American Journal of Otolaryngology and Head and Neck Surgery

Case Report
Published: 30 Apr, 2018

Pediatric Submandibular Gland Sialolithiasis with Salivary–Cutaneous Fistula: Case Report on Gland-Preserving Treatment with Combined Endoscopic–Transcutaneous Surgery

Achim Stegmann*, Heinrich Iro and Michael Koch

Department of Otorhinolaryngology, Friedrich Alexander University of Erlangen, Germany

Abstract

Objective: Salivary stones in the parotid and the submandibular glands are often seen diseases in ENT-Departments. More rarely these are seen in children. Complications generated by these stones are very seldom, especially in children. In the following case, a 6-year-old girl presented for further treatment due to a salivary–cutaneous fistula that had developed as a complication of left-sided submandibular gland sialolithiasis. Ultrasonography revealed a hyperechoic reflex 5 mm in size in the submandibular glandhilum.

Procedure: The Treatment consisted of endoscopically guided basket extraction of the stone and simultaneous excision of the fistula channel, with transcervical closure of the glandular capsule, preserving the gland.

Results: Even 10 years after the procedure, the patient is still without symptoms. There was no further stone seen in the gland and no signs of obstruction.

Conclusion: This case shows that gland-preserving treatment is possible even in complex situations in the submandibular gland - a very useful approach particularly in pediatric patients.

Keywords: Sialolithiasis; Complication; Submandibular gland; Salivary-cutaneous fistula; Treatment

OPEN ACCESS

****Correspondence:***
Achim Stegmann, Department of Otorhinolaryngology, Friedrich Alexander University of Erlangen, Germany, Tel: 49-9131-85-43812; Fax: 49-9131-85-33833;
E-mail: Achim.Stegmann@uk-erlangen.de

Received Date: *13 Mar 2018*
Accepted Date*: 20 Apr 2018*
Published Date*: 30 Apr 2018*

Citation:
Stegmann A, Iro H, Koch M. Pediatric Submandibular Gland Sialolithiasis with Salivary–Cutaneous Fistula: Case Report on Gland-Preserving Treatment with Combined Endoscopic–Transcutaneous Surgery. Am J Otolaryngol Head Neck Surg. 2018; 1(1): 1003.

Introduction

The development and introduction of minimally invasive treatment methods has led to fundamental changes in the way in which the large salivary glands are treated in patients with obstructive salivary gland diseases, particularly those with sialolithiasis [1,2]. Sialoendoscopy has also been used successfully as a diagnostic and therapeutic method in pediatric patients [3,4]. Combined endoscopic–transcutaneous procedures have proved to be a useful way of avoiding resection of the salivary glands in complex and treatment-resistant situations. To date, they have only been described in connection with the parotid gland [5-8]. Sialolithiasis in the submandibular gland is much rarer in children than in adults [3,4]. Fistulas caused by sialoliths represent a rare complication, and they develop more often with salivary–cutaneous fistulas toward the surface of the skin, rather than intraorally toward the mucosal surface. Complications of this type are extremely rare, particularly in pediatric patients, and they often mean that resection of the affected gland is required. They may be caused by an abscess with perforation of the glandular capsule, due to obstruction by the stone, or by migration of the stone itself [9-11].

Case Presentation

A 6-year-old patient presented 2006 with a space-occupying mass in the left submandibular area, with fistula formation to the skin surface. It had been present for approximately 4 months, occasionally with putrid secretion and sometimes causing pain. At the time of presentation, there was no pus emerging from the fistular orifice. Resistance in the posterior floor of the mouth was noted during bimanual palpation, but it was not possible to express saliva *via* the Wharton duct. Ultrasound examination revealed a hyperechoic reflex 5 mm × 4 mm in size, with dorsal signal loss, in the hilum of the left submandibular gland. The parenchyma showed hypoechoic loosening.

Figure 1: Preoperative ultrasound examination. There is a hyperechoic reflex 5.4 mm in size with a signal loss in the hilum of the left submandibular gland A) indicating the presence of calculus. Between the glandular parenchyma and the skin level, there is a 17-mm long hypoechoic structure, B) indicating a salivary–cutaneous fistula.
GSM: Submandibular Gland; Fistel: Fistula

Figure 2: A) Sialoendoscopic images of the hilar stone before, B) after interventional sialoendoscopy. The stone was mobilized with a drill and extracted using a basket.

Figure 3: A) The intraoperative view of the left submandibular fistular orifice before, B) after excision. The glandular capsule (arrow) has not yet been closed.

In the hilum, a hyperechoic reflex with signal loss was seen, and a hypoechoic cordal structure 17 mm long to the surface of the skin was noted (Figure 1A and B). The diagnosis was sialolithiasis of the submandibular gland with development of a salivary–cutaneous fistula, against the background of a previous abscess. Treatment was carried out under general anesthesia. During ductal endoscopy, the stone was located in the hilum. Its size was reduced intraductally with a microdrill, and the fragments were removed using a basket (Figure 2A and B). As the obstruction had been removed, an effort was made to preserve the submandibular gland. An oval incision was made around the fistula, with dissection as far as the capsule of the submandibular duct. The fistula was then excised in to and the glandular capsule was closed with 4-0 Vicryl interrupted sutures (Figure 3A and B).

