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Case Report

Multiple meningiomas with varying MRI features and postsurgical outcomes: A case report ☆

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ABSTRACT

Meningiomas, which are typically benign tumors originating from the meninges, can present as multiple lesions in rare cases, occurring in 1%-10% of patients without neurofibromatosis. This report details a case involving a 49-year-old woman who initially presented with headaches followed by blurred vision, leading to the discovery of multiple meningiomas through MRI which appears as some solid mass outside the axial plane. The DWI ADC shows varying results. The patient then underwent transsphenoidal surgery for tumor resection. Histopathological analysis confirmed the presence of a meningothelial meningioma (WHO grade I) in the sellar region. Postsurgery, the patient had a CT scan showing a residual meningioma mass and experienced significant relief from her symptoms. The patient underwent outpatient treatment and was planned to undergo reoperation and follow-up MRI. The case underscores the complexity of managing multiple meningiomas. It highlights the necessity for a multidisciplinary approach to ensure accurate diagnosis and effective treatment strategies.

Introduction

Meningiomas are tumors that originate from the meninges, the protective membranes surrounding the brain or spinal cord. Harvey Cushing was the first to identify and characterize this type of tumor [1]. By definition, meningioma is the most common primary intracranial benign tumor that develops from arachnoid cells. The main predisposing factors for meningioma development are inactivation of the NF2 gene

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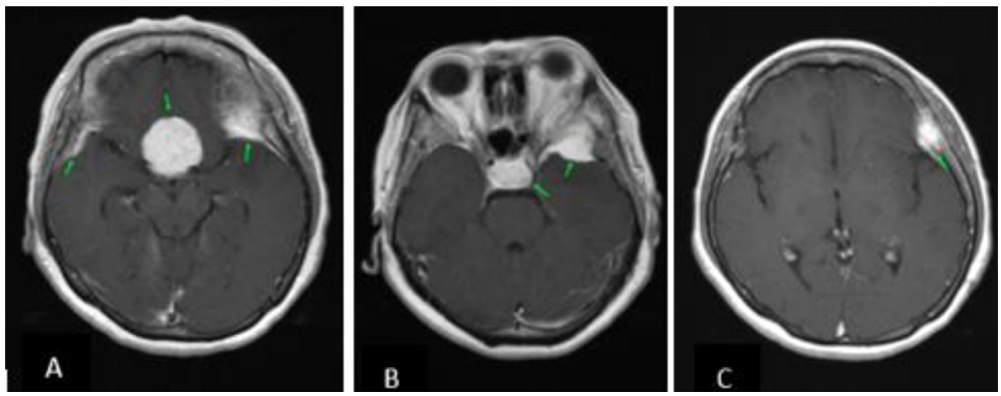

Fig. 1 – The axial T1 weighted scan showed a mass in the sellar region, 2 concavities in the temporal lobes (A and B), and a concavity in the left frontal lobe (C). It also showed a CSF cleft sign (+) and a dural tail sign (+). Hyperostosis is present in both the bilateral sphenoid wing and the left frontal bone.

and exposure to ionizing radiation, which can be the cause of some neoplasms [2]. Meningioma tumors are a relatively common finding in brain imaging with a prevalence of 53 per 100,000 people. The World Health Organization (WHO) classifies meningioma tumors based on their histological characteristics and recurrence risk as grade I, benign (80%), grade II, atypical (18%), and grade III, anaplastic/malignant (2%). The prevalence ratio of women and men affected by meningioma tumors is 3:1 [3].

Patients who have 2 or more meningioma tumors, synchronous or metachronous, that are spatially separated are called “multiple meningiomas." They defined multiple meningiomas as “at least 2 spatially separated meningiomas in a patient without signs of neurofibromatosis’’ [4]. Meningiomas can occur randomly or as part of neurofibromatosis type 2 syndrome or multiple familial meningiomas. Additionally, multiple meningiomas are observed in only a tiny subset, between 1% and 10%, of patients with meningiomas [5,6]. They also make up 5%-10% of all orbital tumors, and bilateral cases of optic nerve compression are sporadic, representing only 5% of all reported cases [7].

