

Case Report**Subperiosteal and Citelli abscesses as rare complications of chronic suppurative otitis media****Edi Handoko, Selina Hans Sunanto**Department of Otorhinolaryngology-Head and Neck Surgery, Faculty of Medicine,
Brawijaya University/ Dr. Saiful Anwar General Hospital, Malang**ABSTRACT**

Background: Citelli abscess, is a rare extracranial complication of malignant chronic suppurative otitis media (CSOM), constitutes of a subperiosteal abscess extending from the mastoid medial aspect into the digastric fossa. Its infrequency often leads to underreporting, and delayed recognition can have severe consequences. **Purpose:** To present a case of malignant CSOM complicated by subperiosteal and Citelli abscesses. **Case report:** A 31-year-old male was diagnosed with left ear CSOM, with subsequent extracranial complications. Computed Tomography (CT) of the head and neck revealed cholesteatoma-associated ossicular damage, subperiosteal abscess, and Citelli abscess, extending intracranially to the extra-axial zone of the left temporal region. The patient underwent incision and drainage prior to receiving CT scan results, followed by a radical mastoidectomy and broad-spectrum antibiotic treatment. **Clinical question:** "In patients with CSOM, how does the prevalence of extracranial complications compared to intracranial complications in terms of frequency and associated clinical outcomes?" **Method:** A systematic search of PubMed, ProQuest, EBSCOhost, and Medline identified studies on extracranial and intracranial complications of CSOM published between 2019 and 2024. **Result:** Out of identified 84 articles, 6 met the inclusion criteria. Extracranial complications were more common, though regional variations emphasized the need for vigilance regarding intracranial complications due its severity. **Conclusion:** Subperiosteal abscesses, including Citelli abscesses, are rare but serious extracranial complications of CSOM. Prompt surgical intervention and targeted antibiotic therapy are crucial for favorable outcomes.

Keywords: subperiosteal abscess, Citelli abscess, subperiosteal, mastoiditis, CSOM**ABSTRAK**

Latar belakang: Abses Citelli merupakan komplikasi ekstrakranial yang jarang terjadi pada otitis media supuratif kronik (OMSK) maligna. Kondisi ini menggambarkan abses subperiosteal yang meluas dari aspek medial mastoid ke dalam fossa digastrik. Karena jarang kasusnya, komplikasi ini sering kali tidak dilaporkan, dan keterlambatan diagnosis dapat menyebabkan dampak yang serius. **Tujuan:** Melaporkan suatu kasus OMSK maligna yang disertai komplikasi berupa abses subperiosteal dan abses Citelli. **Laporan kasus:** Seorang pria berusia 31 tahun didiagnosis dengan OMSK pada telinga kiri yang berkembang menjadi komplikasi ekstrakranial. Hasil pemeriksaan Computed Tomography (CT) kepala dan leher menunjukkan kerusakan osikular akibat kolesteatoma, abses subperiosteal dan abses Citelli yang meluas di intrakranial ke area ekstra-aksial di regio temporal kiri. Pasien menjalani insisi dan drainase sebelum hasil CT scan diterima, diikuti dengan operasi mastoidotomi radikal dan pengobatan antibiotik spektrum luas. **Pertanyaan klinis:** Pada pasien dengan OMSK, bagaimana perbandingan prevalensi komplikasi ekstrakranial dengan komplikasi intrakranial dalam hal frekuensi dan hasil pengobatan? **Metode:** Pencarian sistematis dilakukan di PubMed, ProQuest, EBSCOhost, dan Medline untuk menemukan studi tentang komplikasi ekstrakranial dan intrakranial OMSK, yang diterbitkan antara tahun 2019 hingga 2024. **Hasil:** Dari 84 artikel yang ditemukan, 6 memenuhi kriteria inklusi. Komplikasi ekstrakranial lebih umum ditemukan, meskipun terdapat variasi regional yang menekankan pentingnya kewaspadaan terhadap komplikasi intrakranial karena tingkat keparahannya. **Kesimpulan:** Abses subperiosteal, termasuk abses Citelli, adalah komplikasi ekstrakranial OMSK yang jarang namun serius. Intervensi bedah yang cepat dan terapi antibiotik yang tepat, sangat penting untuk hasil klinis yang memuaskan.

Kata kunci: abses subperiosteal, abses Citelli, subperiosteal, mastoiditis, OMSK.

