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OTOLOGY

Complications of chronic otitis media with cholesteatoma during a 10-year period in Kosovo

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Abstract We review and discuss the results of treatments for complications of cholesteatomatous chronic otitis media (CCOM) in a tertiary health care center. In a retrospective study, the medical records of patients with complications of CCOM who had undergone surgical treatment at the ENT Clinic of the University Clinical Center of Kosovo for the period 1994–2004 were reviewed. From a total of 1,803 patients suffering from CCOM, in 91 patients, 55 (60.4%) men and 36 (39.6) women, one or two complications are recorded. The mean age of the subjects was 30 years, and the age range was from 1 to 76 years. Extracranial (EC) complications were observed in 52 cases (57.1%), and intracranial (IC) complications were seen in 29 patients (31.9%). Twelve patients (11%) had multiple complications. For the EC cases, we found that subperiostal mastoidal abscess occurred in 26% of the all patients, facial nerve palsy was seen in 16.48% and labyrinthine fistula occurred in 10%. For the IC cases, meningitis (19.7%) and perisinusal abscess (15.3%) were the most common complications. The most often isolated pathogen from ear swabs was *Proteus mirabilis* in 33.3% of cases. The most frequent radiological diagnostic procedures were mastoid tip X-rays, which were performed in 77% of the patients, and computed tomography in 24%; magnetic resonance imaging was not performed on any of the patients during the study period. Patients with EC complications were treated in the ENT Clinic, whereas patients with IC complications, after otologic surgical procedures, were transferred to the Neurosurgery Clinic or to the Clinic for Infectious Diseases. In this series, three patients (3.3%) died as a result of complications, while the remaining 96.7% survived. Complications of COM with cholesteatoma can represent life-threatening conditions, and close cooperation between otosurgeons, neurosurgeons and infectious disease specialists is mandatory.

Keywords Complications of cholesteatoma · Chronic otitis media · Surgical treatment

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Introduction

Cholesteatoma is an epithelial-lined growth containing mainly cellular debris in which cholesterol crystals can generally be found. Cholesteatomas are benign and occur mainly in the middle ear and mastoid region, and through pressure, cause destruction of surrounding structures [1].

Once established in the middle ear, mastoid or petrous bone, cholesteatoma is a destructive lesion that gradually expands and destroys adjacent structures, leading to complications. The pathogenesis is diverse, with different pathways leading to the same destructive lesion [2]. The majority view of pathogenesis now favors retraction of Shrapnel's membrane or the posterosuperior quadrant of the pars tensa, which develops into a gradually expanding cyst containing epithelial debris having locally erosive properties [3].

Although the incidence of complications due to otitis has decreased in recent decades, they still pose a challenge for

clinicians, because the complications have an insidious presentation that is usually hidden by the indiscriminate use of antibiotics [4]. The objective of surgery for chronic otitis media with cholesteatoma and chronic otomastoiditis is to eradicate the disease and create anatomic conditions to prevent recurrence and complications. To achieve this purpose, two surgical techniques exist: canal wall-up (CWU) and canal wall-down (CWD) mastoidectomies [5].

Complications of CCOM are classified as extracranial (EC) or intracranial (IC). EC complications are recorded more often than IC complications, but IC complications are more severe and lethal. Consequently, studying EC and IC complications is clinically important.

The goal of the present study was to analyze the management of complications of cholesteatoma for a 10-year period at our center retrospectively. Because the University Clinical Center of Prishtina is the only referral center for otogenic complications in Kosovo, the incidence of complications due to cholesteatoma can be estimated accurately. The present study, which included all patients hospitalized for complications of cholesteatoma over a 10-year period, describes the diagnosis and treatment of complications due to CCOM. In particular, we focus on the cause and duration of chronic ear signs and symptoms and discuss otosurgical approaches.

Material and methods

We reviewed the clinical records of patients diagnosed as having CCOM and treated at the ENT/Head and Neck Surgery Clinic, University Clinical Center, Prishtina, Kosovo, from January 1994 through December 2003. The diagnosis of a complication of CCOM was established by experienced otolaryngologists based on the clinical examination, otomicroscopy and radiology examinations and was confirmed during the surgical procedures. Cases with COM without an intraoperative finding of cholesteatoma were excluded from the study.