Diffusion-Weighted Imaging (DWI) is an MRI technique that measures the movement of water molecules in human tissues to estimate the cellular makeup of the tissue. DWI offers the capacity to quantify water diffusion across tissues through the measurement of ADC values. Another highlight of DWI is that ADC value can differentiate between benign and malignant tumors. Malignant tumors generally have lower mean ADC values due to their high cellularity, high nuclear-to-cytoplasmic ratio, and densely packed intracellular space [8].

Case report

A 49-year-old female patient came with a history of chronic headaches that were felt to be intermittent for approximately four years. The patient experienced a stabbing sensation throughout her head, accompanied by a throbbing sensation on her forehead, with a Numeric Rating Scale (NRS) of 4-5. The pain intensified when she bent down or exerted strain. Head pain is relieved by sleeping and taking painkillers, then followed by symptoms of blurred left eye vision 3 years ago and followed by blurred eye vision 2 years ago. The patient did not complain of limb weakness, loss of balance, or changes in facial sensation. The patient has no stigma of neurofibromatosis. The patient has a history of using hormonal injectable contraceptives for approximately 15 years and has stopped since 3 years ago.

The physical examination revealed compos mentis consciousness, a blood pressure of 120/80 mmHg, a pulse rate of 82 beats per minute, a respiration rate of 18 beats per minute, and a temperature of 36.6°. The neurological exam shows that both eyes can’t see in dim light a so it can be concluded that the visual field test shows binocular visual field loss. The funduscopy test reveals papillary atrophy in both eyes, while other cranial nerve exams, motor exams, sensory exams, physiological reflexes, and pathological reflexes all fall within normal limits. Proptosis left eye. The patient had an MRI of the brain with gadolinium contrast, which showed several solid masses outside of the axial planes. The images showed CSF cleft signs which is the cerebrospinal fluid that surrounds the mass and dural tail signs in the bilateral temporal lobe concavities and the left frontal lobe concavity. These masses were accompanied by hyperostosis in the sphenoid wing bilaterally and the left frontal bone in the intrasellar. The mass extends supracellularly (Fig. 1), compressing the optic nerve, optic chiasm, and optic tract bilaterally (Fig. 2), causing visual impairment. The DWI ADC examination yielded varying results. Fig. 3A shows that the solid mass in the sellar area had limited diffusion. On the other hand, Fig. 3B shows that the solid mass in the bilateral temporalis lobe concavity and the left frontal lobe did not do so. The MRI spectroscopy examination showed an increase in choline, glutamine, and a decrease in creatine (Fig. 4).

After a contrast-enhanced MRI of the head, the patient had surgery to remove the sellar tumor. The surgery left behind bits of brownish-white, rubbery tissue that weighed 2.4 grams (Fig. 5). Histopathology was then used to look at the tissue. It showed a tumor mass made up of medium-sized oval to syn-

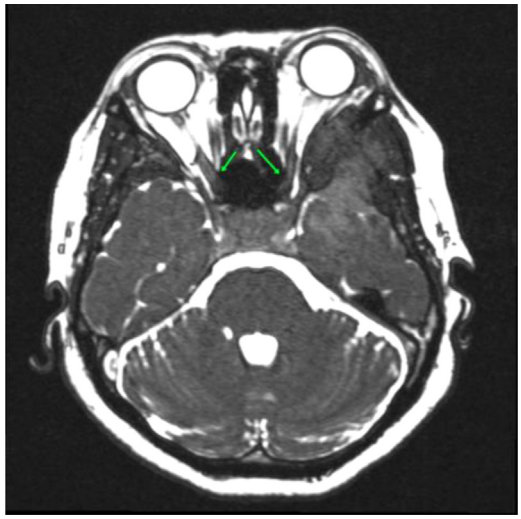

Fig. 2 – Illustrates how the mass affects the optic nerve, optic chiasm, and optic tract bilaterally.

