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Serous Otitis Media Revealing Temporal *En Plaque* Meningioma

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Objectives: To present a series of temporal *en plaque* meningiomas involving the middle ear or mastoid, whose main symptoms suggested a serous otitis media.
Study Design and Settings: Multicentric retrospective study reviewing clinical records originating from eight tertiary referral centers.
Materials and Methods: The clinical records of 10 patients presenting with signs and symptoms suggesting serous otitis media and whose neuroimaging studies revealed a temporal *en plaque* meningioma involving the middle ear or mastoid are reported.
Results: All the patients were women, ranging from 49 to 71 years old. The delay between the onset of symptoms and the diagnosis of meningioma varied from 1 to 10 years. All the patients underwent various procedures usually applied for the treatment of serous otitis media, which failed in all the cases, particularly ventilating tube placement, which was followed by severe episodes of discharge. In all cases, the computed tomographic scans showed three imaging signs: soft tissue mass filling the middle ear or mastoid, hyperostosis of the petrous bone, and hairy aspect of the intracranial margins of the affected bone. This imaging triad must alert the otologist of the possibility of intracranial meningioma. Magnetic resonance imaging was the method of choice to assess the diagnosis of intracranial meningioma involving the middle ear or mastoid. When analyzing management options, it appeared that conventional middle ear procedures were inefficient.
Conclusion: Temporal *en plaque* meningioma involving the middle ear or mastoid can mimic a serous otitis media. A computed tomographic scan is recommended for cases of atypical or prolonged unilateral serous otitis media to investigate indirect signs of a meningioma, which has to be confirmed with magnetic resonance imaging. **Key Words:** Meningioma—Otitis media—Temporal bone.
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Meningiomas are slow-growing benign tumors originating from arachnoid granulations. They account for 13 to 26% of all primary intracranial tumors (1). Extracranial meningiomas are most often located in the orbit, and then the external calvaria, nasal and paranasal cavities, and neck and tympanomastoid cavities (2).

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The most common otologic and neurotologic symptoms of a meningioma of the temporal bone are sensorineural hearing loss, tinnitus, dizziness, vertigo, or facial palsy caused by the compression of the VIIIth or VIIth cranial nerves. Meningiomas extending to the mastoid bowl or to the middle ear cleft are uncommon, with less than 100 cases reported in association with the international literature, and can present as chronic otitis media (2,3). Most of these cases were secondary extracranial meningiomas originating from intracranial meningiomas extending to the temporal bone through various pathways (3). Primary temporal bone meningiomas are extremely rare, and care must be taken not to misdiagnose an

intracranial primary meningioma (3,4). Computed tomographic (CT) scan in conjunction with magnetic resonance imaging (MRI) can confidently determine the diagnosis and extent of a temporal bone meningioma (1). The gross appearance of central meningioma can be a globular form, presenting as a bulky mass, or an *en plaque* form that tends to infiltrate along meninges and nerve sheaths (1,2).

We report 10 cases of patients presenting with the appearance of a serous otitis media (SOM) resistant to conventional therapy and whose underlying diagnosis of *en plaque* temporal meningioma was disclosed after imaging evaluation.

MATERIALS AND METHODS

The clinical records originating from eight tertiary referral centers were retrospectively reviewed. The criteria for inclusion were patients presenting with signs and symptoms suggesting SOM and whose neuroimaging studies revealed a temporal *en plaque* meningioma involving the middle ear or mastoid. Ten cases meeting the criteria for inclusion were collected, reported below, and summarized in Table 1.

Case 1

A previously healthy 53-year-old woman was referred to one of the authors (D.A.) for a left SOM resistant to conventional therapy. SOM was diagnosed 4 years ago with the following sign and symptoms: aural fullness, conductive hearing loss, and Type B tympanogram. Aural fullness and hearing loss were not modified despite three courses of antibiotics and steroids. Ventilating tube (VT) insertion did not improve hearing and was followed by purulent discharge. Otorrhea did not respond to medical therapy (local and oral antibiotics) and ceased only when the tube was removed. At initial examination, the patient complained of aural fullness and hearing loss. On otomicroscopic examination, the ear was not discharging and fluid filled the middle ear behind an intact tympanic membrane. Audiometry demonstrated a 40-dB left conductive hearing loss. Computed tomography (CT) demonstrated a nonspecific opacity of the left middle ear and mastoid bowl, with an hyperostotic reaction along the temporal bone and greater wing of the sphenoid, and a hairy aspect of the margins of the affected bone. MRI showed an enhancement and thickening of the temporal and sphenoidal dura associated with an enhancing soft tissue mass of the middle ear, corresponding to a temporosphenoidal *en plaque* meningioma invading the middle ear. Regarding the extent of the dural lesion on MRI and because of the absence of brainstem compression and cranial nerve involvement, it was decided to follow the patient without surgical intervention.