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INTRODUCTION

Chronic Suppurative Otitis Media (CSOM) is a chronic middle ear inflammation characterized by the persistent perforation of the tympanic membrane and the discharge of purulent fluid, lasting at least six weeks.¹ The ear discharge can either be continuous or intermittent. In many cases, CSOM is associated with varying degrees of hearing impairment.^{2,3}

Around 31 million CSOM cases are reported annually, with an overall incidence rate of 4.76%. Epidemiological assessments revealed that low-income and low-resource setting countries contribute over 90% of CSOM cases, including regions like Southeast Asia, the Western Pacific, and Africa.^{4,5} Factors such as densely populated living conditions, limited healthcare access, inadequate availability of antibiotics, poor hygiene, and inadequate nutrition significantly influence the high incidence rates.⁶

CSOM can be broadly classified into two types, benign and malignant. Benign CSOM involves inflammation confined to the mucosa, with a centrally located perforation that generally avoids affecting the bone. In contrast, malignant CSOM is characterized by perforation at the margins or attic, often accompanied by the presence of cholesteatoma.^{2,7}

Delayed management, particularly in cases of malignant CSOM, can lead to severe complications. However, with the availability of empirical antibiotics, complications of CSOM are now rare. Nonetheless, in developing countries, accurate reporting of incidence rates may be lacking.⁸

Complications in CSOM arise due to the spread of inflammation beyond the middle ear mucosa and the pneumatization of the temporal bone. Based on their distribution, complications are classified into intracranial and extracranial categories. Extracranial complications include petrositis, labyrinthitis, facial nerve paralysis, and subperiosteal abscesses.^{8,9}

Various abscesses can occur as complications of CSOM due to mastoid inflammation. Subperiosteal abscesses occur when the temporal bone undergoes erosion, accumulating pus beneath the skin and periosteum above the cortex of the mastoid in the region of MacEwen's triangle. Additionally, subperiosteal abscesses can extend through the medial aspect of the mastoid cavity into the digastric fossa, representing an expansion of the mastoid infection.^{10,11}

The localization of the inflow of pus in mastoiditis depends on the extent of the pneumatization. Some abscesses associated with mastoiditis are retro-auricular, zygomaticus, Bezold, Luc, Citelli, and occipital abscess.¹²⁻¹⁵ Citelli abscess is an extratemporal complication that occurs when pus from the mastoid apex drips downward along the posterior digastric muscle to the occipital and cervical regions.¹⁶

In cases of acute mastoiditis with swelling around the ear, the primary step of medical management involves draining the pus from the swelling, followed by mastoidectomy. Antibiotics are prescribed for two weeks. Additional treatments include administering analgesics, antipyretics, and topical antibiotic/steroid combinations.¹⁶

This case report presented the case of a 31-year-old male patient diagnosed with malignant CSOM, experiencing extracranial complications, including subperiosteal and Citelli abscess extending into the left temporal region. This comprehensive report underscored the significance of early diagnosis and effective management of CSOM and the understanding of its potential complications.

CASE REPORT

A 31-year-old male was presented with a chief complaint of pain behind and below the left ear, worsening for two weeks before seeking medical treatment at the hospital. He had felt this discomfort since three months previously, accompanied by the appearance of a lump behind the left ear. The lump had grown downward, extending beneath the ear and spreading to the posterior neck.

The patient reported recurrent ear discharge from the same ear for the past ten years. The discharge was yellow, thick, sticky, and foul-smelling. He also experienced progressive hearing loss in the left ear over the past three months. Nausea, vomiting, headaches, and vertigo were denied. Nasal and throat complaints were also absent. The patient had never sought care from a specialist before; he had been treated by traditional healers and general practitioners, and had been prescribed ear drops.

On physical examination, the patient appeared in good general condition and was alert. The axillary body temperature was measured at 37.8°C, while other vital signs were within normal limits. An otoscopic examination of the left ear revealed edema, erythema of the ear canal, and bulging due to pressure from the retroauricular area. The tympanic membrane of the left ear was perforated by 50%, and there was no light reflex. The right ear showed no abnormalities, with a clear ear canal, no discharge or wax, and an intact tympanic membrane. The left

retroauricular area showed swelling, redness, warmth, tenderness, fluctuation, and positive induration. The inferior region of the left auricle, the left neck, and the posterior neck displayed swelling, redness, tenderness, warm on palpation, fluctuation, and positive induration.



Figure 1. A 31-year-old patient with subperiosteal abscess.

The image showed a reddish lump under the left ear extending to the retroauricular and back of the neck (red arrow and circle).

A puncture was performed in the left retro auricular area and the posterior neck, yielding approximately 3 ml of pus mixed with blood. Laboratory examination revealed an elevated white blood cell count (22,680/ μ L). Cervical plain radiography showed a soft-tissue mass in the left middle-posterior side of the neck region, without accompanying airway narrowing. An incision and drainage of the abscess were carried out on the patient under general anesthesia. Computed Tomography (CT) scan of the head and neck was conducted after the incision, both with and without contrast, revealing extension into the intracranial extra-axial region of the left temporal region.