cytial cells that become hyperplastic, clumped, compacted, and arranged in whorls. Cell nuclei are oval and monomorphic; some give a picture of pseudoinclusion, but mitosis is not found. The cytoplasm is abundant. Fibrocollagenous connective tissue stroma with dilated blood vessels and hemorrhage was also seen. There were no signs of malignancy, such as marked vascularity, loss of organoid structure, mitotic figures, nuclear pleomorphism, prominent nucleoli, focal necrosis, or infiltration to the adjacent brain, leading to the conclusion that the condition was meningothelial meningioma (WHO grade I) in the sellar region (Fig. 6).

Three days postsurgery, the patient underwent another CT scan of the brain with contrast for evaluation. There was a residual meningioma mass in the suprasellar region of the scan. There were also extra-axial solid lesions in the left frontal lobe, the concavity of both temporalis lobes, and the suprasellar region (Fig. 7A). Additionally, there was hyperostosis in the bilateral sphenoid wings and left frontal bone (Fig. 7B). The patient was treated for 9 days after transsphenoidal surgery, and the complaints of headache were significantly reduced, but the unchanged visual acuity in both eyes is probably due to irreversible damage caused by the compression of the bilateral optic nerve. Subsequently, the patient underwent outpatient treatment, followed by planning for surgical treatment to remove the mass in the concavity of the temporal lobe and was scheduled for a follow-up MRI 6 months later.

Discussion

Meningiomas are tumors that originate from the meninges, the protective membranes surrounding the brain or spinal cord. Meningiomas can occur randomly or as part of neurofibromatosis type 2 syndrome or multiple familial meningiomas. Additionally, multiple meningiomas are observed in only a tiny subset, between 1% and 10%, of patients with meningiomas [6]. In the case of multiple meningiomas, it is crucial to understand their potential impact on the patient's health, including common symptoms such as chronic headaches and blurred vision. Magnetic resonance imaging is the primary diagnostic tool for identifying meningiomas, providing detailed information about their location, size, and appearance [7]. A person with multiple meningiomas may require a combination of surgery, radiation therapy, and occasionally systemic therapies or participation in clinical trials. The exact treatment plan will depend on the patient's neurological symptoms, such as the number, size, and location of the tumors, as well as the presence and severity of headaches, vision changes, and other focal deficits [9].

While the underlying mechanisms behind the development of multiple meningiomas are not fully understood, some research has shed light on potential contributing factors. Genetic predisposition, exposure to ionizing radiation, and hormonal factors have all been implicated in the

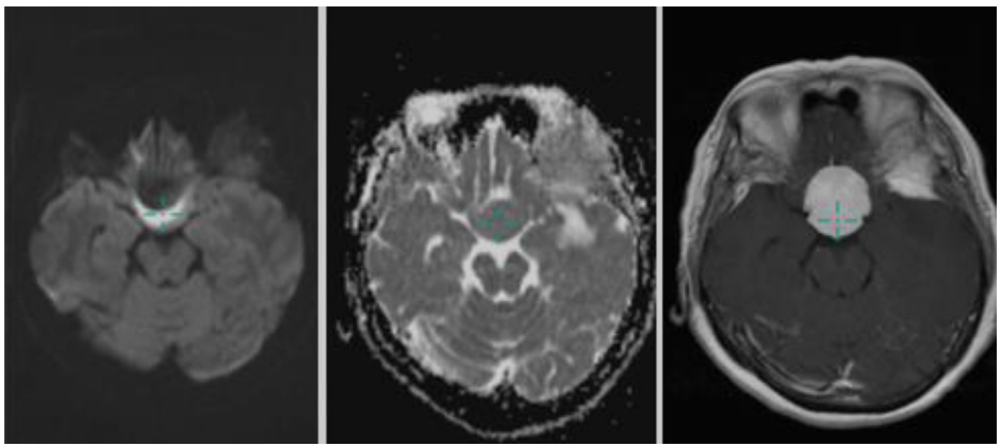

Fig. 3 – (A) The DWI ADC examination conducted in the solid mass sellar region revealed restricted diffusion. (B) The DWI ADC examination revealed no restricted diffusion in the solid mass left temporal lobe concavity region (A) and left frontal lobe concavity region (B).

