

COMPLICATIONS OF CHRONIC CHOLESTEATOMATOUS OTITIS MEDIA: ABOUT EIGHT CASES

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ABSTRACT

A total of 156 cases of the middle ear cholesteatoma treated in our ENT department since 2005, 8 patients presented for the first time for cholesteatomatous complications (5.13% of cases). Intracranial complications in three patients (37.5%), intrapetrous in two patients (25%), extrapetrous in two cases (25%), intra- and extracranial in one patient (12.5%). Children represent 37.5% of cases (three cases), adults under 30 represent 37.5% of cases (three cases), two cases are over 30 years old (25%). Two patients had more than one complication (25%). Radical mastoidectomy was performed for Bezold's abscess, Tympanoplasty with canal wall down was performed for all four patients with intracranial abscess and tympanoplasty with canal wall up was performed in three situations (labyrinthine fistula, facial paralysis and temporo-zygomatic abscess). The cholesteatoma excision was performed simultaneously at, the evacuation of the intracranial abscess in only one case, the drainage of extracranial abscesses in two cases, the repair of the bone defect concerning the facial palsy and the labyrinthine fistula; it was also carried out in a deferred manner in three situations of intracranial complications. The microorganism was isolated in two cases. Mortality in this series was estimated at 12.5%.

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INTRODUCTION

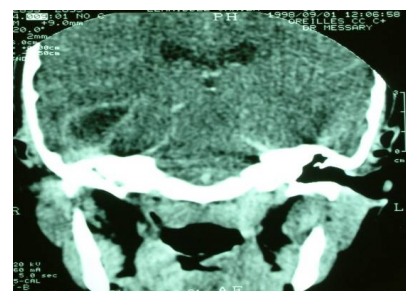
The complications of cholesteatoma have become rare since the advent of antibiotic therapy. However, these complications are still present with an always high prevalence in the developing countries [1]. They may be isolated or associated in the same patient. Diffusion of the infection from the middle ear cavities by different mechanisms can lead to intracranial and / or extracranial complications [2] [3]. We report a series of eight patients with cholesteatomatous otitis who presented eleven complications. These complications were intracranial and extracranial; in two patients, they were associated complications.

Clinical Cases

CASE 1: Right temporal abscess

A twelve-year-old girl with history of right chronic otitis media, she was admitted in ENT for febrile right purulent otorrhea. The otoscopy has objectified a cholesteatoma of the middle ear. The evolution was marked by the installation of a meningeal syndrome with obtundation and signs of localization (aphasia and convulsions). CT scan diagnosed a temporal abscess in contact with a cholesteatoma of the middle ear (Figure1) and the patient was rapidly given

ceftriaxone 500g / 8hours and metronidazole 250mg / 12hours. The drainage of the abscess took place immediately, the bacteriological study of which was able to isolate the proteus mirabilis.



A



B

Figure 1 Coronal CT scan showing (A) right temporal abscess encapsulated in continuity with a right middle ear cholesteatoma; (B) After drainage of the temporal abscess, the appearance of a cholesteatomatous filling of the middle ear.

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Ten days later a tympanoplasty with canal wall down was able to eradicate the cholesteatoma of the right ear. The patient had no recurrence during a three-year follow-up.

CASE 2: Subdural empyema and thrombophlebitis of the lateral sinus

A 21 year old male patient was admitted into internal medicine for an isolated febrile syndrome. Then a meningial syndrome marked the first 48 hours of evolution; Otoscopy was able to recognize neglected cholesteatomatous otitis. The cranial CT scan found a subdural empyema and a lateral sinus thrombophlebitis complicating a left ear cholesteatoma (Figure2). The patient received a combination of Ceftriaxone1g / 12H and metronidazole 500mg / 12H for a period of 6 weeks, followed by tympanoplasty with canal wall down. The following clinical and paraclinical controls were very satisfactory with a two-year follow-up.



Figure 2 Axial CT scan showing a subdural empyema (white arrow) with lateral sinus thrombophlebitis (black arrow) in contact with the cholesteatomatous otitis.

CASE 3 Cerebellar abscess

A 30-year-old female patient with a history of left chronic otitis media, presented in ENT for left fetid otorrhea, cerebellar syndrome and intracranial hypertension syndrome. CT has found a cerebellar abscess in contact with cholesteatomatous otitis. Under wide-spectrum antibiotherapy, the patient underwent emergency drainage of the abscess by a transmastoid approach with the removal of cholesteatoma by canal wall down. The evolution is made towards death after aggravation of the neurological state.