Figure 2. Anteroposterior and lateral cervical radiographs

High-resolution CT (HRCT) findings were consistent with left-sided osteomastoiditis extending into the infratemporal fossa and involving the surrounding muscles, extending further into the intracranial extra-axial region of the left temporal region.



Figure 3. Mastoid HRCT results

An isodense lesion was seen on the left mastoid air cells, with a defect in the pars mastoid os temporalis on the posteroinferior side, ± 4.5 mm wide. It extended to the infratemporal fossa, and involved left sternocleidomastoid, left levator scapulae, left longissimus capitis, left semispinalis capitis, splenius capitis, scalenus medius, left longus capitis, and left digastric muscle. The lesion extended to the intracranial extra-axial left temporal region, through a defect in the pars mastoid of temporal bone ± 3.7 cm wide.

Based on the history, physical examination, and diagnostic tests, the patient was diagnosed with left-side malignant CSOM, and complications of subperiosteal abscess and Citelli abscess. Post HRCT results, a radical mastoidectomy was performed.

During the radical mastoidectomy surgery, cholesteatoma was found in the mastoid antrum and the tympanic cavity; with destruction of the mastoid tip and involvement up to the level of the digastric muscle. Granulation tissue was observed in the attic space. The auditory bones were found to be surrounded by cholesteatoma mass.

The patient was treated with intravenous ampicillin-sulbactam 4 times a day at 1.5 grams, and metronidazole 3 times a day at 500 mg for 5 days. The patient's condition remained stable after the operation, and further management awaited the results of the abscess base swab and histopathology of the tissue.

CLINICAL QUESTION

Based on the case above, the clinical question formulated was, "In patients with Chronic Suppurative Otitis Media (CSOM), how does the prevalence of extracranial complications compare to intracranial complications in terms of frequency and associated clinical outcomes?". From this clinical question, the PICO (Patient/population/problem, intervention, comparison, outcome) was as follows:

P : Patients with Chronic Suppurative Otitis Media (CSOM).

I : Analysis of extracranial complications.

C : Analysis of intracranial complications.

O : Frequency of complications.

METHOD

A systematic search was conducted using multiple databases, including PubMed, ProQuest, EBSCOhost, and Medline, focusing on articles published within the last five years (2019–2024). No language restrictions were applied, and articles published in languages

other than English were translated using online translation tools. To ensure a comprehensive review, the reference lists of selected studies were examined, and preprint data were manually searched to supplement database results. Eligible studies included cohort, case-control, or cross-sectional designs that analyzed extracranial and intracranial complications of Chronic Suppurative Otitis Media (CSOM). The search utilized the keywords “extracranial,” “intracranial,” “complication,” and “chronic suppurative otitis media.” Studies excluded from the review included duplicates, scoping reviews, systematic reviews, narrative reviews, and conference abstracts.

The selection process involved independent screening of titles and abstracts by the authors based on predefined criteria. Full texts of relevant articles were subsequently reviewed to confirm eligibility. Studies that did not meet the inclusion criteria were excluded. Data extraction was carried out using Microsoft Excel and included information such as author, year, location, study design, population, age range, number of cases/patients, and primary outcomes. The risk of bias was independently assessed by researchers using standardized tools.

RESULT

Initially, 84 articles were identified, with 46 duplicates removed. Following a title screening, 20 articles were excluded. Subsequently, 18 full-text articles were evaluated for eligibility, and 6 were found to meet the inclusion criteria.

Table 1 provided an overview of six studies investigating complications of CSOM conducted in various countries, including India, Nepal, Pakistan, Egypt, and Nigeria, with patient ages spanning from 1 to 89 years. Five of the studies employed a cross-sectional design, while one was a prospective study. Most studies identified extracranial complications as more common than intracranial ones. However, an exception was observed in the study by Umar et al.¹⁷ in Nigeria, which reported a higher number of intracranial complications (32) compared to extracranial ones (22). While extracranial complications were generally more prevalent, the findings highlight regional differences and emphasize the need to remain vigilant about intracranial complications due to their severity and associated risks.