Case 2

A 54-year-old woman was evaluated by one of the authors (D.A.) for a 6-year history of right SOM, persisting despite several courses of antibiotics and steroids and two VT insertions. Hearing was not modified neither with medical therapy nor with VT. VT insertion was complicated by purulent discharge. The patient complained of hearing loss and aural fullness. Otoscopic examination revealed a very narrow right external auditory canal; the tympanic membrane was intact and the middle ear was filled with fluid. Audiometry showed a right moderate mixed hearing loss and Type B tympanometry. CT showed an opacity of the right mastoid and middle ear cavities with an hyperostotic reaction of the temporal bone with a hairy aspect of the margins of the bone without bony erosion. MRI demonstrated an enhancing intracranial *en plaque* meningioma invading the temporal bone and extending within the mastoid and middle ear cavities. The decision of conservative management without surgery was taken regarding the extent of the meningioma.

TABLE 1. *Serous otitis media revealing a temporal en plaque meningioma patient data*

Case	Sex/Age	Symptoms/Signs	Delay before diagnosis	Treatment before diagnosis	Diagnostic	Treatment option
1	F/53	AF, HL	4 yr	VT ⇒ Discharge	CT + MRI	Clinical + Imaging follow-up
2	F/54	AF, HL	6 yr	VT ⇒ Discharge	CT + MRI	Clinical + Imaging follow-up
3	F/67	Pain, tinnitus, vertigo	2 yr	Tympanoplasty	Biopsy + CT	Clinical + Imaging follow-up
4	F/55	Headache, AF, HL	2 yr	VT ⇒ Discharge	CT + MRI	Clinical + Imaging follow-up (4 yr)
5	F/70	Discharge, HL	8 yr	Mastoidectomy Tympanoplasty VT ⇒ Discharge	CT + MRI	Clinical + Imaging follow-up
6	F/49	HL, positional vertigo, mild FP, trigeminal neuralgia	2 yr	VT ⇒ Discharge	CT + MRI	Surgery (combined petrosal approach)
7	F/53	Pain, HL	2 yr	Mastoidectomy VT ⇒ Discharge	CT + MRI	Clinical + Imaging follow-up
8	F/60	AF, Tinnitus	10 yr		Scintigraphy (breast carcinoma) CT + MRI + Biopsy	Clinical + Imaging follow-up
9	F/71	AF, HL, visual loss		VT ⇒ Discharge	MRI	Radiotherapy
10	F/58	HL	1 yr	Mastoidectomy + Biopsy	CT + MRI + Biopsy	Clinical + Imaging follow-up

AF, aural fullness; HL, hearing loss; FP, facial palsy; VT, ventilating tube; ME, middle ear; MRI, magnetic resonance imaging; CT, computed tomography.

Case 3

A 67-year-old woman was referred to one of the authors (V.D.) after having undergone two previous surgical procedures for a right SOM that started 2 years ago. She experienced pain, tinnitus, and vertigo. The first diagnosis was SOM with retraction pocket. The first surgical procedure using a transcanal approach found both "inflammatory mucosa" and serous fluid within the middle ear. No biopsy was done. There was no improvement, and a VT was inserted for a second time a few months later. Pain and vertigo remained and a second operation was proposed. Revision surgery using a postauricular approach was performed 1 year later. The surgeon faced an important bleeding coming from the middle ear; he performed a mastoidectomy and found the same "inflammatory" tissue that he sent to the pathologist. The diagnosis was meningioma of meningoepithelial type. At otoscopy, the tympanic membrane was intact, but it hid a reddish mass filling the middle ear. At audiometry, there was a 45-dB conductive deafness. CT revealed an hyperostotic bone infiltrating the whole petrous bone. It was decided to simply watch and scan.

