



Journal Homepage: -www.journalijar.com

INTERNATIONAL JOURNAL OF ADVANCED RESEARCH (IJAR)

Article DOI:10.21474/IJAR01/19990
DOI URL: <http://dx.doi.org/10.21474/IJAR01/19990>



RESEARCH ARTICLE

SURGICAL TREATMENT OF RARE SUBMANDIBULAR SIALOLITH OF EXUBERANT DIMENSIONS

Marianna Miranda Pereira, Fabiana Sindeaux Araujo, Karen Gonçalves da Silva, Mariana Frota de Castro, Daniely Francine Fagundes Marques, Nathanael Barbosa Oliveira, Yasmin de Medeiros França, Mariana Souto Lima, Luiz Felipe Costa de Moura, Tiago Gomes de Lucena, Gabriel Luiz Schneider De Lima, Luana Gaspar Prates, João Vitor Dias Andrade, Jessé Jarlison Morais Oliveira Bezerra, Anna Karolyne Grandó Silveira

Manuscript Info

Manuscript History

Received: 05 October 2024

Final Accepted: 07 November 2024

Published: December 2024

Key words:-

Sialolithiasis, Obstruction, Calcification,
Salivary Glands

Abstract

Sialolithiasis is characterized by the formation of stones that obstruct the salivary ducts, leading to inflammation, infection, and reduced salivary flow. Etiopathogenesis is currently unknown, but evidence suggests an interaction with the chemical composition of saliva, specifically due to the combination of various electrolytes such as sodium, potassium, calcium, and phosphates. The stones are generally unilateral, with a higher prevalence in the submandibular gland. Clinical and radiographic exams are essential for diagnosis, with CT and MRI serving as auxiliary methods in cases where stones are not visible in conventional X-rays. Treatment varies depending on the size and location of the stone, ranging from conservative measures like salivary stimulation and compresses to surgical interventions. The aim of this study was to report a rare case of an extensive sialolith with a minimally invasive approach. A female patient sought the maxillofacial surgery service reporting swelling in the floor of the mouth region and difficulty chewing. During the physical examination, hardening of anatomical structures on the right side was identified. Complementary exams, such as occlusal radiography, revealed a symmetric radiopaque area of 35.8 mm, confirming the presence of a remarkably large salivary stone in the duct of the submandibular gland. The treatment of choice was the excision of the sialolith, where tissue synthesis was performed by anatomical planes. In the postoperative period, complete restoration of salivary function and relief of initial symptoms were observed. Therefore, large stones significantly affect salivary flow and gland function, justifying surgical removal as the most appropriate treatment. The surgical technique used in this case, besides being minimally invasive, proved effective in removing the sialolith and preserving the gland's structures, resulting in satisfactory recovery and the complete restoration of the patient's salivary function.

Copyright, IJAR, 2024,. All rights reserved.

Corresponding Author: Anna Karolyne Grandó Silveira

Introduction:

Sialoliths are calcified structures located in the secretory salivary ducts or in the glandular parenchyma. They can affect major salivary glands and, in rare cases, minor salivary glands (FOLCHINI & STOLZ, 2016, KUHN-DALL MAGRO *et al.*, 2014).

The etiology is not yet fully understood; however, two main pathways are believed to contribute to its emergence: the anatomical structure of the gland and ducts and the chemical composition of saliva (AVISHAI *et al.*, 2020, LIMA *et al.*, 2013, JADU & JAN, 2014).

Sialolithiasis, in turn, is characterized as the inflammation of a salivary gland due to the obstruction of a sialolith. This is a relatively common condition and, although it is not directly related to age and gender, it is more common in men after the age of 40. The symptoms include edema, especially during meals; erythema, pain and purulent secretion originating in the duct. Depending on the severity of the obstruction or the size of the calcified structure, some patients may present hyperemia, malaise and localized lymphadenopathy (SANTANA *et al.*, 2021, SILVA *et al.*, 2021, LOMMEN *et al.*, 2021, SOUZA *et al.*, 2015).