The demographic data included sex, age and residence (rural or urban). Signs and symptoms on admission, white blood count (WBC), erythrocyte sedimentation rate (ESR) and the results of middle ear aspirate culture obtained on admission were also recorded. In all cases with a suspected complication, X-rays of the mastoid were performed, and occasionally, computed tomography (CT) of the temporal bone, brain and neck was performed. The surgical procedures were recorded, and materials were harvested for pathology confirmation.

With regard to patient consent, we obtained written permission from the clinic director to conduct research on this topic, because our institution lacks an institutional review board. In addition, every patient admitted to the clinic must sign a consent form giving permission to conduct all diagnostic and therapeutic procedures, including the publication of images or other medical records.

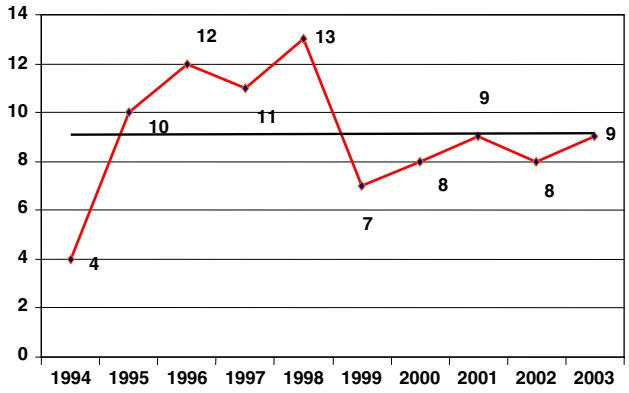

Fig. 1 Complications of CCOM by year

Results

During the 10-year period from January 1994 to December 2003, 1,803 patients were admitted with diagnosis of otitis media. We found that 92% of these patients had COM, while 8% had acute otitis media. From a total of 1,803 patients, in 272 of them, an intraoperative cholesteatoma is confirmed. From this 272 patients with CCOM, 91 (33.5%) came to the clinic with either one or two complications. We also found that complications of COM (without cholesteatoma) occurred in only 6.7% of cases.

The number of patients with complications of cholesteatoma decreased during the last 5 years of the study period as shown in Fig. 1. This decrease corresponds to the end of the military conflict in Kosovo in 1999 and was likely due to the subsequent rapid reconstruction of the health care system, which improved primary health care and medical education.

The detailed data of 91 patients with complications of CCOM included in the study are presented in the Table 1, where 55 (60.4%) were men and 36 (39.6%) women, that presents statistically significant difference (χ^2 test = 3.97, $P < 0.05$). The mean age was 30 years, with a standard deviation (SD) of 16.5 years (SD $\pm$ 16.54), ranging from 1 to 70 years. The percentage of pediatric patients (<15 years old) was 20.9%. According to residence, urban versus rural, there were no significant difference: 50.5% versus 49.5% (χ^2 test = 0.01, $P > 0.05$).

The duration of disease, from the first visit to the ENT doctor to the moment of surgery, is presented in Table 2. The mean duration of disease was 8.6 years, with a standard deviation of 8.9. Only 17.6% of patients carried the cholesteatoma for less than 1 year. The same percentage of patients had episodes of exacerbation for more than

Table 1 Patient by gender, residence and age

	Number *N* (%)	*P* value
Gender		
Men	55 (60.4%)	0.046
Women	36 (39.6%)	
Residence		
Urban	46 (50.5%)	0.917
Rural	45 (49.5%)	
Age (years)		
Mean ± SD	29.90 ± 16.54	
Rank	1–70	
<15 years	19 (20.9%)	

Table 2 Duration of disease (CCOM)

Duration in years	Number (*N*)	Percent (%)
<1	16	17.6
1–5	35	38.5
6–10	17	18.7
11–15	5	5.5
>15	16	17.6
No data	2	2.2
Total	91	100.0
Mean	8.6 years	
SD	8.9	

15 years. Most of the patients (38.5%) carried the cholesteatoma from 1 to 5 years.