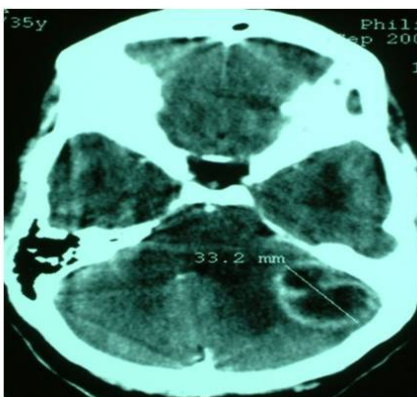
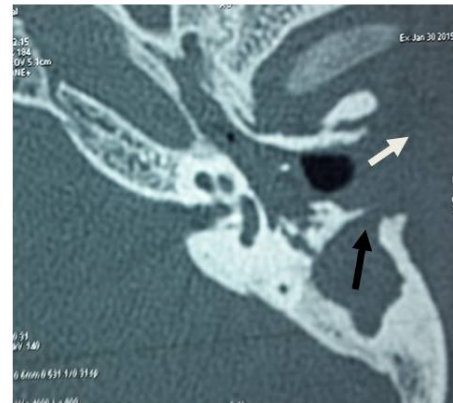


Figure 3 Axial CT revealing a cerebellar abscess Complicating left ear cholesteatoma.

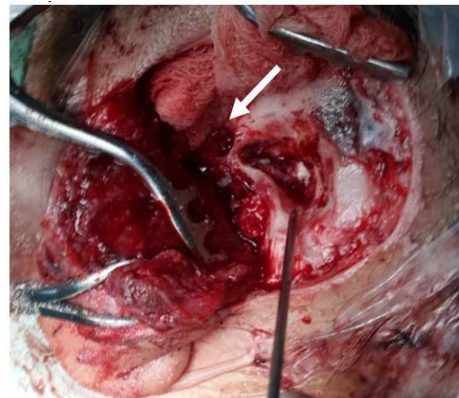
CASE 4: Temporo-zygomatic abscess

A 14-year-old mal patient, followed for poorly labeled chronic otitis, consulted in the ENT department for subcutaneous fluctuating swelling of the temporo-zygomatic region. The emergency examination revealed a painful subcutaneous collection, above the left ear lobe that reached the temporo-mandibular joint. The otoscopy diagnosed a cholesteatoma of the left middle ear. The cranial CT confirmed the destructive character of the cholesteatoma which is in continuity with the temporo-zygomatic collection (Figure4A). Under the antibiotic cover, we carried out drainage of the purulent collection, which was simultaneously associated to tympanoplasty with a canal wall up (Figure 4B).



A

Figure 4A Axial CT scan showing a voluminous antro-attico-tympanic cholesteatoma with lysis of the external attical wall (black arrow) putting the cholesteatoma in continuity with a temporo-zygomatic collection (white arrow).



B

Figure 4B The posterior approach, which allowed a tympanoplasty with canal wall up and drainage of the abscess (lysis of the external attical wall: white arrow).

CASE 5: Bezold's Abscess

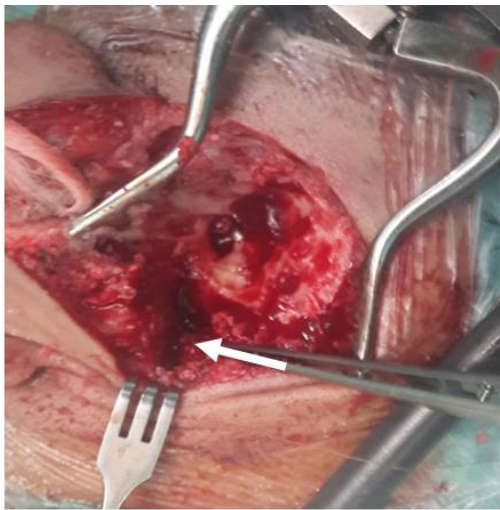
A 15-year-old male patient with a history of foul otorrhea neglected and poorly treated. It was received in ENT department for a small swelling under the left ear region, painful with fever at 39 °, limitation of the mouth opening and trismus. The auricular examination revealed a soft and fluctuating swelling with a painful mastoid on palpation. CT scan of the ear showed a left antro-attico-tympanic filling with destruction of the intercellular partitions of the mastoid and rupture of its internal and anterior cortical (Figure5A). This cortical rupture has placed the mastoid cavities in continuity with a deep cervical collection evoking an abscess of Bezold.