Although the frequency of sialolithiasis is relatively high, the occurrence of giant sialoliths measuring more than 1.5 cm in any of their diameters is rare. For this reason, there are few studies published in the relevant medical literature (OLIVEIRA *et al.*, 2016, AVISHAI *et al.*, 2024).

Sialolithiasis most frequently affects the submandibular glands, because they have a more alkaline, thick and calcium-rich secretion (OLIVEIRA *et al.*, 2019, YING *et al.*, 2015, MEIRA *et al.*, 2021).

The identification can be performed on routine occlusal radiographs, presenting as a radiopaque area, although the composition of the calculus and its location may influence its radiopacity. For diagnosis confirmation, the sequence consist of a clinical examination, medical history analysis and imaging tests such as panoramic radiography, occlusal radiography, computer tomography (CT), ultrasonography and magnetic resonance imaging (MRI) (SILVEIRA JÚNIOR *et al.* 2020, MORTAZAVI *et al.*, 2024, KOCH *et al.*, 2021).

Treatment for sialolithiasis depends on the size and location of the calcification. Smaller calcifications can usually be treated with non-invasive techniques such as hydration, use of mechanical sialogogues to stimulate salivation, massage involving the gland, warm compresses, non-steroidal anti-inflammatory drugs and antibiotics. Planned surgical removal by radiological apparatus or camera-guided sialendoscopy is recommended when conservative devices are successful (ASHINDOITIANG *et al.*, 2023, BEUMER *et al.*, 2023, SODNOM-ISH *et al.*, 2024, BASRA *et al.*, 2024, CHIESA-ESTOMBA *et al.*, 2024;).

In cases of giant salivary glandular calculi (GSGC) affecting the submandibular gland, treatment consists of a surgical approach, including a complete sialoadenectomy, through a transverse access (CASTRO *et al.*, 2021, SILVA *et al.*, 2024, BEUMER *et al.*, 2024).

This article deals with a case of a rare, large sialolith and demonstrates the effectiveness of a minimally invasive technique for its removal, highlighting the recovery of salivary function.

Case Report:

A 42-year-old female patient sought the oral and maxillofacial surgery service complaining of increased sublingual volume and difficulty chewing. During the anamnesis, the patient denied having any previous illnesses. Physical examination revealed hyposialia and hardening in the sublingual space. For better investigation of the case, an occlusal radiograph was requested, which revealed a single radiopaque calculus in the region of the Wharton duct (Figure 1). A provisional diagnosis of sialolithiasis in the duct of the right submandibular gland was suggested.

In this case, being considered a GSGC, the proposed definitive treatment was minimally invasive surgical excision in order to preserve the glandular structures and salivary flow. The treatment was performed under local anesthesia with lingual nerve block, via an intraoral approach. An incision was made, and careful tissue dissection was carried out to preserve the anatomical structures, followed by complete removal of the sialolith (Figure 2). The presence of

complete mineralization facilitated the excision without intraoperative fragmentation of the sialolith (Figure 3). Finally, the tissues were sutured in anatomical layers (Figure 4).



Figure 1: Occlusal radiograph revealing a well-defined mineralized tissue, mostly radiopaque with homogeneous density, measuring approximately 35 × 8 mm.



Figure 2: Intraoperative moment of sialolith excision.