Table 1. Characteristics of included studies

Author, Year	Country	Study design	Population	Age	Number of Patient	Intervention	Comparison
Parmar et al. (2020)	India	Cross-sectional	All patients of CSOM with intra or extracranial complication	4-52 years	36 patients with 61 complications	44 extracranial complications	17 intracranial complications
Dongol et al. (2020)	Nepal	Cross-sectional	Patients of all age groups and genders with a complication of otitis media were included in the study	1-89 years	164 patients with	155 extracranial complications	27 intracranial complications

Shah et al. (2021)	India	Prospective study	All the consenting patients with complicated CSOM were included.	4-52 years	50 patients	30 extracranial complications	20 intracranial complications
Haqdad et al. (2022)	Pakistan	Cross-sectional	All patients of chronic suppurative otitis media with intra or extra-cranial complications	5-58 years	42 patients	23 extracranial complications	19 intracranial complications
Hamed et al. (2023)	Egypt	Cross-sectional	All patients of chronic suppurative otitis media with intra or extra-cranial complications	1-75 years	53 patients	47 extracranial	4 intracranial
Umar et al. (2024)	Nigeria	Cross-sectional	Patients who were diagnosed as having CSOM with complications	3-50 years	46 patients with 54 complications	22 extracranial	32 intracranial

DISCUSSION

A case of a 31-year-old male with a chief complaint of pain behind and below the left ear, which worsened two weeks prior to seeking medical attention at the hospital. The complaint was accompanied by a lump behind the left ear, which enlarged up to the lower ear, and extended into the back of the neck. The patient complained of recurrent ear discharge for the past ten years, with secretions being yellow, viscous, sticky, and putrid from the left ear. A decreased hearing in the left ear was also experienced and had been worsening over time. The patient's symptoms led to a diagnosis of longstanding CSOM with complications. CSOM is a chronic middle ear inflammation with perforation of the tympanic membrane and a history of secretion drainage (otorrhea) lasting more than two months, whether continuous or intermittent. The discharge may be thin or thick, clear or pus-filled, and can be accompanied by varying degrees of hearing impairment.^{4,9,18}

Several episodes of acute otitis media often precede CSOM, which develops when the natural repair mechanism of the tympanic membrane fails to heal the perforation completely. After CSOM develops in childhood, it can persist into adulthood. This is the primary cause of recurrent episodes of fluid drainage from the ear unless the tympanic membrane is repaired through surgery.¹⁹ Therefore, adult patients as the one in this case report, typically had a long history extending over several years or even decades.^{20,21}

Factors contributing to the high proportion of CSOM cases include population density, malnutrition, poor hygiene, colonization of potentially pathogenic microorganisms in the nose and nasopharynx, and lack of access to standard healthcare facilities.^{5,6}

Generally, CSOM is classified into benign and malignant types. Benign CSOM, also known as the mucosal or safe type in international literature, is characterized by

its limited mucosa involvement, absence of cholesteatoma, and rare complications. On the other hand, malignant CSOM, also referred to as the epithelial or unsafe type, typically leads to serious complications.¹⁰ In this case report, the patient experienced malignant CSOM with a subperiosteal abscess complication, which occurred due to erosion of the temporal bone and the accumulation of pus beneath the skin and periosteum above the cortical bone in the MacEwen's trigonum area.

In CSOM, the causative bacteria can be aerobic (e.g. *Pseudomonas aeruginosa*, *Escherichia coli*, *S. aureus*, *Streptococcus pyogenes*, *Proteus mirabilis*, *Klebsiella species*) or anaerobic (e.g. *Bacteroides*, *Peptostreptococcus*, *Propionibacterium*).¹⁸ Bacteria are rarely found in the skin of the outer ear canal, but can reproduce in the presence of trauma, inflammation, lacerations, or high humidity. These bacteria can enter into the middle ear through chronic perforations. Among these bacteria, *P. aeruginosa* is specifically accountable for the profound and progressive destruction of the middle ear and mastoid structures through its toxins and enzymes.¹⁰

Complications of CSOM can be both intra- and extracranial.⁹ In this patient, an extracranial complication was found as a subperiosteal (Citelli abscess), extending into the left temporal region's intracranial extra-axial region. This specific extracranial complication, especially subperiosteal abscess, is rare and is often accompanied by other otitis media complications.^{22,23}

Subperiosteal abscesses can result from complications of both acute and chronic otitis media. Subperiosteal complications due to chronic otitis media are often caused by the blockage of the aditus and antrum by granulation tissue and cholesteatoma. This blockage hinders the flow of pus from the mastoid cavity to the tympanic cavity. The accumulation of pus increases the pressure within the mastoid cavity. This increased

pressure triggers the destruction of the lateral wall of the mastoid cortex (MacEwen's trigonum), which had previously been eroded by cholesteatoma or granulation tissue. Pus is forced out of the mastoid cavity through the destroyed lateral wall of the mastoid cortex into the periosteum, forming a subperiosteal abscess.^{10,24}