Under broad spectrum antibiotherapy (3rd-generation cephalosporins and metronidazole), the surgical management consisted of a performing simultaneously by a posterior approach, a drainage of the Bezold's abscess and a radical mastoidectomy (Figure5B). A good evolution without recurrence was observed with a four-year follow-up.



A

Figure 5 A Coronal CT scan showing a mastoid filling with rupture of the internal cortex (black arrow) which was complicated by a Bezold abscess (white arrow).



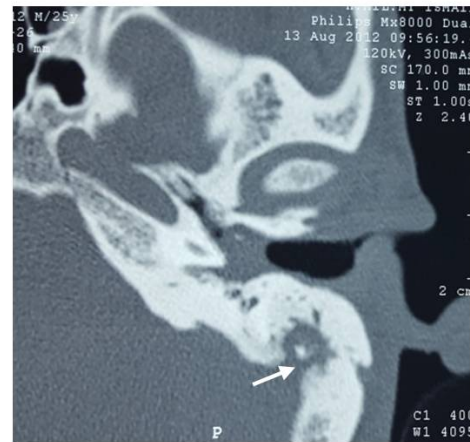
B

Figure 5 B Intraoperative view showing the seat of the Bezold's abscess (white arrow).

CASE 6: Thrombophlebitis of the lateral sinus (LS) and internal jugular vein (IJV) associated with a double abscess (cerebellum and deep cervical). (Clinical case already published in the journal research)

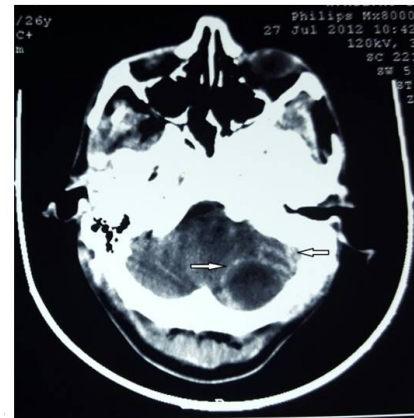
It is a 24-year-old patient who suffered from a neglected and poorly treated otorrhea. The patient was hospitalized in our department through emergencies for a left lateral swelling of the neck, renitent, voluminous and febrile. During his hospitalization, he developed a febrile meningeal syndrome with obtundation. A cranial CT, which was then supplemented with cranio-cervical MRI, concluded that cholesteatoma of the middle ear cavities was diagnosed with complete lysis of the internal mastoid cortex in contact with the LS. The cholesteatoma was directly in contact with a LST; the latter is in continuity above by a cerebellar abscess and below by a deep cervical abscess (Figure6). Therapeutic

management consisted, under broad-spectrum antibiotics, of simultaneous drainage-evacuation of cerebellar and cervical abscesses. The resection of the cholesteatoma was carried out two weeks later by tympanoplasty with canal wall down. Post-operative follow-up revealed no recurrence with a follow-up of five years.



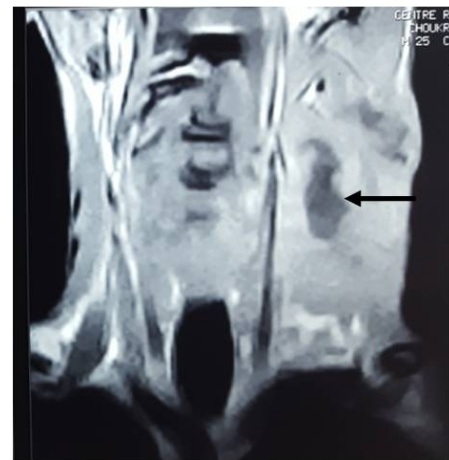
A

Figure 6 A Axial CT showing an antro-attico-tympanic cholesteatoma with complete erosion of the sinus plate (white arrow).



B

Figure 6 B Axial CT showing cholesteatoma in contact with LST (left arrow) and cerebellar abscess (right arrow).



C

Figure 6 C Coronal Cervical MRI in T2 showing a bulky cervical abscess (black arrow) in continuity with thrombophlebitis of the lateral sinus and internal jugular vein.

CASE 7: Peripheral facial paralysis.

