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Case Report

A novel drainage approach in patients with cholesterol granuloma: From petrous apex to mastoid air cell

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Abstract

Background: Cholesterol granulomas (CG) of the petrous apex (CGPA) are benign lesions that have high recurrence rates after surgical intervention. We describe the use of a robust silicon drain between the petrous apex and mastoid air cells to allow constant aeration of the lesion for preventing recurrence.

Case Description: A retrospective analysis was performed using the data of four patients treated at the Maastricht University Medical Centre between 2014 and 2016. Using the middle fossa approach, the petrous apex was reached, the cyst was opened, and the content aspirated. Subsequently, a robust silicon drain was placed between the cyst and mastoid air cell system. The outcome measures were clinical improvement of the symptoms and radiological parameters. The patients were female ($n = 2$) and male ($n = 2$) with an age range between 33 and 53 years at the time of the operation. Computed tomography and magnetic resonance imaging scans were used to confirm CG diagnosis. The most common presenting symptoms in our population were diplopia and headaches. The symptoms improved after surgery and there were no complications. Thus far, no recurrence has been observed and imaging shows aeration in the lesion area.

Conclusion: The use of a robust drain seems to be an effective, safe, and feasible option to prevent recurrences in patients with CG.

Key Words: Cholesterol granuloma, drainage, mastoid, petrous apex

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INTRODUCTION

Cholesterol granulomas (CG) are rare and slowly growing cystic lesions surrounded by fibrous tissue. These lesions are formed via the reaction of foreign body giant cells against cholesterol crystals.[6,11] The promoting factor is most likely to be an immune reaction to crystal deposits within the air cells.[10] There are two hypotheses linked to this. The first is the obstruction vacuum hypothesis. A chronic obstruction of pneumatization leads to the formation of negative pressure. This in turn results in

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the extravasation of intravascular fluid into the air cells' mucosa and the formation of edema. Degradation of blood products of the edema, hemosiderin, in particular, triggers an inflammatory reaction.[4,10] A newer hypothesis, known as the exposed marrow hypothesis, states that the hemorrhage stems from the erosions of the marrow-filled cavities in the petrous apex.[7]

Symptoms of CGs vary based on the location, size, and involvement of surrounding anatomical structures. Most lesions become symptomatic when they compress the adjoining structures, usually the cranial nerves V, VI, VII, or VIII. As a result, presenting symptoms are often related to a cranial nerve function deficit and include trigeminal neuralgia, diplopia, facial weakness, facial spasms, deafness, vertigo, tinnitus, headaches, and/or seizures.[10] Most common presenting symptoms are hearing loss, vertigo, and headaches.[12,13]

Computed tomography (CT) images show a well-defined expansive and erosive lesion with a density similar to that of the brain. On magnetic resonance imaging (MRI), the lesions appear with high intensity signal on both T_1 and T_2-weighted images due to presence of cholesterol. The rim of the lesion on T_2-weighted imaging appears with a low intensity signal because of hemosiderin. No attenuation is seen on fluid attenuated inversion recovery (FLAIR) sequences. Apparent diffusion coefficient (ADC) sequences show no restriction of diffusion.[2] Fat suppression on MRI results in the disappearance of the granuloma. In early stages of CG, CT scans may not show erosions and MRI may show different intensities than the ones seen in later stages.[1,5,6]

When a lesion becomes symptomatic, surgical intervention is the preferred management strategy.[10] The goal of the surgery is to achieve adequate decompression of the cystic content. Because recurrence rates are as high as 60%,[5] surgeons have tried to keep the cyst constantly oxygenated and drained. A vascularized temporal muscle flap was used for this purpose in our clinic[5] and unfortunately, was ineffective in preventing recurrence. In our last four surgeries, we have used an alternative approach, which is based on the hypothesis that constant aeration of the cyst allows for pneumatization.[3] We placed a robust silicon drain, used for ventricular CSF drainage in patients with subarachnoid hemorrhage (SAH), between the cyst and the mastoid air cell system. Here, we describe the surgical details of the cases and the outcome.

METHODS AND RESULTS

Patients with the diagnosis of petrous apex CG who were referred to our Skull Base surgery team at the Maastricht University Medical center (MUMC+) in the Netherlands were included in this study after informed consent had been obtained. Due to the small sample size, formal statistical analyses were not performed. Detailed documentation on clinical and surgical information, complications, pre- and postoperative imaging findings, revision surgery, and audiometric data were analyzed.

Surgical procedure

The cyst was reached via a middle fossa extradural approach, unroofed by drilling and aspirated.[2] After a connective canal was drilled between the apex of the os petrosum and the mastoid, the SAH drain (Medtronic 26020; Medtronic, Minneapolis, MN, USA) was placed with one end in the cyst and another in the mastoid air cell system [Figure 1].

CASES DESCRIPTIONS

The cases are briefly described below. No major comorbidities were present in the patients described here. The order of the cases is based on the duration of follow-ups.

Case 1

A 53-year-old male patient, with the diagnosis of CG since 2003 with slow progression, presented with diplopia and headache for a couple of weeks. MRI showed a 2.2 × 1.4 cm lesion in the transversal plane above the apex of the right os petrosum. The lesion appeared with a high intensity signal on both T_1 and T_2-weighted imaging. The rim of the lesion showed a low intensity signal on T_2-weighted imaging.

Case 2

A 43-year-old female patient, previously operated for CG in 2009, was experiencing facial spasms for 2 months along with an occipital headache. MRI showed a 1.5 × 0.9 cm lesion in the apex of the left os petrosum. The lesion demonstrated a high intensity signal on both T_1 and T_2-weighted images which was indicative for recurrence.

