

# Multiple sialoceles of the parotid gland in chronic parotid sialadenitis – a case report

# Mnogie sialocele przyusznicy w przebiegu przewlekłego zapalenia – opis przypadku

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ABSTRACT:	<b>Introduction:</b> Chronic parotid sialadenitis is a disorder of multifactorial etiology. The main cause of this condition is usually a presence of deposits that narrow the parotid duct lumen. Obturative lesions and ongoing inflammation may lead to the development of retention cysts.
	<b>Case report:</b> In the reported case, a 56-year-old patient experienced a massive polycystic hyperplasia of the parotid gland secondary to chronic calculous sialadenitis, with total parotidectomy as the only effective method of treatment. The methods of treatment and procedural models in the management of chronic parotid sialadenitis are discussed.
KEYWORDS:	chronic parotid sialoadenitis, parotid gland, parotid lithiasis
STRESZCZENIE:	Wstęp: Przewlekłe zapalenie ślinianki przyusznej jest chorobą o etiologii wieloczynnikowej. Przyczynę stanowi najczęściej zwężenie światła przewodów wyprowadzających przez zalegający złóg. Na tle zmian obturacyjnych oraz toczącego się proce- su zapalnego może dochodzić do powstawania torbieli retencyjnych.
	<b>Opis przypadku:</b> W opisanym przypadku u 56-letniego pacjenta doszło do masywnego wielotorbielowatego rozrostu ślinian- ki na tle kamiczego przewlekłego zapalenia, w którym jedyną skuteczną metodą leczenia była parotidektomia całkowita. W niniejszej pracy omówiono metody leczenia i model postępowania w przewlekłym zapaleniu przyusznicy.
SŁOWA KLUCZOWE:	kamica ślinianki przyusznej, przewlekłe zapalenie ślinianki przyusznej, ślinianka przyuszna

# INTRODUCTION

Chronic parotid sialadenitis in adults involves inflammation and stenosis of parotid ducts leading to recurrent swelling and periodic purulent discharge from the ostium of the parotid duct [1]. According to the reports, the main causes include local duct obturation resulting from the presence of deposits, anatomical pathologies of the parotid ducts, ostial stenosis, foreign bodies, or external impingement. Other reasons include the impairment of the parotid duct function secondary to radio- and iodotherapy and autoimmune processes (e.g. the natural history of Sjögren's syndrome). Obturative lesions and inflammation lead to further stenosis and development of retention cysts. The reported case involved chronic calculous sialadenitis leading to hypertrophy of the parotid gland and multiple sialoceles.

# **CASE REPORT**

A 58-year-old male patient had presented at the emergency room in September 2016 with swelling and inflammation of the right

lation to the diameter of 24 mm and shadowing deposit sized ca.  $12 \times 5$  mm located at the ostium. Following the failure of conservative treatment involving antibiotic therapy combined with massage and administration of relaxants, the patient was hospitalized in October 2016. The ostial deposit was removed from transoral access. The patient experienced resolution of symptoms following surgery; follow-up ultrasound scans revealed regression of Stensen's duct dilatation to 18 mm in January 2017 and 6.5 mm in November 2017.

parotid gland. The ultrasound scan visualised Stensen's duct di-

In late 2018, the patient returned to the Clinic due to parotid swelling which had been worsening for about two months. Ultrasound scan revealed numerous, significantly dilated, sinuous parotid ducts with diameters of up to 30 mm. No shading deposit was visualized. Computed tomography scan revealed numerous cystically dilated ducts and cystic lesions up to 32 m wide within both lobes of the right parotid gland (Fig. 1.). On 11.01.2019, an attempt was made to dilate the right parotid duct in order to make the connection with the cyst. Despite the widest probe being installed,

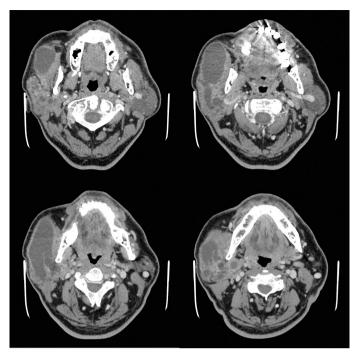


Fig. 1. CT scan of the head: Transverse cross-sections reveal multiple sialoceles within the right parotid gland.

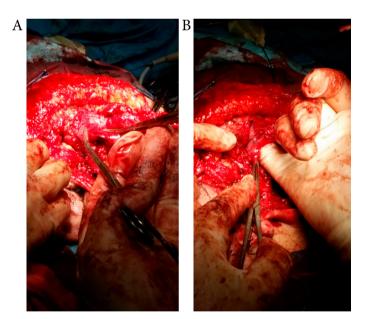


Fig. 2. Dilated Stensen's duct - intraoperative image.

no communication could be obtained with the duct and its contents could not be evacuated. No consent for radical surgical treatment was obtained from the patient at that time.