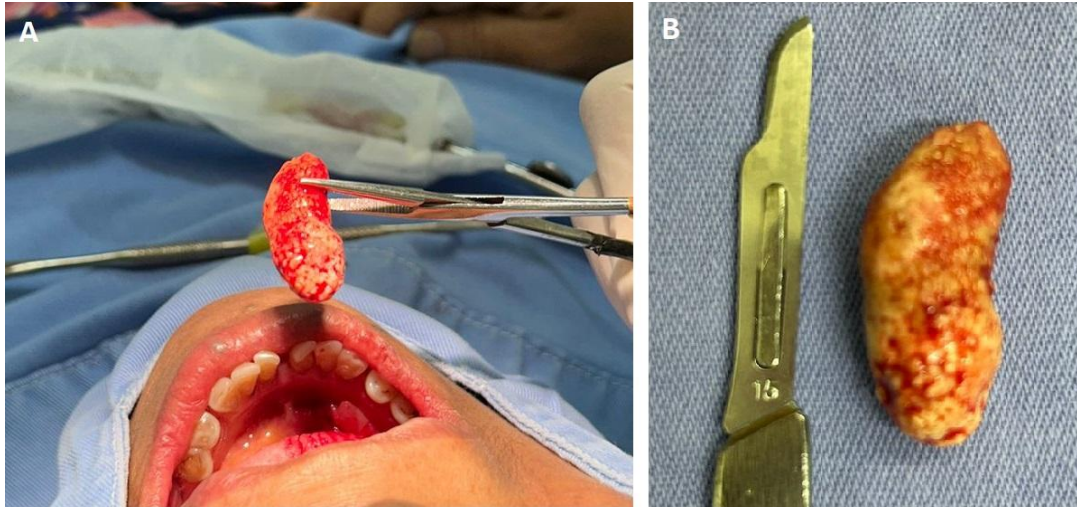


Figure 3:(A) Complete removal of the calculus. (B) Comparison of the size of the n°15 scalpel blade and the sialolith.



Figure 4:Layered closure of the surgical area.

During post-surgical monitoring, satisfactory tissue repair was observed, with a significant and gradual reduction in volume increase and discomfort. Healing occurred adequately, without the formation of adhesions or stenosis in the glandular ducts, which ensured full restoration of salivary function and complete relief of initial symptoms.

Discussion:

Sialolithiasis is the most common disease affecting the salivary glands, characterized by the presence of a stone in the gland or duct, which obstructs the ducts and results in decreased salivary flow, inflammation and infection (MOHSIN, RIYAZ, ALQAZLAN, 2022; BARBEIRO *et al.*, 2022; CASTRO *et al.*, 2021; LOMMEN, *et al.*, 2021; SODNOM-ISH *et al.*, 2023) this was the diagnosis in the present case, the sublingual swelling, difficulty in chewing, and reduced saliva production due to duct obstruction-induced were observed.

The etiology of sialolithiasis remains unclear, however, potential causes may be mechanical, traumatic, infectious, chemical, neurotic origin or some foreign body (FATIMA *et al.*, 2014). Most cases (80%) occur in the submandibular gland, as in presented case, which was diagnosed with sialolithiasis in the right submandibular gland duct. Possible factors explaining this higher prevalence include a higher calcium concentration, more alkaline pH and easier obstruction of salivary flow due to the anatomy of Wharton's duct, where secretion flows against gravity (CASTRO *et al.*, 2021; SANTANA *et al.*, 2021). A minority of cases (between 1-2%) are located in the sublingual gland and minor salivary glands, with trauma being a primary factor, followed by stone formation in the ducts (BASRA *et al.*, 2024).

According to Castro *et al.*, 2021, there is no predilection for sex or age. However, sialolithiasis is more prevalent in men aged between 30 to 40 years. This contrasts with the present case of a 42-year-old female patient.

Sialolithiasis is often asymptomatic in most patients, but in cases of total duct obstruction, symptoms may become a way noticeable, including inflammatory sign, purulent discharge, and difficulty swallowing (SANTANA *et al.*, 202; YING *et al.*, 2015). In this study, an obstruction in the duct, leading to sublingual swelling, hyposalivation and difficulty in chewing. Sialoliths are typically unilateral (affecting only one gland per time) and simple representing 75,3%, with rare cases of multiple stones. Some sialoliths are elongated and cylindrical, being found on the ducts and others with oval shape, found in the glandles (SILVA *et al.*, 2021; Basra *et al.*, 2024). In most cases, the stones reach up to 10mm and are classified as GSGC. Nevertheless, in this case, the sialolith measured approximately 35mm, a really rare size. (CASTRO *et al.*, 2021).