The postoperative course was completely uneventful (Figure 4). Almost 10 years after the procedure, the patient is still free of stones and completely free of symptoms. Glandular function was preserved.

Figure 4: Postoperative ultrasound examination. The glandular parenchyma is still showing hypoechoic changes, but there is no evidence of a stone or fistula.
GSM: Submandibular Gland; DW: Wharton's Duct; Z: Tongue; MM: Mylohyoid Muscle

Discussion and Conclusion

Gland-preserving therapy is an established method that can be used in over 90% of cases, including children. Following gland-preserving treatment, glandular function recovers in nearly 90% of cases [12,13]. Recent studies have shown that both symptoms and quality of life significantly improve after treatment for obstructive sialadenitis, particularly after the removal of stones [14]. Following resection of the submandibular gland, there is an increased risk of caries developing, as a result of reduced salivary flow [15]. Particularly in pediatric patients undisturbed dental development can be achieved through glandular function preserving treatment.

The combined endoscopic–transcutaneous approach is an additional minimally invasive procedure that can be used to preserve the gland even in cases of treatment resistance. Although there have been earlier reports in connection with transoral surgery on endoscopically assisted procedures in the submandibular gland [7,16], a combined endoscopic–transcutaneous approach in this area has not previously been described. The approach has been successful particularly in patients with complications in the parotid gland, and especially after the development of a salivary–cutaneous fistula [8]. The positive and complication-free course in the present case and the patient's improved quality of life after treatment indicate that both in obstructive sialadenitis, particularly in the presence of sialolithiasis, minimally invasive gland-preserving therapy is useful and should be the treatment of choice. The development of a complication in such cases no longer represents a strict indication for gland resection. The decisive aspect is to relieve the cause of the obstruction in order to restore anterograde salivary flow. This can be achieved both by using interventional sialoendoscopy and also through transoral ductal incision.

References

1. Iro H, Zenk J, Escudier MP, Nahlieli O, Capaccio P, Katz P, et al. Outcome of minimally invasive management of salivary calculi in 4,691 patients. Laryngoscope. 2009;119(2):263-8.
2. Koch M, Zenk J, Iro H. Algorithms for treatment of salivary gland obstructions. Otolaryngol Clin North Am. 2009;42(6):1173-92.
3. Nahlieli O, Eliav E, Hasson O, Zagury A, Baruchin AM. Pediatric sialolithiasis. Oral Surgery Oral Medicine Oral Pathology Oral Radiology. 2000;90(6):709-12.

4. Faure F, Querin S, Dulguerov P, Froehlich P, Disant F, Marchal F. Pediatric salivary gland obstructive swelling: sialendoscopic approach. Laryngoscope. 2007;117(8):1364-7.

5. Nahlieli O, London D, Zagury A, Eliav E. Combined approach to impacted parotid stones. J Oral Maxillofac Surg. 2002;60(12):1418-23.

6. McGurk M, MacBean AD, Fan KFM, Sproat C, Darwish C. Endoscopically assisted operative retrieval of parotid stones. Br J Oral Maxillofac Surg. 2006;44(2):157-60.

7. Marchal F. A combined endoscopic and external approach for extraction of large stones with preservation of parotid and submandibular glands. Laryngoscope. 2007;117(2):373-7.

8. Koch M, Bozzato A, Iro H, Zenk J. Combined endoscopic and transcutaneous approach for parotid gland sialolithiasis: indications, technique, and results. Otolaryngol Head Neck Surg. 2010;142(1):98-103.

9. Drage NA, Brown JE, Makdissi J, Townend J. Migrating salivary stones: report of three cases. Br J Oral Maxillofac Surg. 2005;43(2):180-2.

10. Saha S, Jha A, Kaur N. Unusual cause of orocutaneous fistula in the neck. Case Rep Surg. 2012;2012:658536.

11. Kieliszak CR, Gill A, Faiz M, Joshi AS. Submandibular ductal fistula: an obstacle to sialendoscopy. JAMA Otolaryngol Head Neck Surg. 2015;141(4):373-6.

12. van den Akker HP, Busemann-Sokole E. Submandibular gland function following transoral sialolithectomy. Oral Surg Oral Med Oral Pathol. 1983;56(4):351-6.

13. Makdissi J, Escudier MP, Brown JE, Osailan S, Drage N, McGurk M. Glandular function after intraoral removal of salivary calculi from the hilum of the submandibular gland. Br J Oral Maxillofac Surg. 2004;42(6):538-41.

14. Gillespie MB, O'Connell BP, Rawl JW, McLaughlin CW, Carroll WW, Nguyen SA. Clinical and quality-of-life outcomes following gland-preserving surgery for chronic sialadenitis. Laryngoscope. 2015;125(6):1340-4.

15. Cunning DM, Lipke N, Wax MK. Significance of unilateral submandibular gland excision on salivary flow in noncancer patients. Laryngoscope.1998;108(6):812-5.

16. Nahlieli O, Shacham R, Zagury A, Bar T, Yoffe B. The ductal stretching technique: an endoscopic-assisted technique for removal of submandibular stones. Laryngoscope. 2007;117(6):1031-5.