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Original Article

Transoral submandibulotomy plus duct marsupialization; an appropriate approach for the treatment of proximal submandibular sialolithiasis; a long-term follow-up study

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Abstract: Background: Sialolithiasis is the most common salivary gland-related disease worldwide, leading to sialadenitis. Although there is no serious problem with surgical removal of stones at the middle and distal parts of the Wharton's duct, the approach for treating proximal stones located near to hilum and its adjacent parenchyma is a challenge. The current study has aimed to evaluate the outcome of transoral submandibulotomy for submandibular gland stone treatment. Methods: This study was conducted on 63 patients with proximal (or deep hilar) submandibular sialolithiasis treated with transoral submandibulotomy and duct marsupialization from January 2011 to April 2019 in Pars hospital. Complications of the old fashion surgery (transcervical submandibulectomy) were assessed in this method (number and size of the stone(s), relapse of sialolithiasis-related obstructive symptoms, partial or complete removal of the stone(s), sialolithiasis recurrence, postoperative surgical or nonsurgical intervention, foreign body sensation, and taste sensation quality). Results: The stone(s) was located at the proximal of the duct in 68% of the cases, while 32% of the stones were located at the deep hilar region. The mean size of the stones was 1.28±0.51 cm. Postoperative inflammation, postoperative pain, sialolithiasis recurrence, obstructive symptoms, postoperative nonsurgical intervention, postoperative surgical intervention, reduced taste sensation, and foreign body sensation were presented in 24%, 29%, 29%, 44%, 14%, 6%, 11%, and 10% respectively. Eighty-nine percent of recurrences were healed spontaneously. Conclusion: Considering the cosmetic advantage due to the lack of cervical surgery scar (which is mandatory in old fashion transcervical method), long-term follow-up of transoral submandibulotomy showed successful outcomes regarding neurological impairment, postoperative pain and inflammation and incredibly complete stone removal that was found in all of the patients. In addition, submandibular duct marsupialization is recommended based on our study.

Keywords: Submandibular salivary gland calculi, submandibular salivary duct calculi, transoral submandibulotomy, stone removal, submandibular gland

Introduction

Sialolithiasis, or salivary gland stone, is the most common salivary gland-related disease worldwide, leading to sialadenitis [1]. It has been estimated that up to 1.2% of the general population are affected by the disease, among which 80% of symptomatic ones affect the submandibular gland. Over half of the submandibular glands are located in the hilum or its adjacent parenchyma [2].

While submandibular gland removal may pose symptom relief, novel techniques regarding saving this gland. Among them ultrasound and sialendoscopy-assisted transoral submandibulotomy can be mentioned [3].

Although there are no serious issues with surgical removal of stones located at the middle and distal parts of the Wharton's duct, the value of these novel techniques can be better clarified for stones located in the proximity of the submandibular gland near to hilum and its adjacent parenchyma due to its anatomical position. The clinical manifestations of salivary gland calculi include pain and swelling of the gland, dull pain of the gland and swelling of the face or neck, dry mouth and difficulty swallowing or opening the mouth. The Wharton's duct originated from

Table 1. Comparison of alternative methods of salivary stone removal complication in contrast with routine trance cervical approach

Variable	Skin Scar	Recurrence	Nerve related side effects	other
Trans cervical method (routine type)	+	Remnant of stone in the salivary duct Possible recurrence in the remnant on gland	Probable risk of marginal mandibular nerve injury, as well as lingual nerve injury with lower probability	Recurrence, abscess formation and other complication due to the salivary duct stone remnant (focusing on the gland rather than duct)
Trans oral method (in our method with duct marsupialization)	-	Probable as the gland itself is preserved in this method	Lower nerve injury probability as imply different approach to access salivary gland (0% in our study)	Lower ductal stone remnant related complication as focusing on the duct instead of gland
Intraductal lithotripsy	-	Probable as the gland itself is preserved in this method	-	Limitation in multiple, large or proximal stones that decrease the success an confine the method for selected cases
Laser lithotripsy	-	Probable as the gland itself is preserved in this method	-	Limitation in deep hilar stones that decrease the success an confine the method for selected cases
pneumatic lithotripsy	-	Probable as the gland itself is preserved in this method	-	Acceptable just for selected cases

the distal parts of the submandibular gland and then curves around the mylohyoid posterior border over the lingual nerve [4]. Therefore any injury to any part of this pathway can cause considerable complications [5].