The stones are typically yellowish, soft or thick, and easily palpable in the peripheral portions of the salivary ducts (SANTANA *et al.*, 2021). In this study, a firm, yellowish stone was observed after the surgical procedure. Imaging exams aid in diagnosing this condition, although sialoliths may not always be visible on radiographs due to the stone's degree of mineralization, often requiring computed tomography or magnetic resonance imaging to close the diagnosis. Treatment choice depends on the sialolith's size and anatomical position (CASTRO *et al.*, 2021, MEIRA, *et al.*, 2022). More conservative options include warm compresses, massages, salivary flow stimulation, duct milking or catheterization, duct dilation, increased fluid intake and citrus beverages to remove the sialolith. However, due to its substantial size, surgical removal was the best treatment option in this case (SILVA *et al.*, 2021; Basra *et al.*, 2024).

Conclusion:

Therefore, giant sialoliths require an accurate diagnosis and individualized planning, in which the most conservative treatment should always be prioritized. In this case, the surgical technique adopted was effective in removing the sialolith, preserving the glandular structures, and ensuring a satisfactory postoperative outcome, free from complications. The minimally invasive approach not only allowed for the elimination of the sialolith but also ensured the preservation of important structures, resulting in proper recovery and the complete restoration of the patient's salivary function.

References:

1. AVISHAI, G.; BEN-ZVI, Y.; GHANAHEM, O.; CHAUSHU, G.; GILAT, H. Sialolithiasis—do early diagnosis and removal minimize post-operative morbidity?, Archives of Health Investigation, v. 56, p. 332, 2020. DOI: <https://doi.org/10.3390/medicina56070332>.Disponível em: <https://www.mdpi.com/1648-9144/56/7/332> . Acesso em: 30 ago. 2024.
2. ASHINDOITIANG, J. A.; NWAGBARA, V. I. C.; UGBEM, T.; ODOKS, R. K.; UDO SOLOMON, A.; AKPAN, S. O.; OGAMBA, N.; ASUQUO, M. E. Huge sialolith of the submandibular gland: a case report and review of literature. The Journal of International Medical Research, v. 51, n. 1, 2023. DOI: <https://doi.org/10.1177/03000605221148443> .Disponível em: <https://pubmed.ncbi.nlm.nih.gov/36624984/>. Acesso em: 30 ago. 2024.
3. BARBEIRO, C. O.; BARBEIRO, R. H.; BUFALINO, A.; LEÓN, J. E. Suppurative minor salivary gland sialolithiasis. Autopsy and Case Reports, v. 12, 2022. DOI: <https://doi.org/10.4322/acr.2021.397>.Disponível em: <https://www.autopsycasereports.org/article/doi/10.4322/acr.2021.397> .Acesso em: 30 ago. 2024.
4. BASRA, Arshjot s *et al.* Management of a Submandibular Sialolith: a case report. Cureus, Wardha, p. 1-7, 6 jun. 2024. Springer Science and Business Media LLC. <http://dx.doi.org/10.7759/cureus.61812>. Acesso em: 31 ago. 2024.