Citelli abscess is a rare complication of CSOM. The current incidence rate is difficult to determine; older literature with limited samples reported a Citelli abscess occurrence rate of <1%.⁹ Most case reports had been published in developed countries, with low CSOM complications due to medical advancements. However, reports from low-income countries are difficult to find, possibly leading to underreporting in these nations.^{14,15}

Subperiosteal abscess and Citelli abscess are rare in the era of available antibiotics. Delayed treatment of CSOM complications may be associated with more significant morbidity. Early recognition and treatment are essential. The classic features of a neck space abscess include painful neck swelling and reduced neck movement.¹⁵ In this patient, swelling and pain in the neck are suspected of suggesting a deep neck abscess.

Citelli's abscess was first described in 1901 by Salvatore Citelli. Citelli's abscess is an extratemporal complication of otitis media. It occurs when pus from the mastoid tip trickles down along the posterior belly of the digastric muscle to the occipital and cervical region. This abscess is located posterior to the mastoid process between the mastoid and occipital bones. Another hypothesis of the Citelli abscess involves the route of the mastoid vein, or the route of the occipitomastoid suture.¹⁶

Gravity causes the abscess to spread from the mastoid process either inferiorly or posteriorly along the sternocleidomastoid muscle or digastric. In addition, the space between the two muscles contains the

attachments of the splenius and longissimus capitis muscles, which may contribute to the spread of the abscess along these structures.²⁵

Currently, mortality and morbidity due to CSOM complications have decreased dramatically due to the discovery of modern antibiotic therapy. Still, with the increasing number of pathogens resistant to various drugs, the incidence of complications is increasing again and needs to be watched out for. In a retrospective study conducted over 22 years (1987-2008) by Wu JF et al.²⁶ in China, 285 (12.1%) patients with acute and chronic otitis media with and without cholesteatoma developed intracranial and extracranial complications. The most extracranial complications were labyrinthitis (90 cases), mastoid abscess (79 cases), facial paralysis (47 cases), Bezold's abscess (5 cases), and apicitis pyramidalis (1 case).

The diagnosis of CSOM complications such as subperiosteal abscess is established based on physical examination and supporting examinations. CT scan of the head and neck is an important modality, as it has been shown to detect deep abscesses that are not seen clinically. Subperiosteal abscess and Citelli abscess usually complicate the course of chronic otomastoiditis. This type of abscess accounts for less than 6% of otogenic abscesses. In clinical suspicion of subperiosteal abscess, temporal bone CT and contrast-enhanced CT of the neck are the gold standards for determining two main diagnostic aspects: (1) the site of erosion of the mastoid bone in the context of mastoiditis, and (2) the anatomical boundaries of the neck suppurative collection. CT of the temporal bone usually shows signs of mastoiditis, such as mastoid cell opacities and erosion of the mastoid bone trabeculae. High Resolution (slice thickness <1 mm), neck CT Acquisition Reconstruction coronal plane, high-frequency bone kernel, and bone window are suggested to allow the detection

of minor bone interruption at the end of the mastoid, commonly in the digastric groove.²⁴

If a subperiosteal abscess is present or suspected, broad-spectrum antibiotic therapy with good CSF (cerebrospinal fluid) penetration should be initiated, and appropriate imaging should be performed to evaluate the location and size of the abscess collection. Given the incidence of polymicrobial infections, routine empiric broad-spectrum antibiotic therapy should cover most Gram-positive and Gram-negative aerobic and anaerobic pathogens. Early surgery is often mandatory to establish middle ear and mastoid drainage (also through myringotomy). Sampling of purulent material needs to be done.⁵

Pathogen test results make it possible to replace as soon as possible the broad-spectrum antibiotic that was first administered with an antibiotic to which the pathogen is susceptible. In conditions where a collection of deep neck fluid is present with coalescent mastoiditis, a post-auricular incision is made, and a complete mastoidectomy should be performed in addition to drainage of the deep neck abscess via a transcervical approach. After surgical drainage of a deep neck abscess collection, contrast-enhanced imaging control is recommended 48-72 hours before removal of the suction tube.^{13,27}

In conclusion, Citelli's abscess is rare but significant extracranial-extratemporal complication of otitis media. It occurs when pus from the mastoid tip trickles down along the posterior belly of the digastric muscle to the occipital and cervical region. This report detailed a case of a 31-year-old male diagnosed with a Citelli abscess. Timely surgical intervention, combined with targeted antibiotic therapy proved essential for achieving a positive outcome.

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