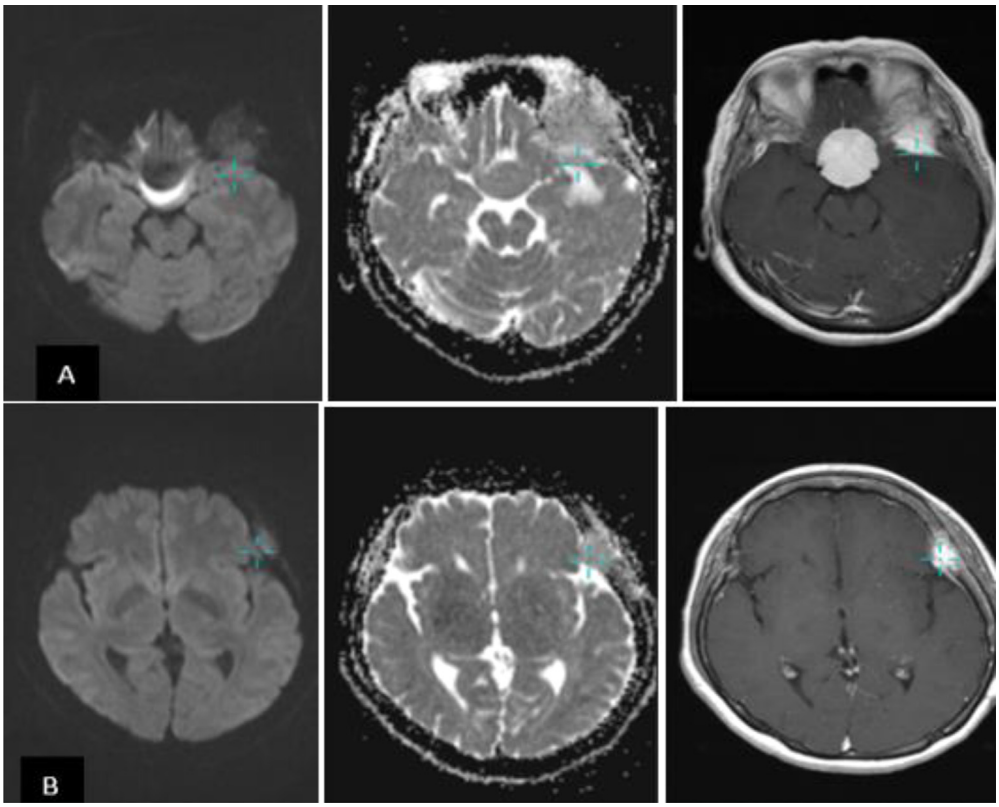

Fig. 3 – Continued

pathogenesis of meningiomas [9]. In this case, a 49-year-old woman, who had been using hormonal birth control for approximately 15 years, presented with complaints of chronic headaches and blurred eyes. During the exam, an MRI of the brain showed several solid masses outside of the skull, along with hyperostosis. A histopathologic examination confirmed the diagnosis of a meningothelial meningioma (WHO grade I).