5. BEUMER, L. J. *et al.* Success rate of sialendoscopy. A systematic review and meta-analysis. *Oral Diseases*, Leeuwarden, v. 30, n. 4, p. 1843-1860, 24 jul. 2023. Wiley. <http://dx.doi.org/10.1111/odi.14662>. Acesso em: 31 ago.2024.
6. CHIESA-ESTOMBA, Carlos Miguel *et al.* Laser-Assisted Lithotripsy With Sialendoscopy: systematic review of yo-ifos head and neck study group. *Ear, Nose & Throat Journal*, Donostia, v. 100, n. 1, p. 42-50, 22 maio 2020. SAGE Publications. <http://dx.doi.org/10.1177/0145561320926281>. Acesso em: 31 ago. 2024.
7. FOLCHINI, S.; STOLZ, A.B; Sialitosnaglandula submandibular: relato de caso. *Sialolitos the submandibular gland: a case report. OdontologiaClínico-Científica (Online)*, v.15, n.1. Recife. Jan/Mr. 2016. ISSN 1677-3888.
8. JADU, Fatima M.; JAN, Ahmed M.. A meta-analysis of the efficacy and safety of managing parotid and submandibular sialoliths using sialendoscopy assisted surgery. *Saudi Medical Journal*, Jeddah, v. 35, n. 10, p. 1118-1194, 12 ago. 2014. Acesso em 31 ago. 2024.
9. KOCH, M.; MANTSOPULOS, K.; MÜLLER, S.; SIEVERT, M.; IRO, H. Treatment of sialolithiasis: what has changed? An update of the treatment algorithms and a review of the literature. *Journal of Clinical Medicine*, v. 11, p. 231, 2022. DOI: <https://doi.org/10.3390/jcm11010231> . Disponível em: <https://www.mdpi.com/2077-0383/11/1/231>. Acesso em: 30 ago. 2024
10. KUHN-DALL MAGRO *et al.* Sialólito de glândulasalivar submandibular: relato de caso. Sialolith of submandibular salivary gland: case report. *RFO UPF*, v.19, n.3. Passo Fundo, Set/Dez. 2014. ISSN 1413-4012.
11. LIMA *et al.* Sialolitíaseemglândula submandibular: relato de casoclínico. Sialolithiasis in the submandibular gland: a clinical report. *Revista de Cirurgia e Traumatologia Buco-maxilofacial*, v.13, n.1, Camaragibe, Jan/Marc. 2013. ISSN 1808-5210.
12. LIRA PEREIRA DE CASTRO, C. C.; RODRIGUES, Éwerton D. R.; VASCONCELOS, B. C. do E.; MOREIRA, T. C. A. Abordagemintrabucal para exérese de sialolitonaglandula sublingual: relato de caso. *ARCHIVES OF HEALTH INVESTIGATION*, Piauí, v. 10, n. 3, p. 427-430, 2021. DOI: 10.21270/archi.v10i3.5185. Disponível em: <https://www.archhealthinvestigation.com.br/ArcHI/article/view/5185>. Acesso em: 30 ago. 2024.
13. LOMMEN, J., SCHORN, L., ROTH, B. *et al.* Sialolithiasis: retrospective analysis of the effect of an escalating treatment algorithm on patient-perceived health-related quality of life. *Head Face Med* 17, 8 (2021). DOI: <https://doi.org/10.1186/s13005-021-00259-1> . Disponível em: <https://head-face-med.biomedcentral.com/articles/10.1186/s13005-021-00259-1#citeas>Acesso em: 30 ago.2024
14. MEIRA, Clarina Louis Silva *et al.* Sialolitíase da glândula submandibular associada a defeitoósseo de stafne: relato de caso. *Jornal Brasileiro de Patologia e Medicina Laboratorial*, Belém, v. 58, p. 1-4, 22 set. 2021.. Acesso em: 31 ago. 2024.
15. MOHSIN, Syed Fareed; RIYAZ, Mohamed Abdulcader; ALQAZLAN, Abdulmalik Ali. Detection and Management of Giant Submandibular Gland Sialolith. *Pakistan Journal Of Medical Sciences*, Pakistan, v. 38, n. 7, p. 2030-2033, 2 ago. 2022. *Pakistan Journal of Medical Sciences*. <http://dx.doi.org/10.12669/pjms.38.7.5988>.. Acesso em: 31 ago. 2024.
16. MORTAZAVI, Hamed *et al.* What is the impact of previous cholelithiasis on sialolithiasis: a systematic review and meta-analysis. *The Saudi Dental Journal*, Ann Arbor, v. 36, n. 1, p. 44-51, jan. 2024. Elsevier BV. <http://dx.doi.org/10.1016/j.sdentj.2023.08.010>. Acesso em: 31 ago. 2024.
17. OLIVEIRA, Thiago de Paula *et al.* Giant sialolith of submandibular gland duct treated by excision and ductal repair: a case report. *Brazilian Journal Of Otorhinolaryngology*, Juiz de Fora, v. 82, n. 1, p. 112-115, jan. 2016. Elsevier BV. <http://dx.doi.org/10.1016/j.bjorl.2015.03.013>.. Acesso em: 31 ago. 2024
18. OLIVEIRA, M.; ACIOLY, R.; SOUZA, D.; SILVA, B.; CARVALHO, D.. Sialolito de grandesdimensõeseducto de glândula submandibular: relato de caso. *Revista de Cirurgia e Traumatologia Buco-Maxilo-Facial*, Camaragibe, v. 19, n. 2, p. 33-36, abr./jun. 2019. Disponível em: *Brazilian Journal of Oral and Maxillofacial Surgery*. Acesso em: 30 ago. 2024.
19. SANTANA, Daiana Cristina Pereira; GARCIA, Juliana Jorge; SILVA, Juliana Maria Araújo; BARBOZA, Alana Del'Arco; DE AZEVEDO, Roberto Almeida. Tratamentocirúrgico de sialolito intraglandular: relato de caso. *Journal of Health & Biological Sciences*, Bahia, v. 9, n. 1, p. 1-4, 2021. DOI: 10.12662/2317-3076jhbs.v9i1.3727.p1-4.2021. Disponível em: <https://periodicos.unichristus.edu.br/jhbs/article/view/3727>. Acesso em: 30 ago. 2024.
20. SILVA, C. B. da; CABRAL, L. N.; PINHEIRO, T. N.; VASCONCELOS II, A. J. A. de. Sialolitíaseemregião sublingual direita: relato de caso. *ARCHIVES OF HEALTH INVESTIGATION*, Manaus, v. 10, n. 3, p. 368-372, 2021. DOI: 10.21270/archi.v10i3.5181. Disponível em: <https://www.archhealthinvestigation.com.br/ArcHI/article/view/5181>. Acesso em: 30 ago. 2024.

