubMed and web of science. The search returned 652 total peer-reviewed articles, and each item was reviewed to evaluate for cases with minimal or nonenhancing meningioma. Screening guidelines included recoverable English-written articles with basic demographic and radiographic information, including images to support the interpretation of radiographic features. The last query was 04 April 2020. Selection and reporting biases are acknowledged. Cystic and necrotic meningioma was excluded from the review. Minimal enhancement was defined as faint or hardly noticeable contrast enhancement on radiologic images or interpretation. Seven articles, including case reports and case series, had 14 verifiable cases included in the systemic review [Table 1].<sup>[9-11,13,15,17,21]</sup> The average age was 48.1 years, and the majority were female (71.4%). Convexity meningioma was the common location at 57.1%. Two cases involved skull base, and one case involved the lumbosacral region. All cases had a faint or nonenhancing meningioma. Microcystic meningioma was the most common histological type at 85.7% (12/14). Clear cell and fibrous meningioma were other histological types of meningioma, respectively [Table 1]. Two patients had radiotherapy for adjuvant treatment.

## DISCUSSION

Meningioma is a common intradural tumor that avidly enhances with contrast. Almost all meningiomas either enhance homogeneously or heterogeneously. The meningioma presented in this report had faint enhancement with contrast, which is very rare. Dural tail, found in 72% of meningioma,<sup>[1]</sup> was not present, and hyperostosis, found in 50–60% meningioma,<sup>[14]</sup> was also not present. The constellation of these features made meningioma an unlikely diagnosis based on imaging alone. Given the typical location, near-absence of contrast enhancement, and hypointensity in T1 without contrast, the most likely primary diagnosis was an epidermoid cyst. The main confounding finding was

**Table 1:** A literature review of minimal or nonenhancing meningioma.

Articles	# Cases	Age	Gender	Location	CT	T1-weighted	T2-weighted	MRI enhancement	Histological type
Shimoji <i>et al.</i>	1	35	M	Convexity	Hyperdense	Hypointense	Hyperintense	Minimal	Microcystic
Lin <i>et al.</i>	7	51.9	F	4 convexities/1 skull base	N/A	Hypointense	Hyperintense	Minimal	Microcystic
Wang <i>et al.</i>	1	35	M	Lumbosacral	Hyperdense	Isointense	Isointense	No	Clear Cell
Seung <i>et al.</i>	1	59	M	Convexity	Hypodense	Hypointense	Hyperintense	Minimal	Microcystic
Paek <i>et al.</i>	1	37	M	Convexity	Hypodense	NR	NR	Minimal	Microcystic
Paek <i>et al.</i>	1	62	F	Convexity	NR	NR	NR	Minimal	Microcystic
Zhang	1	42	M	Convexity	Hyperdense	Hypointense	Hypointense	Minimal	Fibrous
Kubota	1	63	F	Skull Base	Hypodense	Hypointense	Hyperintense	Minimal	Microcystic
This study	1	57	M	Skull Base	Hypodense	Hypointense	Hyperintense	Minimal	Microcystic

the relative lack of diffusion restriction within the tumor [which is essentially pathognomonic for an epidermoid cyst, Figure 1]. A technical error in the administration of contrast could explain a lack of contrast enhancement in some cases. However, the fact that the nasopharyngeal mucosa, choroid plexus, and vessels enhance with contrast contradicts this hypothesis.

The histology and the immunostaining demonstrated that the tumor was a microcystic meningioma. The tumor stained for typical meningioma markers such as Vimentin, Reticulin, and Cyclin D1.<sup>[12]</sup> However, an important atypical molecular finding was the expression of the GFAP, which is mostly expressed in astrocytes.<sup>[5]</sup> The expression of GFAP was avid and diffusely expressed by the tumor cells. Meningioma cells do not typically express GFAP<sup>[4,12,19]</sup> except in the cases of brain invasion, which produces distinct meshwork of cells; one is meningothelial cells from the tumor, which are interwoven with the brain astrocytes expressing GFAP.<sup>[2]</sup> In this case, there was no evidence of brain invasion, and the GFAP was exclusively expressed by the tumor cells. The expression of GFAP by meningothelial cells in meningioma is rare<sup>[7,16,18]</sup> and the expression of GFAP by the meningothelial cells in this case suggests a possible precursor cell type distinct from precursor cells associated with the classic meningioma.

Meningioma is one of the most incidentally diagnosed intracranial lesions.<sup>[20]</sup> And yet, the literature review identified only 14 verifiable cases with minimal or nonenhancing type, which is rare.<sup>[9-11,13,15,17,21]</sup> Most of the tumors in the review had faint enhancement, and only one, a lumbosacral meningioma, was completely unenhanced. The majority of the meningiomas were located at the convexity with a small number of cases at the skull base and the spine. Microcystic meningioma comprised the majority of the minimally enhancing meningiomas, followed by fibrous and clear cell types. Apart from the contrast enhancement, there were other imaging sequences that distinguished these tumors from each other. The microcystic type was hypointense on T1-weighted and hyperintense on T2-weighted images like in our case. The fibrous type was hard with calcifications and was hypointense on both T1- and T2-weighted sequences. The clear cell meningioma was isointense on T1- and T2-weighted images.

Microcystic meningioma is rare WHO Grade 1 meningioma, and it constitutes about 1.6% of all intracranial meningiomas. Although the microcystic type comprised the majority of the minimally enhancing meningiomas, only 10.2% of microcystic meningiomas had a faint enhancement in MRI, and thus, the majority of them had typical radiological features of meningiomas.<sup>[10]</sup> These different radiological features do not seem to affect clinical management or prognosis.

This case demonstrates that microcystic meningioma should be considered part of the differential diagnosis for nonenhancing cerebellopontine mass with intermediate or no signal or diffusion-weighted imaging. The goal of treatment remains similar regardless of the tumor type; maximal safe surgical resection.

## CONCLUSION

Meningioma should be considered a differential diagnosis for a non-enhancing lesion at the cerebellopontine and petroclival regions.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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