Types of complications of CCOM are shown in Table 3. EC complications occurred in 52 patients (57.1%), IC complications in 29 (31.9%) and multiple complications (IC + EC) in 10 patients (11%). In IC group, there were two cases (2.2%) with multiple complications (IC + EC), considering sepsis as result of an intracranial complication. For the EC cases, we found that subperiostal mastoidal abscess (SMA) occurred in 26% of the all patients, facial nerve palsy (FNP) was seen in 16.48% and labyrinthine fistula occurred in 10%. For the IC cases, meningitis (19.7%) and perisinusal abscess (15.3%) were the most common complications. In group of multiple complications (IC + EC), combination of perisigmoid sinus abscess and SMA was the one that most often occurred (4.4%). Meningitis and SMA was the second most often occurred one.

Diagnostic procedures applied for this series of patients was not standardized and differs from patient to patient. For some procedures, e. g. CT scans, determinative was availability. For the period 1994–2003, CT was not available in our center in all cases and is done only in 24% of cases; pure tone audiometry can be performed only in conscious patients. The Table 4 shows the percentage of diagnostic

Table 3 Types of CCOM complications

Type of complication	Number (*N*)	Percent (%)
Extracranial complications		
SMA	24	26.4
FNP	13	14.3
Perilabyrinthine fistula	9	9.9
Acute mastoiditis	4	4.4
Zygomatitis/perizygomatic abscess	1	1.1
Bezold's abscess	1	1.1
Total	52	57.1
Intracranial complications		
Meningitis	11	12.1
Perisigmoid sinus abscess	10	11.0
Cerebral abscess	3	3.3
Meningoencephalitis	2	2.2
Subdural abscess	1	1.1
Perisigmoid sinus abscess + sepsis	1	1.1
Meningitis + sepsis	1	1.1
Total	29	31.8
Multiple (IC/EC) complications		
Perisigmoid sinus abscess + SMA	4	4.4
Meningitis + SMA	2	2.2
Cerebral abscess + FNP	1	1.1
Meningitis + petrositis	1	1.1
Meningitis + FNP	1	1.1
Perisigmoid sinus abscess + jugular vein thrombophlebitis + SMA	1	1.1
Total	10	11.0
Total	91	100.0

SMA subperiostal mastoid abscess, *FNP* facial nerve paralysis

Table 4 Diagnostic procedures

	Total number of patients*n* = 91	
	N	%
ESR	91	100.0
WBC	91	100.0
CT	22	24.2
Mastoid X-rays	64	70.3
PTA	44	48.4
Ear swabs	44	48.4

ESR erythrocyte sedimentation rate, *WBC* white blood count, *CT* computed tomography, *PTA* pure tone audiometry

procedures performed in patients of our series. Laboratory analyzes, erythrocyte sedimentation rate (ESR) and white blood count (WBC) was taken in all cases and elevated values indicated developing complications. In 70.3% of patients, mastoid tip X-rays were performed; in cases

suspected of IC complications, CT scans were performed. PTA shows conductive hearing loss in diseased ears.

The microbiology analysis results are shown in Table 5. Swabs from draining ears, myringotomy incisions or the mastoid cavity were only taken in 44 patients, and pathogenic bacteria were isolated in only 33 patients. The most frequently isolated pathogens were *Proteus mirabilis* in 33.3%, *Staphylococcus aureus* in 15% and *Pseudomonas* sp. and *Proteus vulgaris* in 12% of the 44 cases.

The surgical procedures for the treatment of complications are summarized in Table 6. CWD radical tympanomastoidectomy (RTM) was performed in 86.8% of the patients and was the most frequently applied procedure. CWU procedures were only applied in 13.2% of the patients.

The modifications of the CWD procedure in each of the 79 applicable cases were different and depended on the spread of disease and the type of complications. The modifications of the CWD procedure are listed in Table 7. The most common open techniques applied were RTM in 34.2% of the patients, RTM with middle fossa dura exposure in 14% and RTM with lateral sinus exposure in 11.4%.

The end results regarding the management of this series of patients were the following: seventy-seven patients (84.6%) were cured and followed as outpatients for at least 1 year. Conversely, 10 patients (11%) were transferred to other departments, three died and one patient left our department. In addition, one patient was transferred to the Abdominal Surgery Department because of acute abdominal distress. The three patients who died had intracranial complications: two had meningitis and one had meningitis and a brain abscess.