Although the technique of transoral submandibulotomy has been raised with the aim of submandibular gland preserving in hilar and its adjacent parenchyma stones treatment, most of the stones are treated with the submandibulectomy approach worldwide [6].

It should be notified that the gland's function could be successfully recovered following the removal of the calculi. Furthermore, the transoral submandibulotomy technique is more economical due to the shorter duration of hospitalization and more favored by patients due to less postoperative pain [7, 8]. Comparison of alternative methods of salivary stone removal complication in contrast with routine trance cervical approach is shown in **Table 1**.

The number of studies assessing the technique of transoral submandibulotomy is limited. Based on our knowledge, this study is the first effort to evaluate the long-term efficacy and postoperative early and late complications of transoral submandibulotomy for submandibular gland stone treatment in Iran.

Methods and material

Data and parameters

This is an analytic-descriptive study on 63 patients referred to the Department of Otorhinolaryngology; Pars Hospital; affiliated with Isfahan University of Medical Sciences, from January 2011 to April 2019. The study protocol was approved by the ethics committee of Isfahan University of Medical Sciences (Ethics code: IR.MUI.MED.REC.1398.068).

Inclusion and exclusion criteria

The inclusion criteria were salivary gland calculi less than 3 cm in diameter, treatments with the technique of transoral submandibulotomy, having access to patient's medical records, and signing the written informed consent to participate in this study. The exclusion criteria were medical record defects and patients' will to exit the study.

Preoperative and postoperative assessments

Clinical examination, and neck computerized tomography (CT) scan without contrast was performed for all of the patients to select the best surgical procedure option. The transoral approach for the surgical procedure was considered for stones located in the deep hilar and/or beginning of the submandibular gland intraparenchymal region, found through CT scan.

Postoperative management included seven days of treatment of amoxicillin/clavulanate or clindamycin in case of allergy to penicillins, and anti-inflammatory medications (500 mg naproxen, twice daily for five days). In addition, a daily operated gland massage was performed.

Figure 1. After incision on the on the floor of mouth cavity, salivary duct and gland hilum are shown.

Surgical interventions

Wharton's duct was prepared for dissection within 3-5 mm posterior to the papilla at the site of the papillary or retropapillary region. Afterwards, it was dissected at the hilar region of the gland and the incision was made up to the hilum. As shown in **Figure 1**, salivary duct and gland hilum are observed after incision on the floor of mouth cavity.

Then further preparation was considered for lingual nerve dissection to assess anatomical variations and minimize probable neural injuries. Following the access to the hilar region, an external mild manual pressure was applied to elevate the submandibular gland's hilum beyond the mylohyoid muscle posterior border. In cases where the stone could not be detected following the incision made to the hilum still, the stone(s) was palpable within the adjacent gland, submandibulotomy was performed in a term that an incision in the adjacent parenchyma of the gland directly over the stone and under direct visual control was made. After primary incision on the left side of on the floor of mouth, salivary duct pathway is revealed to the gland hilum (**Figure 2**). After the explosion of the stone, it is removed by the small incision (**Figure 3**).