It is a 55-year-old patient who suffered from chronic otitis media with poor follow-up. He was received in ENT consultation for recurrent fetid otorrhea and facial asymmetry. Microscopic otoscopy confirmed the diagnosis of middle ear cholesteatoma. In addition, the facial paralysis gradually settled over a period of five months. CT revealed a filling of all cavities of the middle ear with lysis of their walls in particular a dehiscence of the second portion of the Fallope duct (Figure7). The treatment consisted of a cholesteatoma excision by tympanoplasty with canal wall up and coverage of the denuded portion of the facial canal by a fragment of the temporalis fascia. Under antibiotherapy and eight months of facial muscles re-education, the patient recovered a normal facial function.



Figure 7 Coronal CT showing cholesteatomatous filling with lysis of the second portion of the Fallope's duct (white arrow).

CASE 8: The fistula of the left lateral semicircular duct.

It is a 61-year-old patient followed for recurrent and fetid right chronic otorrhea with iterative rotatory vertigo. In otolaryngology, otoscopy was able to diagnose a left cholesteatomatous otitis. The pressure of the tragus against the external auditory meatus allowed to trigger a brief rotatory vertigo. The skull base CT scan, revealed a complete filling of the antro-attico-mastoid cavity with ossicular lysis and smoothing of the middle ear walls, in particular a lysis of the lateral semicircular duct. Treatment consisted of tympanoplasty with canal wall up which allowed the eradication of cholesteatoma and the repair of labyrinthic fistula.

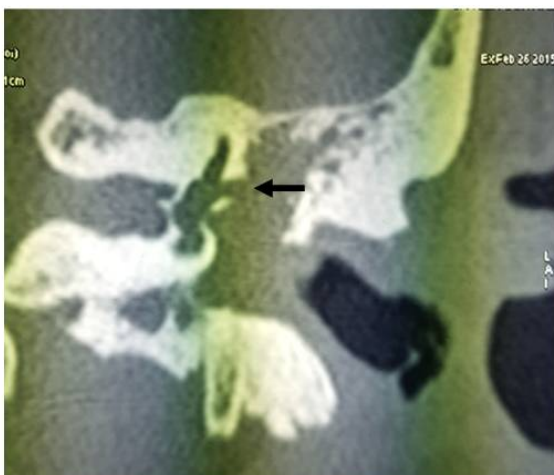


Figure 8 Coronal CT scan showing a complete cholesteatomatous filling of the middle ear cavities with a fistula of the lateral semicircular duct (black arrow).

DISCUSSION

Since the introduction of the vaccination program and antibiotics, there has been a reduction in the incidence of complications (from 10 % to 0.15 %) and mortality (from 25% to 8%) due to Chronic suppurative otitis media (CSOM)[4][5]. Despite this overall decline, the fatal complications of CSOM still persist. This is relatively common in developing countries because of poverty, ignorance, and lack of health infrastructure. In addition, the emergence of bacterial resistance to antibiotics has been implicated in some cases.

The complications of chronic middle ear otitis are related to cholesteatoma in about 36% to 84.2%. They can occur at any age but more frequently between 20 and 30 years with a male predominance [6].

The complications of cholesteatoma can be divided according to their localization into intracranial and extracranial complications.

Endocranial complications

In theory, the spread of an infection from middle ear cholesteatoma to the endocranium can be accomplished via three channels [7]: preformed, along a pre-existing anatomical pathway; neoformed, along a pathway created by the cholesteatoma; And vascular, usually venous [6].

The intracranial complications observed in this study account for 54.5% of all complications reported in this series. In half of the cases were multiple complications. Among these complications, the temporal lobe abscess (16.7%), the cerebellar abscess (33.3%), the lateral sinus thrombophlebitis (33.3%), and the subdural abscess (16.6%). Abscess occurred before the age of 30 years. There were as many women as men.

Intraparenchymal abscesses; are frequent in the development of Cholesteatoma (20% of meningoencephalitis complications) and more often temporal (75%) than cerebellar (25%, [6]. (This is the most serious complication of chronic otitis media, which requires early diagnosis to institute effective therapy.

Its frequency varies according to the authors from 17.5% (Rupa [8]) to 57.4% (Osma [9]) of intracranial complications [10]. In our series, the cerebral abcès was the first intracranial complication in order of frequency with a rate of 50%.

The bone lysis caused by cholesteatoma allows a direct diffusion of the infection through this neoformed pathway. This mechanism has been found in all cases of cerebral abscesses in this series.