Computed tomography imaging helps detect calcification and hyperostosis in meningiomas, but magnetic resonance imaging is more effective for evaluating tissue characteristics and associated edema. On MRI, meningiomas typically appear hypointense to isointense on T1-weighted sequences and isointense to hyperintense on T2-weighted sequences. Additionally, meningioma contents exhibit no diffusion restriction, demonstrate a signal comparable to cerebrospinal fluid, and are often not accompanied by surrounding edema. A well-defined border, homogeneous contrast enhancement, calcification, and a dural tail frequently characterize benign meningiomas. While a dural tail is not a pathognomonic feature of meningiomas, it is a common characteristic observed in approximately 72% of cases. [10] Meningiomas, which account for only 5% of all reported cases, are relatively uncommon. They also make up 5%-10% of all orbital tumors, and bilateral cases of optic nerve compression, like the one observed in this patient, are sporadic, representing only 5% of all reported cases [11]. In our case, the unchanged visual acuity in both eyes is probably due to irreversible damage caused by the compression of the bilateral optic nerve.

ADC values had any relationship with prognostic factors, especially tumour grade and lymphovascular invasion. Tumour characteristics are crucial in the management of tumours and DWI is a very sensitive method in determining tissue structure; this structure may accurately represent the aggressiveness of the tumour, thus important data may be derived from DWI [8]. In this case, there was a difference in the DWI ADC examination, wherein the solid mass in the sellar area and diffusion restriction were found. However, diffusion restriction was not found in the solid mass in the bilateral temporalis lobe concavity and the left frontal lobe concavity. The increased ADC value of these meningiomata may be explained by the presence of increased amounts of fluid within these lesions. The presence of the increased fluid may allow for free movement of water molecules or less restriction to water diffusion, much in the same way that CSF diffuses freely without restriction and appears hypo intense on the diffusion-weighted MR sequences [12].

There are 2 distinct hypotheses for multiple meningiomas. The first suggests that the tumors arise independently from each other. This theory is backed by histological and cytogenetic examinations, which revealed microscopic and karyotypic differences in multiple tumors in the same patient. The second hypothesis suggests that a single triggering event causes the original clone of cells to spread throughout the meninges, resulting in multiple and clonally-related tumors [13].

Banga et al., reported a case of a 50-year-old female presented with complaints of headache over the previous year