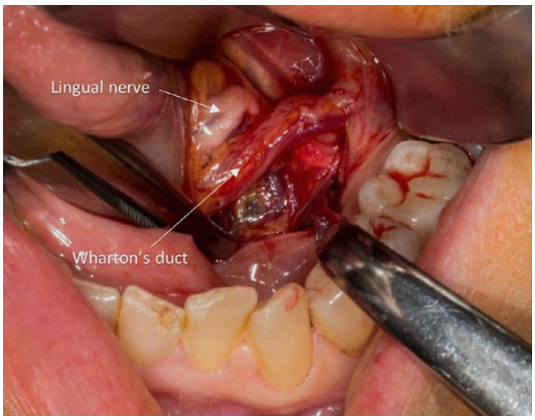

Figure 2. After primary incision on the left side of on the floor of mouth, salivary duct pathway is revealed to the gland hilum.

Eventually, the duct system and incised submandibular gland parenchyma were sutured to the epithelium of the oral cavity. Therefore a wide neo-ostium with the proper salivary flow was created. Eventually, salivary duct marsupialization was performed for all of the patients.

During the surgical procedures, the stone characteristics including the location, number of stones, and size of stones were collected.

Follow-up

Postoperative and follow-up study of sialolithiasis treatment was conducted by postoperative visits 4 weeks after surgeries. The following items were evaluated:

- Presence of postoperative inflammation (evaluated by physician's opinion).
- Presence of postoperative pain (according to patient's interview).
- Sialolithiasis recurrence (based on physical examination).
- Obstructive symptoms recurrence (according to patient's interview).
- Postoperative nonsurgical intervention requirement (based on patient's files).
- Taste sensation reduction (according to patient's interview).
- Foreign body sensation (according to patient's interview).

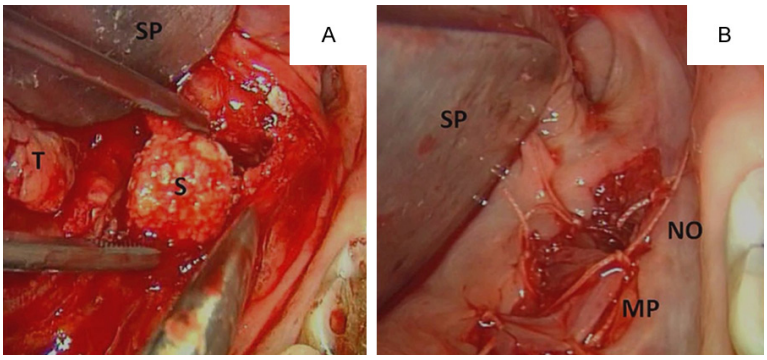

Figure 3. After the explosion of the stone (A), it is removed by the small incision over the mentioned area. Incision site on salivary gland is marsupialized to the oral mucosa of the on the floor of mouth cavity (B). SP: spatula; T: tongue; S: stone; NO: new ostium; MP: marsupialized duct [27].

- The willingness of the same operation if necessary (according to patient's interview).

- Postoperative surgical intervention requirement (based on patient's files).

- Postoperative stone remnant (using neck CT scan without contrast).

- The acceptability of the procedure (ranked from 1 to 6 as very comfortable to very uncomfortable, respectively) [9].

All of the symptoms were explained to the patients thoroughly.

Analysis

The obtained data were entered into the Statistical Package for Social Sciences (SPSS) (version 24, SPSS Inc., Chicago, IL). Quantitative data were reported as mean ± standard deviation and qualitative data as frequency distribution (percentage). *P*-value < 0.05 was considered as significance threshold.

Results

Study population

In the current study, 63 patients with the chief complaint of the proximal submandibular gland and deep hilar sialolithiasis who had been surgically operated on with the transoral submandibulotomy were assessed and followed. The mean age of the studied population was 45.2±14.15 years, and 68% were male.

Stone characteristics

The stone(s) was located at the proximal of the duct in 68% of the cases while 32% of the stones were located at the deep hilar region.