21. SILVA, Camila Barbosa da *et al.* Sialolitíase em região sublingual direita: relato de caso. Archives Of Health Investigation, Manaus, v. 10, n. 3, p. 368-372, 2 mar. 2021. Archives of Health Investigation. <http://dx.doi.org/10.21270/archi.v10i3.5181>. Acesso em: 31 ago. 2024.
22. SILVEIRA JUNIOR, J. ; MATIAS NETO, J.; ANDRADE JUNIOR, I.; CAPISTRANO, H.. Multiple sialolithiasis in submandibular gland duct: a rare case report. RGO - Revista Gaúcha de Odontologia, v. 68, p. e20200029, 2020. DOI: <https://doi.org/10.1590/1981-863720200002920180103>. Disponível em: <https://www.scielo.br/j/rgo/a/HCyphxjjzm9vvCJwJHqTyTk/?lang=en#ModalHowcite>. Acesso: 30 ago. 2024.
23. SODNOM-ISH, B.; EO, M. Y.; CHO, Y. J.; *et al.* Identification of biological components for sialolith formation organized in circular multi-layers. Scientific Reports, v. 13, p. 12277, 2023. DOI: <https://doi.org/10.1038/s41598-023-37462-w>. Disponível em: <https://www.nature.com/articles/s41598-023-37462-w#citeas>. Acesso em: 30 ago. 2024.
24. SODNOM-ISH, Buyanbileg *et al.* Elemental characteristics of sialoliths extracted from a patient with recurrent sialolithiasis. Journal Of The Korean Association Of Oral And Maxillofacial Surgeons, Seoul, v. 50, n. 2, p. 94-102, 30 abr. 2024. The Korean Association of Oral and Maxillofacial Surgeons. <http://dx.doi.org/10.5125/jkaoms.2024.50.2.94>. Acesso em: 31 ago. 2024.
25. SOUZA, I. F., KAWATAKE, M. M., SOARES, A. B., MORAES, P. de C., ARAÚJO, V. C. de ., & PASSADOR-SANTOS, F.. (2015). Sialolithiasis of minor salivary glands. RGO - Revista Gaúcha de Odontologia, v. 63, n. 1, p. 63-68, jan. 2015. DOI: <https://doi.org/10.1590/1981-863720150001000092592>. Disponível em: <https://www.scielo.br/j/rgo/a/J7mWcd44pbcYFSVyD3XySZP/?lang=en#> . Acesso: 30 ago. 2024.
26. YING, Xiaoxia *et al.* Recurrent sialoliths after excision of the bilateral submandibular glands for sialolithiasis treatment: a case report. Experimental And Therapeutic Medicine, Dalian, v. 11, n. 1, p. 335-337, 10 nov. 2015. Spandidos Publications. <http://dx.doi.org/10.3892/etm.2015.2849>. Acesso em 31 ago. 2024.