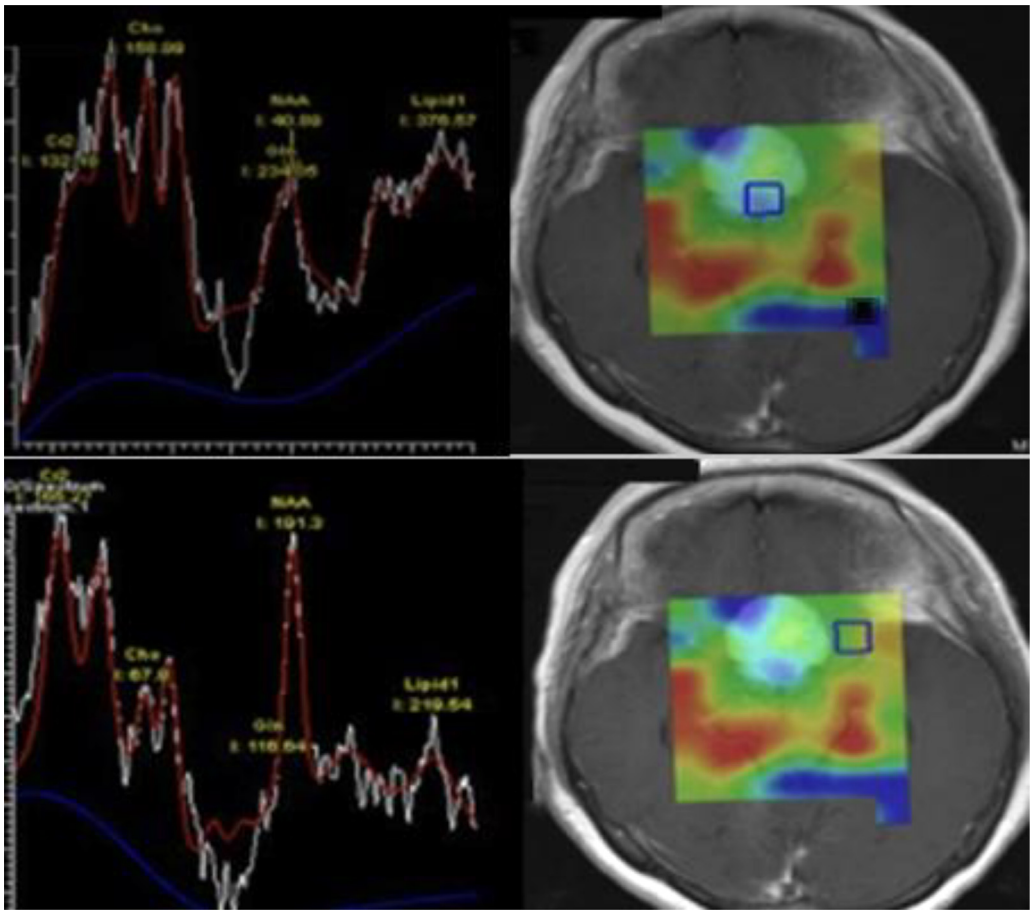

Fig. 4 – The MRI spectroscopy examination revealed an increase in choline, glutamine, and a decrease in creatine.

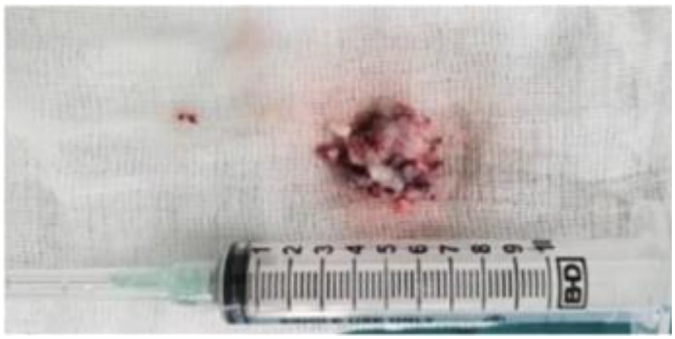

Fig. 5 – The tumor mass was removed after undergoing transsphenoidal surgery.

and right-sided weakness for 1 month. Clinical examination revealed right-sided hemiparesis. MRI of the brain demonstrated a well-defined, homogenously enhancing extra-axial mass located in the left frontoparietal convexity, and an intradiploic mass in the left parietal bone abutting the parietal lobe. She underwent a large left frontoparietal craniotomy with excision of both the masses (Simpson's Grade 1) with mesh cranioplasty. Histopathologic section of the frontoparietal mass showed, predominantly, a meningothelial pattern, and the intradiploic lesion showed sheets of fibroblastic cells separated by collagen bundles consistent with a fibroblastic variety. Both tumors were of WHO grade I [13].

In 2001, Koh et al., published a report of a patient with multiple meningiomas having both benign and malignant pathology. Pathological diagnosis showed that the patient had a psammomatous meningioma in the left parietal region and an atypical meningioma in the right frontal falx region. The authors claimed it to be the first

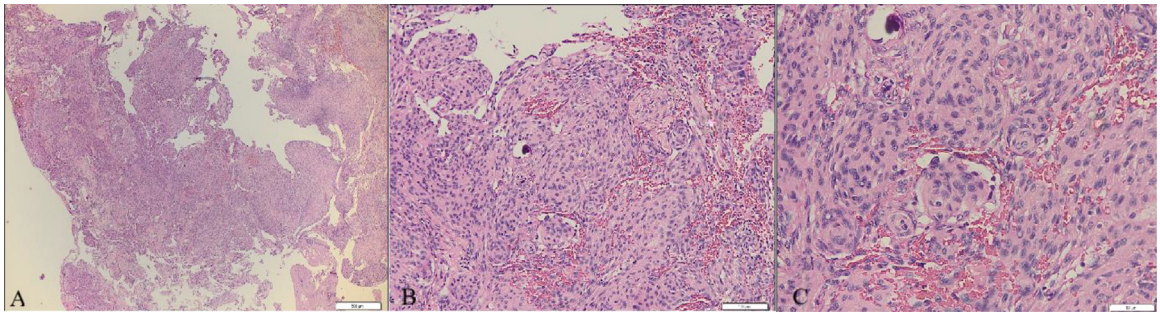

Fig. 6 – The histopathologic examination revealed meningothelial meningioma (WHO grade I) at magnifications of (A) 20x, (B) 100x, and (C) 200x.