Case 4

A 55-year-old woman was referred to one of the authors (F.T.) for a 1-year history of unilateral SOM in the left ear. She complained of aural fullness, hearing loss, and headaches. The audiogram revealed a moderate conductive hearing loss on the left side (pure-tone average, 0.5–3 kHz; 40 dB, Type B tympanogram). Imaging evaluation, with CT and MRI, showed *en plaque* meningioma of the middle fossa floor on the left side. CT scan presentation was typical with the association of an opacity of the tympanomastoid cavities and a hyperostotic reaction of the petrous bone. A long-term VT was inserted in the left tympanic membrane 2 years after the onset of symptoms, with a moderate improvement of hearing loss to a 23-dB pure-tone average (0.5 to 3 kHz). Four years after the VT insertion, hearing loss had been stabilized, but the patient experienced recurrent episodes of purulent discharge. It was decided to watch and scan. MRI is performed every year, with the latest (5 yr after the first one) showing a slight progression of the *en plaque* meningioma.

Case 5

A 70-year-old woman was referred to one of the authors (F.T.) in 1999, with a long-lasting history of right chronic otitis media. An SOM was diagnosed in the right ear in 1991, after which she underwent a right mastoidectomy with an exploration of the middle ear in another institution in 1993. In 1999, she presented with a discharging subtotal perforation of the right tympanic membrane, and the audiogram showed a 45-dB right conductive hearing loss There were no other otoneurological signs or symptoms. Imaging evaluation with CT and MRI led to the diagnosis of *en plaque* meningioma of the right temporal bone. CT reported signs of right mastoidectomy, opacity of the tympanomastoid cavities, and hyperostotic reaction of the middle fossa floor. It was decided to perform a right myringoplasty to avoid chronic discharge. This procedure was successful in closing the tympanic membrane and stopping discharge, but hearing did not improve because of the recurrence of effusion. Six months later, a VT was inserted in the right tympanic membrane improving hearing to a 29-dB conductive hearing loss but resulting in recurrent episodes of discharge. On the last MRI in 2004, a slight progression of the right *en plaque* meningioma occupying the middle fossa floor and extending to the right cavernous sinus was observed (with the patient still being symptom-free).

Case 6

A 49-year-old woman was evaluated by two of the authors (B.G., J.P.L.) for a 2-year history of left SOM, persisting despite medical therapy and insertion of a VT. She complained of left hearing loss, recurrent episodes of discharge since VT had been inserted, and positional vertigo. During several months, she experienced headaches and trigeminal neuralgia. Clinical examination revealed a mild facial palsy. Otomicroscopic examination showed a mass bulging behind the pars flaccida and extrusion of the VT on the left side. CT showed an opacity of the middle ear and a hyperostosis of the petrous bone. MRI revealed a left *en plaque* meningioma of the temporal bone involving the middle ear and the cavernous sinus. Surgery was decided because of a disabling trigeminal neuralgia. A subtotal resection of the tumor was recently performed

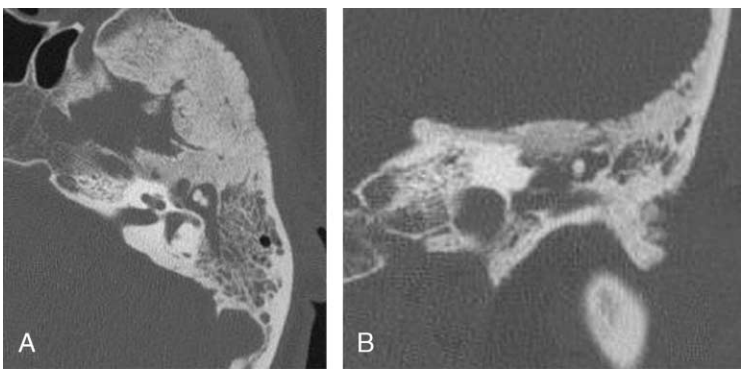

FIG. 1. Axial (*A*) and coronal (*B*) CT scans of the left temporal bone (Case 1) showing a complete opacity of the tympanomastoid cavities associated to an hyperostosis involving the temporal bone and the greater wing of the sphenoid. Notice the typical hairy aspect of the margins of the affected bone.

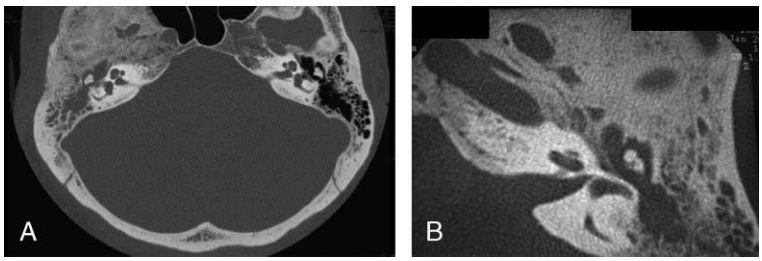

FIG. 2. Axial CT scans showing the typical aspects of a right temporal *en plaque* meningioma in Case 2 (*A*) and a left temporal *en plaque* meningioma in Case 6 (*B*).

through a combined petrosal approach. The initial outcome is encouraging—the disappearance of trigeminal neuralgia without facial or hearing complications.