Fifty-two patients (82%) had only one stone, eight of them (13%) had 2 stones, and remained three patients (5%) had three or more stones. The mean number of stones was 1.24±0.59, and their mean size was 1.28±0.51 cm, with the range of 0.4-5.5 cm. Postoperative inflammation was presented by fifteen patients (24%) and postoperative pain complaints by 18 patients (29%).

Follow-up data

Sialolithiasis recurrence occurred in 18 patients (29%), and the participants observed obstructive symptoms were observed in 28 (44%). Furthermore, 14% of patients required nonsurgical postoperative interventions, four (6%) patients needed postoperative surgical management, one of them being due to a reason other than sialolithiasis.

Complications

11% and 10% of patients presented postoperative complaints of reduced taste and foreign body sensation, respectively; none of them complained about complete paresis of the lingual nerve. In addition, an antibiotic requirement due to infection was only found in 3% of the patients.

Among 18 patients who presented sialolithiasis recurrence, only two required surgical procedures for the treatment. The other postoperative surgical intervention was performed because of mass detection in the submandibular salivary gland (**Table 2**).

Further postoperative evaluations revealed complete stone removal in the study population, and 92% of the patients presented their willingness for the same reoperation if necessary. 76% of the patients were very comfortable, and only 5% of patients were very uncom-

Table 2. Postoperative and follow-up study of sialolithiasis treatment with the technique of submandibulotomy regarding early and late complications

Variable		Frequency	Percent	Variable		Frequency	Percent
Postoperative inflammation	no	48	76	Taste sensation reduction	no	56	89
	yes	15	24		yes	7	11
Postoperative pain	no	45	71	Foreign body sensation	no	57	90
	yes	18	29		yes	6	10
Sialolithiasis recurrence	no	45	71	The willingness of the same operation if necessary	no	5	8
	yes	18	29		yes	58	92
Obstructive symptoms recurrence	no	35	56	Postoperative surgical intervention requirement	no	59	94
	yes	28	44		yes	4	6
Postoperative nonsurgical intervention requirement	no	54	86	Postoperative stone remnant	no	63	100
	yes	9	14		yes	0	0

Table 3. Assessment of submandibulotomy acceptability based on the numerical ranking scale

Acceptability of the procedure	Frequency	Percent
Very comfortable	48	76
Comfortable	9	14
Partially comfortable	3	5
Partially uncomfortable	0	0
Uncomfortable	0	0
Very uncomfortable	3	5

fortable following the surgical procedure (using the numerical ranking scale) (**Table 3**).

Discussion

In our method of surgery, besides having a better cosmetic outcome, we focus on the salivary duct other than the gland itself in salivary gland stone removal and preserving the main gland.

Sialolithiasis, or salivary gland stone, is the most common salivary gland affecting disease and is also the most frequent etiology responsible for submandibulectomy [10]. Although submandibular glands are responsible for approximately more than 50% of unstimulated salivary flow, the impaired function following removal of this gland may be compensated within the time, and submandibular gland removal is not necessarily accompanied by xerostomia [11, 12]. However, its perseverance is superior to save the ultimate salivary glands function as we have done in our study.

The treatment technique is still a significant concern worldwide as gland resection without complete stone removal, preserved in the remnant of the duct, may cause abscess formation within several years after the surgical procedure [8, 10, 11]. Therefore, complete removal of the stone, not only the gland, should be precisely considered Sialolithiasis. Complete removal is the main aim of the surgical procedure [3]. This should be why we had a lesser percentage of the stone remnant in contrast with other studies, as we concentrate on the duct instead of the salivary gland.

A common approach for the treatment of submandibulectomy in sialolithiasis is the transcervical approach about which the following points should be considered:

1. General anesthesia requirements [8].

2. The significant potential risk for marginal mandibular nerve injury [13].

3. Aesthetic manner of cervical scar formation [14].

Moreover, this technique is not entirely safe for the lingual nerve. In a review article by McGurk and colleagues, Permanent lingual nerve paresis has been estimated to be up to 2% [6] that is significantly higher than the studies assessing transoral approach [3, 15, 16]. We have found this complication in 0% of our study population.