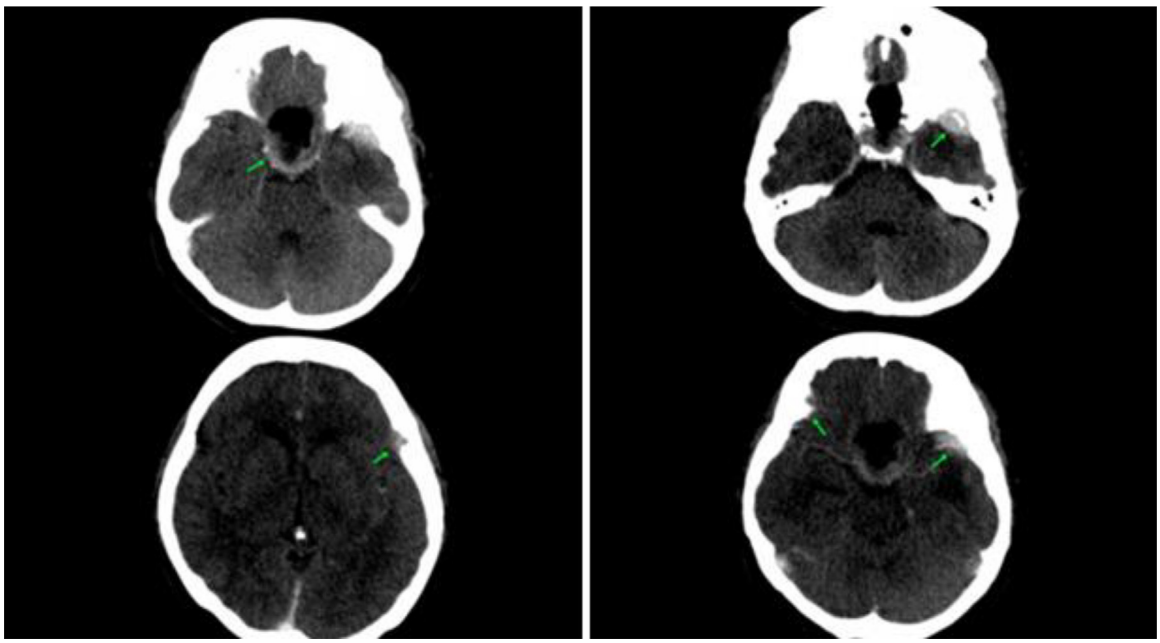

Fig. 7 – (A) We conducted an axial brain CT scan examination using contrast after undergoing transsphenoidal surgery. A leftover mass was discovered in the sellar area, and several solid lesions outside of the axes were found in both the left frontal and temporal lobes, showing a lot of uniform enhancement. (B) A bone window head CT scan revealed defects and hyperostosis in the left frontal bone and bilateral sphenoid wing.

case reported where 2 tumors existed concurrently, with each having different histology (benign versus malignant) [13].

According to Sanverdi et al., grade II and III tumor meningiomas may show more significant than expected restricted diffusion. In their study, the mean ADC values for benign meningiomas were higher than those for atypical and malignant meningiomas. However, the difference was not significant ($P > .2$) in distinguishing between benign and atypical and/or malignant meningiomas [14]. DWI and ADC measurements, even when performed under the best conditions, do not seem reliable in grading meningiomas or identifying histological sub-types. Therefore, these parameters should not be suggested for surgical or treatment planning [15].