Two months later (in March 2019), the patient returned to the Clinic and expressed his willingness to undergo treatment. Examination upon admission revealed a massive, swollen, tuberous right parotid gland which was not tender upon palpation and from which purulent secretion was periodically discharged into the mouth.

On 14.03.2019, the patient was subjected to total right-sided parotidectomy. Hypertrophic parotid gland tissue was observed



Fig. 3. Dissected facial nerve trunk and branches – intraoperative image.

intraoperatively, with markedly dilated salivary ducts discharging large quantities of seromucous contents (Fig. 2.). The trunk and the main branches of the facial nerve were located and dissected – the nerve branches were very thin and compressed by the significantly hypertrophic parotid parenchyma. The nerves were dissected while maintaining their continuity (Fig. 3.). The significantly hypertrophic superficial part of the parotid gland was removed, followed by the deep part with significantly hypertrophic inferior pole penetrating into the parapharyngeal space. Massively dilated Stensen's duct, ca. 1.5 cm in diameter, was visualized intraoperatively. The duct was dissected, cut at the parotid papilla, and sealed shut.

The resected parotid gland presented as a seven-piece specimen with total dimensions of  $7.5 \times 7 \times 4$  cm (Fig. 4.). Within the parenchyma, numerous cysts and six reactively changed lymph nodes sized 0.5 to 0.7 cm were identified. Microscopic examination revealed parenchymal atrophy with chronic inflammation and fibrosis, particularly in periductal locations.

Following the procedure, House-Brackmann grade IV/V right facial nerve paresis was observed in the patient. Following rehabilitation, the paresis subsided to grade II as assessed at a follow-up visit in September 2019.

#### DISCUSSION

Sialolithiasis is a common disorder of large salivary glands affecting about 1% of the general population. It is most common within the submandibular glands (80-90%), and less common within the parotid glands (5-20%). It may develop at any age, with peak incidence being observed in the fourth, the fifth, and the sixth decade of life. Male patients are affected more frequently [3]. Numerous theories had been developed regarding the pathogenesis of calculi within the salivary ducts. The most likely causes include the disturbances in salivary composition leading to precipitation of mineral deposits and the formation of deposits from mucous precipitates. Other hypotheses include calcification originating around a foreign body or microbial fragments [4].

Conservative management can be considered the first line of treatment of chronic sialoadenitis. It consists of antibiotic, analgesic, and anti-inflammatory pharmacotherapy, acidic diet, salivary gland massage, and antibacterial mouth wash being used to prevent secondary ascending infection [5].

The second-line treatment consists of minimally invasive procedures such as sialoendoscopic removal of deposits or ductoplasty.

Parotidectomy is the last treatment option considered to be indicated only in most severe cases or following the failure of other treatments. Parotidectomy can be superficial, subtotal, or total [6].

In 2003, Motamed et al. [7] analyzed distant outcomes of parotidectomy as the treatment of chronic parotid sialadenitis. The analysis of complications and efficacy revealed that disease recurred only in patients subjected to superficial parotidectomy.

In 2019, Rik et al. published a retrospective study of 46 parotidectomies performed in 1999-2012 in patients with chronic parotid sialadenitis. The treatment efficacy rate was 87%. Facial nerve paresis was observed in 12% of patients [8].

One should keep in mind that parotidectomy in chronic parotid sialadenitis poses a significant challenge to the operating surgeon due to chronic inflammation and consequential fibrosis. It is believed that complete remission requires that the parotid gland tissue is excised to the largest extent possible while preserving the continuity of the facial nerve [9].

## **SUMMARY**

Total parotidectomy is considered the last resort in the management of calculous parotid sialadenitis while remaining the only efficacious treatment in justified cases. In the reported case, the conservative treatment followed by minimally invasive procedural treatment

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Fig. 4. Specimen sent for histopathological examination; total dimensions 7.5 × 7 × 4 cm.

involving ductoplasty with transoral extraction had failed. The patient developed irreversible changes in the structure of parotid parenchyma and ducts, leading to a giant-sized gland with numerous sialoceles.

Due to chronic inflammation and consequential anatomical changes, the surgical treatment is very challenging for the operating surgeon. Precision and diligence while dissecting the facial nerve are paramount, and therefore surgeries should be performed at sites with considerable experience in parotid surgeries.

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