Case 7

A 53-year-old woman was evaluated by two of the authors (B.G., S.S.) for a 2-year history of left SOM. During this period, she underwent several courses of antibiotics and steroids, a left mastoidectomy, and a VT insertion, without improvement of symptoms. She complained of disabling left otalgia, left otorrhea since VT had been inserted, and left hearing loss (mixed hearing loss on audiometric examination). CT showed a postoperative aspect of left mastoidectomy with opacity of the middle ear and mastoid cavity and hyperostosis of the petrous bone, suggesting a left *en plaque* meningioma that was demonstrated with MRI. Interestingly, we retrospectively noted that typical CT aspect of temporal *en plaque* meningioma was already present at the beginning of the disease, but diagnosis was not considered at that time. Otoneurosurgical decompression or radiation therapy was discussed with the patient because of the disabling otalgia, but she preferred to wait and scan.

Case 8

A 60-year-old woman was referred to one of the authors (P.B.) with a 10-year history of left aural fullness and tinnitus. Otoscopic examination suggested a left SOM, with a mixed hearing loss on audiometric test. Concurrently, she developed a breast carcinoma. Scintigraphy performed because of the carcinoma disclosed a high-uptake lesion of the left temporal bone. Neuroimaging, with CT and MRI, suggested a left temporal *en plaque* meningioma. A surgical exploration of the middle ear and mastoid was performed to obtain a biopsy to eliminate a metastasis. Pathologic findings revealed a meningioma. It was decided to wait and scan. The meningioma did not progress with a 3-year follow-up.

Case 9

A 71-year-old woman presenting with a left visual loss was referred to one of the authors (P.B.) because it was associated with a left aural fullness. Otologic examination revealed a left SOM. MRI showed a left *en plaque* meningioma involving the posterior surface of the petrous bone, the clivus, and the greater wing of the sphenoid. She received radiation therapy because of the progression of the visual loss, and a VT was inserted. The postoperative course was marked by recurrent episodes of discharge without improvement of the aural fullness.

Case 10

A 58-year-old woman was referred to one of the authors (E.L.) with a 1-year history of right SOM. Audiometry showed a right moderate mixed hearing loss. CT showed an opacity of the middle ear cleft extending to the mastoid and a significant hyperostosis and sclerosis of the tegmen. She underwent a surgical exploration at another institution. The surgical report described difficulties during the mastoidectomy because of uncommon granuloma-like bleeding lesions. A biopsy was undertaken and detected a meningioma. We retrospectively confirmed the CT aspect of *en plaque* meningioma. MRI revealed a well-enhanced

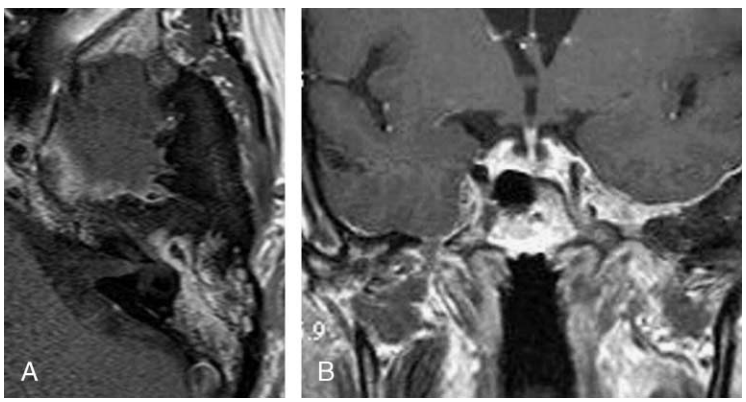

FIG. 3. Axial (*A*) and coronal (*B*) T1-weighted MR images after contrast administration demonstrating meningeal thickening and enhancement along the intracranial aspect of abnormal bone extending along the left petrous bone and greater wing of the sphenoid (Case 1). Thickening is particularly conspicuous on the left side of coronal image performed at the level of foramen ovale. MR images also show a strong enhancement of meningiomatous tissue within the tympanomastoid cavities.