The treatment and prognosis of multiple meningiomas do not differ from those of solitary benign tumours. Surgery is the treatment of choice for multiple meningiomas and depends on the following characteristics: symptomatic meningioma, asymptomatic meningioma greater than 3 cm in size and surgically accessible, and symptomatic expanding tumor. Each tumour should be approached individually, and the mere presence of multiple tumours does not justify their removal. Beyond this, the postoperative course from the initial surgery may be tumultuous and characterized by complications, malignant transformations, the requirement for supplementary therapy to address resistant tumors, and additional surgeries to address the sporadic tumors as they arise [16].

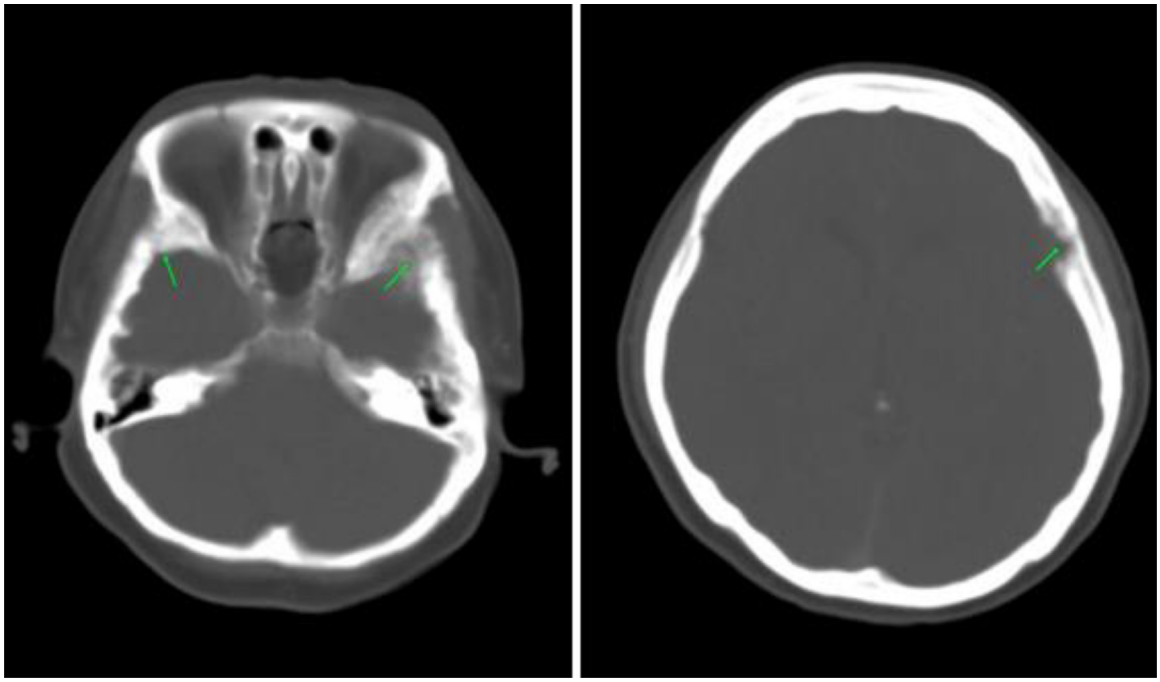

Fig. 7 – Continued

Conclusion

Multiple meningiomas constitute a relatively uncommon and complex condition characterized by numerous meningioma tumors within the central nervous system. This study emphasizes the significance of a multidisciplinary approach for accurate diagnosis and effective treatment planning, encompassing advanced imaging techniques and therapeutic strategies. Continuing to investigate and comprehend the clinical variability and prognostic factors influencing the disease course aims to enhance patients' quality of life and treatment outcomes. Further research and development of more innovative treatment modalities will be instrumental in addressing the challenges associated with multiple meningiomas.

Patient consent

Written informed consent for publication of their case was obtained from our patient.

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