Table 5 Isolated bacteria from ear swabs

Bacteria	Number (*N*)	Percent (%)
Proteus mirabilis	11	33.3
Staphyloccocus aureus	5	15.2
Proteus vulgaris	4	12.1
Pseudomonas spp.	4	12.1
Citrobacter	3	9.1
Escherichia coli	3	9.1
Streptoccocus pneumoniae	1	3.0
Klebsiella pneumoniae	1	3.0
Morganella morganii	1	3.0
Total	33	100.0

Table 6 Surgical procedures applied in patients with complications

Surgical procedure	Number (*N*)	Percent (%)
Radical tympano-mastoidectomy	79	86.8
Simple mastoidectomy + tympanoplasty	6	6.6
Mastoidectomy	5	5.5
Antrotomy	1	1.1
Total	91	100.0

Table 7 The modifications of CWD procedures applied in 79 patients with cholesteatoma complications

CWD procedure	Number (*N*)	Percent (%)
RTM	27	34.2
RTM + MFDE + SSE	12	15.2
RTM + MFDE	11	13.9
RTM + SSE	9	11.4
RTM + labyrinthine fistula closure	7	8.9
Modified RTM	3	3.8
Revision RTM	3	3.8
RTM + facial canal plasty	2	2.5
TOT	1	1.3
TOT + craniotomy	1	1.3
RTM + perizygomatic abscess incision	1	1.3
RTM + SSE + Bezold's abscess evacuation	1	1.3
RTM + Middle fossa extradural abscess evacuation	1	1.3
Total	79	100.0

RTM radical tympano-mastoidectomy, *MFDE* middle fossa dura exposure, *SSE* sigmoid sinus exposure, *TOT* tympanoplasty in open technique

Discussion

Adam Politzer in 1869 (published in 1909) clearly identified the potential seriousness of otitis media in the preantibiotic era based on the anatomic location and boundary with vital structures by stating that "the temporal bone has four sides: the outside is bounded by life, from which there comes the opening of the auditory canal, one form of our appreciation of what life means; on the other three sides this bone is bounded by death". The advent of antimicrobials, an increasing of awareness of physicians and the public regarding the potential seriousness of disease and diagnostic methods yielding earlier identification and treatment immensely reduced the incidence of lethal complications [6, 7].

With regard to the prevalence and incidence of complications, many studies have counted the prevalence of patients having COM with cholesteatoma who develop complications, because calculating the incidence was not appropriate. The results of large series of patients have shown a similar rate of otogenic complications compared to the results of our series. During a 9-year period (1990–

1999), of 2,890 patients with acute and chronic otitis media, Osma et al. found 93 cases involving complications, which consisted of 57 IC cases (59%) and 39 EC cases (41%), suggesting that 3.22% of all patients with middle ear infections develop a complication [8]. Kangsaranak et al. reviewed 17,000 patients with COM and found 102 cases with IC and EC complications; they calculated the prevalence of complications as 0.69% among populations with suppurative COM [9]. This same group reviewed another series of 24,321 patients from a 13-year period and found 87 cases with 140 IC complications; they calculated a prevalence of 0.36% for IC complications [10]. As a comparison, we found complications in 5% of the cases in our series (91 out of the 1,803 patients with acute and chronic otitis media). Our prevalence was high because our series consisted of inpatients. If we had included the outpatients with COM, we would likely have obtained a much lower prevalence that was similar to the results in the literature. The highlight of our results is the finding that CCOM developed complications in 33.5% of the cases in our patient series, whereas COM without cholesteatoma developed complications in only 2% of the cases.

The duration from the onset of disease to the appearance of complications was important in our work. In our series, the mean duration of disease was 8.6 years. More than 62% of the patients reported a duration of 1–15 years, while 17% had duration of less than 1 year, which was similar to the percentage of patients with disease duration of more than 15 years.

Studies on the complications of cholesteatoma have found different percentages of the men-to-women ratio. In the series of Osma et al. involving 93 patients [8], the authors found a significant difference in the men-to-women ratio of 2:1. In contrast, Keles et al. in a series of 51 patients did not find a significant difference in the men-to-women ratio, observing that 57% of the patients were men and 43% were women [11]. In our series, of the 91 patients with complications from cholesteatoma, we found that 60% were men and 40% were women.

Complications due to cholesteatoma can occur in patients of all ages, from infants to older adults [8–12]. In our series, the age structure of the patients was different, with a slight domination in the 11- to 20-year-old group. The mean age was 30 years, and the patients ranged in age from 1 to 70 years.