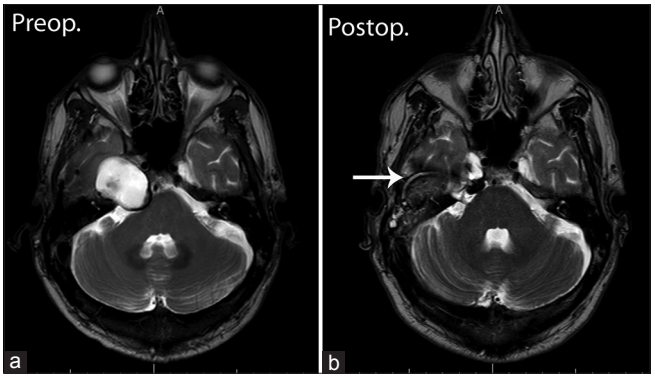

Figure 1: (a) Axial T_2-weighted imaging of a 3.5 × 2.4 cm cystic lesion of the right petrous apex consistent with cholesterol granuloma before intervention. (b) Axial T_2-weighted imaging after intervention.The arrow indicates the SAH drain between the petrous apex and the mastoid air cell

Case 3

An incidental finding in a 33-year-old female patient revealed a CG in the apex of the left os petrosum. The MRI showed a heterogeneous lesion with a size of 2.5 × 1.5 cm. T_1-weighted imaging revealed a bright lesion in the caudal and dorsal parts, but hypointense in the ventral one. The high signal did not disappear on T_1 with fat suppression. Distribution was not restricted. The patient suffered from a generalized epileptic seizure of the left hemisphere, which is unrelated to the CG. After treatment, with the use of carbamazepine, no additional seizures occurred.

Case 4

A 52-year-old male patient had been experiencing paresis of the right abducence nerve for 8 months. The MRI showed a 3.5 × 2.4 cm lesion above the apex of the right os petrosum. The lesion showed a high intensity signal on T_1-weighted imaging. Even though the cranial part of the lesion also showed a high-intensity signal on T_2-weighted imaging, the caudal portion illustrated a low intensity signal [Figure 1]. In addition, the caudal part showed restrictive diffusion, making the diagnosis of CG challenging. Surgical intervention confirmed CG.

RESULTS

Diplopia and headaches were the most common presenting symptoms in our population. All four patients underwent surgery via the middle fossa approach. A summary of patient characteristics at presentation and surgical outcome is presented in Table 1. The cyst was aspirated and a drain was placed. Follow-up periods ranged between 5 months and 2 years. Thus far, no evidence of recurrence was shown upon evaluation. Interestingly, in all patients, pneumatization is enhanced. Two patients became asymptomatic after surgery (cases 1 and 2). The other two showed an improvement in the symptoms. One had an improvement of hearing, even though a hearing aid was recommended (case 3) and the other had an improvement of diplopia (case 4). Case 4 developed a peripheral facial paresis 2 weeks after surgery which was attributed to a viral cause. He received steroid treatment and later made a full recovery.

DISCUSSION

CG is a rare, benign entity that is most commonly found in the petrous apex.[5] Most of the lesions remain clinically and radiologically stable. The likelihood of a sudden growth of the lesion is low.[12] A clinical manifestation of symptoms requires a surgical intervention. Two approaches are used in the clinic for CG – drainage or complete resection of the cyst.[8] Nonetheless, the optimal approach is still debated. Drainage is the more commonly used approach.[9] Drainage has higher recurrence rates than complete resection with a vascularized flap.[9] The average recurrence rate for drainage is 22%, whereas it is 3% in resections. Recurrence in drainage is attributed to a fibrous tissue formation resulting in an obstruction of the drainage site.[9] This in turn prevents long-term aeration. The SAH drain is a robust drain minimizing the risk of obstruction. It will likely remain patent for longer periods of time in comparison with the stents that are currently in use. Securing constant aeration is most probably the critical factor in preventing recurrence. Nonetheless, blockage of the drain remains possible.

The use of a vascularized temporal muscle flap did not seem to be an effective approach in our clinic because we observed muscle atrophy. However, cautious interpretation of results should be applied as information regarding the average period for CG recurrence is scarce, and thus, our follow-up period might also be too short. In addition, the sample size is rather small due to the rarity of the disease and the relatively short inclusion period.

We could not find any information regarding infection rate after drain placement in the literature. However, a superimposed infection of the drain is a possibility. In addition, displacement of the drain can be a possible complication. These complications have not been observed in our case series. Therefore, we will follow-up with our patients for a longer period. Furthermore, to reduce the likelihood of fibrous formation in the drain, we will explore the possibility of designing a three-dimensional printed biocompatible drain between the mastoid air cell and petrous apex.

Table 1: Characteristics of the four patients described in the study

Case	Gender and age	Size (cm) and location	Previous CG	Preoperative symptoms	Duration of follow-up	Symptoms on follow-up	Complications
1	M, 53	2.2 × 1.4, right	0	Diplopia and headache	5 months	Asymptomatic	None
2	F, 43	1.5 × 0.9, left	1	Facial spasms	9 months	Asymptomatic	None
3	F, 33	2.5 × 1.5, left	0	Reduced hearing and tinnitus	14 months	Improvement of hearing and disappearance of tinnitus	None
4	M, 52	3.5 × 2.4, right	0	Diplopia and headache	27 months	Improvement of diplopia. Abduction was still limited	peripheral facial paresis (viral cause)

CONCLUSION

Our approach of using a SAH drain between the petrous apex and mastoid air cell seems to be effective in achieving constant aeration and contributes toward the prevention of recurrence in CG.

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Conflicts of interest
There are no conflicts of interest.

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