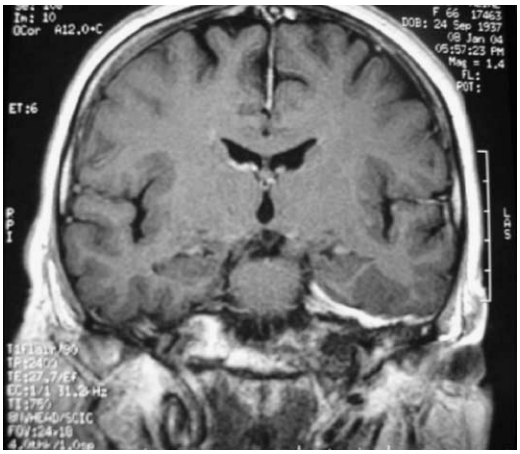

FIG. 4. Coronal T1-weighted MR image after gadolinium injection showing the typical aspect of a left temporal *en plaque* meningioma (Case 3).

mass lesion, which was consistent with CT. This lesion extended along the middle cranial fossa to the geniculate fossa and to the greater wing of the sphenoid. MRI also revealed a second meningioma independent from the right *en plaque* temporal meningioma: It was a symptom-free meningioma of the left orbital roof. Regarding the extent of the right lesion and the asymptomatic feature of the left one, it was decided to wait and scan.

RESULTS

All the patients were women, ranging from 49 to 71 years old at the time of diagnosis of the meningioma. The delay between the onset of symptoms suggesting SOM and diagnosis of temporal *en plaque* meningioma ranged between 1 and 10 years. Most cases experienced various medical treatments or surgical procedures for a SOM before the diagnosis of meningioma was made. In most cases, VT insertion was undertaken and followed by severe episodes of discharge. In all the cases, CT was unable to detect the intracranial component of the meningioma, but three CT features were always found and considered as highly suggestive of a meningioma. This imaging triad includes soft tissue mass filling in the middle ear or mastoid, hyperostosis of the petrous bone, and hairy aspect of the margins of the affected bone. MRI with gadolinium enhancement detected the intracranial meningioma involving the middle ear or mastoid in all the cases.

From a therapeutic point of view, it seemed that conventional middle ear procedures (such as VT insertion, mastoidectomy, or tympanoplasty) were inefficient.

DISCUSSION

Meningiomas are the second most common tumors of the cerebellopontine angle (CPA), comprising 10 to 15% of all CPA tumors (5). There are two types of gross appearance of intracranial meningiomas: globular and *en plaque* meningiomas. The most common form is the globular meningioma presenting as a bulky mass compressing adjacent neural structures. For this reason, the most common symptoms of temporal bone meningiomas are sensorineural hearing loss, tinnitus, vertigo, gait disturbance, headache, or facial palsy (6–8). On the other hand, *en plaque* petrous meningioma infiltrates the dura without bulging into the CPA and spreads along the surface of the temporal bone. Thus, the diagnosis of an *en plaque* meningioma is usually made later when the tumor extends extracranially (2–4,9). The extension into the middle ear or mastoid can occur via a variety of pathways well described by Chang et al. (3): tegmen tympani, posterior fossa plate, internal auditory canal, or jugular foramen. In this case series, the exact route of extension of the intracranial lesion to the middle ear or mastoid was difficult to precisely assess because of the wide infiltration of the dura at the time of diagnosis. Nevertheless, imaging studies allowed to exclude the involvement of the tympanomastoid cavities through the internal auditory canal or jugular foramen. The extension of the meningiomas to the middle ear or mastoid occurred via the tegmen or posterior fossa plate or both in all the cases.

In our series, the tumors extended into the middle ear and mastoid, leading to symptoms suggesting SOM, such as conductive hearing loss, aural fullness or chronic discharge after VT insertion. Sporadic cases of temporal meningiomas presenting as chronic otitis media have already been published (2,3,9). With this clinical presentation, most of the patients underwent various medical treatments or surgical procedures (VT, mastoidectomy, tympanolasty) before the diagnosis of meningioma was assessed with neuroimaging. With Civantos et al. (2) and Chang et al. (3), we think that most primary extracranial meningiomas of the middle ear reported in association with previous articles (4) were misdiagnosed as secondary middle ear meningiomas of intracranial origin. As stated by Chang et al. (3), without MRI evaluation, such cases with an intracranial origin could have been misinterpreted as primary intratemporal tumors.