Brain abscesses are located on the same side as the diseased ear which agrees with the cases of our series. The temporal lobe and cerebellum are the two locations for otogenic brain abscess.

The temporal lobe abscess usually occurs in the middle and basal portions of the temporal lobe, adherent to the dura over the roof of the petrous bone [11][12].

The treatment of choice for brain abscess is neurosurgical drainage. After draining the otogenic brain abscesses, eradication of cholesteatoma should be performed to eliminate the source of infection. The appropriate time for

cholesteatoma resection is controversial [13][12], It depends on the general condition of the patient, the existence of severity signs and the mechanism of spread of the infection. Broad-spectrum parenteral antibiotic therapy covers the surgical procedure, followed by a prolonged oral relay.

All our patients have benefited from neurosurgical drainage in emergency of the abscess, given the poor prognosis related to the cerebral engagement. The resection of the cholesteatoma made in a second time (one to two weeks after). The most frequently encountered germ found in the literature is *Proteus* followed by *Pseudomonas* [14][10].

In this study, the microorganism was isolated in two cases of intraparenchymal abscesses: *Proteus* and *Morganella morganii*.

Mortality from otogenic abscess is highly variable (8-50%), correlated with neurological status [4][15]. In our study, we found 1 case of death on the 4 cases of abscess of the brain (25%).

Lateral sinus Thrombophlebitis (LSTP)

In this series, LSTP accounts for approximately 33.3% of intracranial complications. The erosion of the sinus plate was observed in one case and both cases had the perisinus abscess. LSTP occurs most often by direct spread of infection within the mastoid. More rarely, they are extensive thrombophlebitis of the veins of the middle ear communicating with the lateral sinus. The lateral sinus thrombosis (LST) leads to venous obstruction which may extend to the cerebral veins or internal jugular vein (IJV). Suppurative thrombophlebitis may be responsible for sepsis and septic metastases [6]. The treatment is medico-surgical [16]. Medical treatment is based on antibiotic therapy, whereas anticoagulants are not recommended by most authors because the thrombus is protective against septicemia [17][18]. Surgical treatment includes the cholesteatoma removal and an attitude towards the lateral sinus which depends on its degree of occlusion evaluated by a puncture [6]. In our series we were treated by the broad spectrum probabilistic antibiotherapy without combination of anticoagulants; in all cases, the sigmoid sinus is skeletalized, surrounding granulation tissue and cholesteatoma removed. The IJV was ligated to the only patient who had a combination of IJV suppurative thrombophlebitis and a deep cervical abscess on the same side. The mortality and morbidity associated with LSTP dropped to below 10% [19] and 30% [20] respectively [21].

The subdural empyema

Subdural empyema is the rarest complication of otitis media (less than 20% of cases) [22]. In this series, there was a patient with subdural empyema (16% of intracranial complications) located in the occipital region. Before the introduction of antibiotics, subdural empyema was almost always fatal. Since then, mortality has decreased significantly, and it now ranges from 14 to 28% [12]. In rare situations when there are contraindications to surgery or significant surgical risks, conservative treatment is advised [23]. However, if a patient shows rapid neurological deterioration under medical treatment, surgery may be undertaken as a last resort [12]. In this study, the only case of subdural empyema was treated with broad-spectrum antibiotherapy. After two weeks, the patient underwent canal wall down mastoidectomy.

Extracranial complications

These are all manifestations related to the extracranial extension of infection or inflammation. In this series, extracranial complications account for 45.5% of all complications of cholesteatomatous otitis. Extracranial complications can be further divided into intratemporal and extratemporal complications.

Labyrinthine fistula

In this study, it represents 9.1% of all cholesteatoma complications and 20% of its extracranial complications. Vestibular manifestations and auditory deficit are regularly reported in the literature [10]. The petrous CT scan for cholesteatomatous otitis reveals asymptomatic fistulas [24]. The most common site of fistula was found to be the lateral semi-circular canal followed by the posterior semi-circular canal.

In the literature studies, most of the patients underwent canal wall down surgery [25][26][27].

In our series, Canal wall up procedure was undertaken to eradicate the cholesteatoma and to repair fistula.