COM is a suppurative bacterial infection, and its causative agents can be isolated using ear swabs. This is important because primary surgical therapy must always be complemented with antibiotic therapy chosen according to the microbiology lab results. In a meta-analysis of seven publications, of a total of 280 patients, Dhooge et al. found that *Streptococcus pneumoniae* was the most commonly isolated causative agent, occurring in 23% of cases. *Staphylococcus* coagulase-positive bacteria were seen in 19% of cases. *Streptococcus β-hemolyticus* group A was seen in 16% of cases, and *Haemophilus influenzae* was detected in 2% of the cases. They also found that 20% of cases did not show pathogenic bacteria in the cultures [13]. Other studies have suggested that *Pseudomonas aeruginosa* is the leading causative pathogen in COM with cholesteatoma [14, 15]. In our series, material for microbiology analyses was only taken from 33 patients, and the most frequently isolated bacterial agent was from the Proteus group as follows: *Proteus mirabilis* from 33% of the swabs, *Staphylococcus aureus* from 15%, *Proteus vulgaris* from 12% and *Pseudomonas* spp. from 12%. *Streptococcus pneumoniae* was only isolated from 3% of the swabs. Antibiotic therapy in cases with microbiology lab results is determined by an antibiogram, whereas antibiotic therapy is determined by empirical evidence in cases without lab results. In cases with meningitis, therapy is given after consultation with an infectious disease specialist.

Although the clinical history and clinical exam play important roles in the diagnosis of complications, the main diagnostic tool is clinical imaging. The gold standard is CT, because this imaging provides a good view of bony structures; magnetic resonance imaging (MRI) is better for the visualization of soft tissue pathologic changes. According to the protocols of many centers, the suspicion of a complication necessitates a CT scan within the first 24 h of admittance, in coronal and axial cuts at 1-mm slices, using a high resolution of 30–50 mA.

When an intracranial complication is suspected, a contrast CT scan must be performed. MRI T-1- and turbo T-2-weighted scans allow better visualization of brain soft tissue and blood vessels in coronal and axial sections with or without contrast [16]. The sensitivity of CT in the current diagnosis of intratemporal complications is reported to be 87–100% [17, 18]. In our series, the majority of patients did not receive CT scans for the diagnosis of complications, because these devices were not prevalent in Kosovo in the late 1990s. Temporal bone plain X-rays were applied in 77% of the patients in our series, and CT scans were only applied in 24%. MRI was not applied to any patient. The other diagnostic procedures included laboratory findings (ESR, WBC) and the results of lumbar puncture.

A wide range of surgical techniques are used for the treatment of complications due to cholesteatoma, but the only effective treatment is surgery. For EC complications, otosurgeons must be fully trained to deal with all possible complications and to adapt the approach and surgical method to achieve a clean and dry ear for a long time during the follow-up. For IC complications, close cooperation between otosurgeons and neurosurgeons is mandatory [19]. In recent publications, the authors have agreed that otogenic meningitis must be treated with middle ear surgery (mastoidectomy, simple or radical) in combination with

antibiotics. For lesions located adjacent to the dura or tegmental area, such as extradural abscesses and sigmoid sinus thrombosis, mastoidectomy performed by an otosurgeon is the only necessary approach. For deep cerebral and cerebellar abscesses, craniotomy performed by a neurosurgeon working with otosurgeons is the treatment of choice [20]. In our series, all patients with EC complications were treated in the ENT Clinic by a senior otosurgeon. No deaths were recorded among the EC complications group. The IC complications group was treated primarily in the ENT Clinic with mastoidectomy alone or in combination with sigmoid sinus exploration in cases with thrombosis of the sigmoid sinus. Patients with deep cerebral abscesses were treated surgically twice: first by mastoidectomy in the ENT Clinic and second by craniotomy in the Neurosurgery Clinic. Three deaths occurred in this group: two patients with otogenic meningitis and one with a cerebral abscess died, representing a mortality rate of 3.3% among the patients with complications.

Conclusions

- Patients having COM with cholesteatoma develop complications more often than patients having COM without cholesteatoma; so the management of patients with cholesteatoma must be a priority for the ENT health care system.
- The treatment of the complications due to cholesteatoma is surgical, and trained otosurgeons must be ready to perform all needed interventions.
- Complications of COM with cholesteatoma represent life-threatening conditions, and close cooperation among otosurgeons, neurosurgeons and infectious disease specialists is mandatory.