MRI with gadolinium is the imaging modality of choice to detect intracranial meningioma (1,3). MRI usually shows a dural-based enhancing soft tissue mass in meningiomas (1). Gadolinium enhancement has proven to be particularly useful in delineating *enplaque* meningiomas, where there is no expansive lesion bulging intracranially (1). Nevertheless, the first-line imaging technique for the evaluation of chronic otitis media is CT without contrast injection (10). CT alone cannot detect intracranial *en plaque* meningioma, but some indirect associated radiological signs are highly suggestive of an *en plaque* meningioma and can lead to complete the imaging evaluation with MRI. In most of the cases reported in association with our series, CT showed the association of a nonspecific opacity filling in the tympanomastoid cavities, a hyperostotic reaction of the temporal bone, and a hairy aspect of the margins of the affected bone

(Figs. 1 and 2). Sclerosis limited to the mastoid is usually related to chronic otitis media, but differential diagnoses can be discussed especially if hyperostosis affects not only the mastoid region but other portions of the temporal bone, including fibrous dysplasia, osteopetrosis, meningioma, osteoma, ossifying fibroma, and bony metastases (1,11). To our knowledge, the three radiological signs described previously (soft tissue mass within the middle ear cleft or mastoid cavity and hyperostosis of the temporal bone with hairy aspect of the margins) have only been recognized in cases of *en plaque* meningioma.

Once a nasopharyngeal tumor has been ruled out, we recommend to perform a CT of the temporal bone in cases of atypical unilateral SOM persisting despite medical therapy or VT insertion. In this series, all the patients were women around 50 years old or older. In all the cases, VT insertion was followed by purulent discharge without significant improvement of hearing. CT has also to be considered in case of painful SOM or if neurotologic examination reveals atypical findings such as cranial nerve dysfunction. If CT suggests temporal *en plaque* meningioma, MRI is recommended to assess the diagnosis, thus avoiding surgical procedures to obtain a biopsy (Figs. 3 and 4).

Microsurgery is considered to be the gold standard in treating intracranial meningiomas (1,5,6,12). Meanwhile, even if those benign tumors are slightly radiosensitive (2), the place of radiation therapy in treatment algorithm has recently evolved, with the dramatic development of 3-D conformal dosimetry, such as fractionated radiation therapy, LINAC radiosurgery, or gamma knife radiosurgery (13–16). Thus, radiation therapy is administered using fractionated or single-dose modalities. Usual indications of radiation therapy are as follows: incomplete resection associated with growing tumor remnants; recurrence; histologic findings revealing atypical or anaplasia; unresectable lesions because of a patient's bad general condition; or involvement of structures like cavernous sinus, internal carotid artery, or basilar artery (1,13). Some authors consider radiation therapy to be the first-line treatment of selected cranial base meningiomas (15), but the trend is to promote a combination of microsurgery and radiotherapy in difficult cases (1,8,17).

Generally, in the setting of *en plaque* meningioma, the wide and limitless spread of the lesion and its usual extension toward unresectable structures make total surgical removal impossible. Extension toward the mastoid and middle ear is common when the temporal bone dura mater is especially involved. Attempts to perform complete excision of the *en plaque* tumor would lead to severe postoperative complications related to neural or vascular injuries, and to an increased risk of a cerebrospinal fluid leak after complex dural closure of large dura matter defect. In cases of asymptomatic *en plaque* temporal meningioma or if potential morbidity of the above-mentioned modalities of treatment seems particularly risky, conservative management with serial imaging follow-up can be safely decided (1,8). As presented in most cases of our series, anatomic considerations associated to the infiltrative features of the *en plaque* form make complete resection often impossible and dissection from neural or vascular structures very difficult (1,2,7). Thus, conservative management (i.e., wait and scan) was often the most appropriate option when the extent of the lesion was compared with the relative lack of symptoms.

The otologist must be aware of the inefficiency of VT insertion (like other middle ear procedures) that does not improve symptoms and is frequently followed by chronic or recurrent episodes of discharge. In case of cranial nerve involvement (particularly facial palsy or trigeminal neuralgia), selected otoneurosurgical decompression or conformal radiotherapy or a combination of both can be proposed (1,5,8,16).

CONCLUSION

Temporal *en plaque* meningioma involving the middle ear cleft or mastoid cavities can mimic unilateral SOM. In case of atypical or prolonged SOM, we recommend to perform CT of the temporal. When CT features are highly suggestive of a meningioma, this has to be confirmed with MRI. Conservative management is often the most valuable therapeutic option in cases of temporal *en plaque* meningioma extended to the middle ear or mastoid.

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