Facial nerve paralysis

Paralysis of the facial nerve as a complication of chronic otitis media is most often secondary to the dehiscence or destruction of the bone facial canal by cholesteatoma. In our study, facial paralysis complicates 9% of cholesteatomatous otitis and accounts for 20% of extracranial complications, which is consistent with the literature data [8][10]. The lesion concerned the tympanic segment of the facial nerve. The usual mechanism of paralysis due to cholesteatoma includes direct pressure on the nerve and impaired circulation in the nerves [28]. This is the mechanism we have suspected in the case of our patient. The surgical treatment differs according to the authors, in addition to the mastoidectomy some advocate a systematic decompression of the nerve [8][29], others a simple cleaning of the tympanic cavity [30][10].

The cholesteatoma resection by Canal Wall up approach, allowed the release of the tympanic segment of the facial nerve that was covered by a temporal graft. The rehabilitation of the face's muscles made it possible to recover the nerve motor function.

Bezold's abscess

Bezold's abscess occurs rarely nowadays. In this study, they represent respectively, 9% and 20% of all complications and extracranial complications of cholesteatoma. It is defined as a deep purulent collection to the sterno-cleido-mastoid muscle. This form of abscess differs from the subperiosteal abscess, which arise from the erosion of the outer mastoid cortex [31]. In Bezold's abscess the pus discharge via erosion of the inner side of mastoid process which then spread in the neck along the fascia planes of the digastrics or sterno-cleido-mastoid muscle. As in the case of our series, a well pneumatized mastoid, with thin intercellular septums rapidly destroyed by the cholesteatoma, facilitates diffusion of the infectious process [32]. The clinical symptoms are variable and mainly include pyrexia, swelling and neck pain, otorrhea, restriction of neck movements. Sometimes when the collection fills the parapharyngeal space as the case of our patient, a trismus with limitation of the mouth opening is added. Therapeutic

management is based on adequate broad-spectrum and prolonged antibiotic therapy covering radical mastoidectomy [31][32]. Our patient was treated by radical mastoidectomy under dual antibiotherapy (Ceftriaxone and metronidazole for 5 weeks).

Temporo-zygomatic abscess

The temporo-zygomatic abscess is the rarest mastoid abscess. In this study, the temporo-zygomatic abscess represents, respectively, 9.1% and 20% of all complications and extracranial complications of cholesteatoma. When the pneumatic cells of the zygomatic root and/or the foot of the temporal bone squama are well developed, attic cholesteatoma can easily erode the well-pneumatic outer wall of the attic and reach the temporo-zygomatic space above the auricle [33][34]. Then the infection will diffuse to the extra-auricular region at the deep side of the temporal muscle, through the orifice created by the cholesteatoma. It is the same mechanism by neoformed pathway that was found in our patient. Broad spectrum antibiotherapy and sufficiently prolonged, frames the surgical act. A supra- and retro-auricular incision allowed the drainage of the abscess with resection of the infected and necrotic tissues; intervention was continued by a tympanoplasty with canal wall up [33][34].

Deep neck abscess

Deep neck abscess is a rare complication of middle ear cholesteatoma in the era of antibiotics. Its physiopathological mechanism is different from that of Bezold's abscess. The latter is a subperiosteal abscess that diffuses into the neck through the erosion of the internal mastoid cortex [35][36]. On the other hand the deep neck abscess is linked to a thrombophlebitis of the IJV [37]. Radiological examinations are complementary for a detailed demonstration of the origin and extent of the disease. The eighth case of this study was hospitalized for deep neck abscess in a context of cholesteatomatous otitis, the imaging evaluation led to the diagnosis of a cerebellar abscess and a thrombophlebitis (of lateral sinus and IJV).

Segmental mural lysis of the infected internal jugular vein was considered to be responsible for the development of deep neck abscess. [37] In peroperative, the IJV had an infiltrated, necrotic and thickened wall with a virtual lumen in its cervical trajectory.

Under broad spectrum antibiotherapy; the surgery is carried out by a multidisciplinary team, purulent collections are drained urgently and simultaneously, however, the timing of tympanoplasty with canal wall down depends on the mechanism, the condition of the patient and the coexistence of multiple complications.

CONCLUSION

Studies of cholesteatomatous chronic otitis media complications are currently rare. This small series, like the other studies of literature, separate these complications into two groups: intracranial which remain classical with still existing morbidity and mortality; And extracranial with a microbial diffusion mechanism that reveals other little known entities. The latter are essentially temporo-zygomatic and deep cervical abscesses. These two complications which characterize this study can inaugurate the diagnosis of an unknown cholesteatoma. Therefore, clinicians should be

aware of this possibility and maintain a high index of suspicion to avoid delays in diagnostic and therapeutic management in order to prevent a fatal outcome.

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