References

1. Urdang L (ed) (2000) The Bantam medical dictionary, 3rd revised edn. Bantam Books, New York, p 94
2. Olszewska E, Wagner M, Bernal-Sprekelsen M et al (2004) Etiopathogenesis of cholesteatoma. Eur Arch Otorhinolaryngol 261(1):6–24
3. Robinson JM (1997) Cholesteatoma: skin in the wrong place. J R Soc Med 90:93–96
4. Saffer M, Lubianca Neto JF, Arrarte JLF, et al (1997) Lateral sinus thrombophlebitis and cervical abscess as a complication of chronic otitis media. J Pediatr (Rio de Janeiro) 73(4):269–272. (In Portuguese)
5. Kos MI, Castrillon R, Montandon P, Guynot JP (2004) Anatomic and functional results of canal-wall down mastoidectomy. Ann Otol Rhinol Laryngol 113:872–876
6. Lederer FL (1960) The development of otorhinolaryngology and bronchoesophagology. Int Cell Surg 33:83–97
7. Goycoolea MV, Jung TK (1992) Complications of suppurative otitis media. In: Paparella MM et al (eds) Otolaryngology, vol II: otology and neuro-otology, IIIrd edn. Saunders Company, Philadelphia, p 1382
8. Osma U, Cureoglu S, Hosoglu S (2000) The complications of chronic otitis media: report of 93 cases. J Laryngol Otol 114(2):97–100
9. Kangsanarak J, Foonant S, Ruckphaopunt K, Navacharoen N, Teotracul S (1993) Extracranial and intracranial complications of suppurative otitis media. Report of 102 cases. J Laryngol Otol 107:999–1004
10. Kangsanarak J, Navacharoen N, Foonant S, Ruckphaopunt K (1995) Intracranial complications of suppurative otitis media: 13 years' experience. Am J Otol 16(1):104–109
11. Keles E, Kaygusuz I, Karlidag T, Yalcin S, Sakallioglu O, Alpay HC (2004) The complications of otitis media: retrospective assessment of 51 cases. Turk Arch Otolaryngol 42(4):215–219
12. Albers FWJ (1999) Complications of otitis media. The importance of early recognition. Am J Otol 20(1):9–12
13. Dhooge IJ, Albers FWJ, Van Cauwenberge PB (1999) Intratemporal and intracranial complications of acute suppurative otitis media in children: renewed interest. Int J Pediatr Otorhinolaryngol 49(Suppl 1):S-109–S-111
14. Luntz M, Brodsky A, Nusen S, Kronenberg J, Keren G, Migirov L, Cohen D, Zohar S, Shapira A, Ophir D, Fishman G, Kisilevsky V, Magamse I, Zaaroura S, Joachims HZ, Goldenberg D (2001) Acute mastoiditis—in the antibiotic era: a multicenter study. Int J Pediatr Otorhinolaryngol 57(1):1–9
15. Butbul-Aviel Y, Miron D, Halevy R, Koren A, Sakran W (2003) Acute mastoiditis in children: *Pseudomonas aeruginosa* as leading pathogen. Int J Pediatr Otorhinolaryngol 67(3):277–281
16. Vazquez E, Castellote A, Piqueras J, Mauleon S, Creixell S, Pumarola F, Figueras C, Carreño JC, Javier Lucaya J (2003) Imaging of complications of acute mastoiditis in children. Radiographics 23:359–372
17. Veillon F, Riehm S, Roedlich MN, Meriot P, Blonde E, Tongio J (2000) Imaging of middle ear pathology. Semin Roentgenol 35:2–11
18. Dhooge IJ, Vandenbussche T, Lemmerling M (1998) Value of computed tomography of the temporal bone in acute otomastoiditis. Rev Laryngol Otol Rhinol 119:91–94
19. Ozkaya S, Bezircioglu H, Sucu HK, Ozdemir I (2005) Combined approach for otogenic brain abscess. Neurol Med Chir (Tokyo) 45(2):82–85
20. Bento R, de Brito R, Ribas GC (2006) Surgical management of intracranial complications of otogenic infection. Ear Nose Throat J 85(1):36–39