In the current study, we utilized physical examination and CT scan in order to select the approach of transoral submandibulotomy for our study population. Ninety-two percent of the patients were satisfied with the procedure and

presented their willingness for reoperation with this technique if necessitated. Furthermore, most of the studied population declared being very comfortable following this approach representing its efficacy. This rate was almost the same in the study of Schapher and colleagues (witch have used the transoral approach as well) presenting up to the proportions of 91% postoperative satisfaction [3].

The most notifying finding of our study is regarding the complete removal of stones in all cases regardless of the number of stones due to the better exposure of the salivary duct. In contrast, other studies presented a complete reduction to the maximal rate of 85% [1, 10, 17].

Studies have not achieved a unanimous approach regarding duct marsupialization following sialolithiasis removal, while we have found among the total number of 18 recurrences, only two required surgical intervention and the remaining sixteen stones were excreted spontaneously. While Johannes Zenk and colleagues presented their favor for duct marsupialization [18], Pasquale Capaccio and colleagues presented inverse theory [19].

Furthermore, significant postoperative symptoms, including pain and inflammation, were presented in 29 and 24% of the cases. In addition, in follow-up visits, 44% of the study population regained their obstructive symptoms that recovered spontaneously or with nonsurgical intervention. Based on the literature, choices for those with symptomatic recurrent stones include intraductal lithotripsy [20, 21]. The last surgical option considered for these patients is submandibulectomy [22]. Intraductal lithotripsy has developed dramatically in recent years and its success rate has been estimated to be up to 90%. The only limitation of this technique is inaccessibility to the exact location of the stones. The success rate of this technique declines for the treatment of multiple and large stones (more than 8 mm) and proximal ones [20, 23]. Laser lithotripsy has been developed recently with acceptable outcomes, though further evaluations are required, in the deep hilar region in special [24]. Moreover, Koch and colleagues performed a study using a new technique of pneumatic lithotripsy. Eventually, they presented 100% successful outcomes of stone removal located in the deep hilar region [25]. Nevertheless, these results should be confirmed through further evaluations, especially for stones 8 to 10 mm in size.

In this study, complaints including tastelessness, taste sensation reduction, and foreign body sensation were presented in 0%, 11%, and 10% of patients, respectively. Salles and colleagues performed a similar study demonstrating no permanent neurological disorder among their study population [26]. Johannes Zenk and colleagues conducted another study presenting neurological complaints as paresthesia or anesthesia only in 1% of their study population. They also submitted no complaints of movement dysfunction or foreign body sensation among their study population [18]. Schapher and colleagues conducted their study assessing transoral submandibulotomy on a significant number of 234 patients. Complications including numbness, movement dysfunction, anesthesia, and tastelessness were presented in 9.1% of their study population [3]. Differences in various studies may be attributed to the size of the study population, the experience of the surgeon, and the facilities used during the surgical procedure.

In this study, only a person required antibiotic therapy for postoperative infection. At the same time, Capccio and colleagues performed their analysis with a similar approach for submandibular gland stone removal. They presented that 2.6% (12 cases) of their patients required antibiotic therapy because of developing recurrent infections [1].

The limitations of this study were the lack of comparisons with other types of treatments and the restricted study population. We recommend that further multi-centric studies should be conducted in the future.

Conclusion

In summary, long-term follow-up of transoral submandibulotomy consistent with previous studies showed successful outcomes regarding neurological impairment, postoperative pain, and inflammation, and complete stone removal in special that was found in all of the patients. Also, it has better aesthetic outcomes due to not having a neck scar, which is inevitable in the old fashion transcervical surgery

method. Yet, due to the limited amount of evidence, further studies with a more extensive population are recommended.

Disclosure of conflict of interest

None.

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References

[1] Capaccio P, Gaffuri M, Rossi V and Pignataro L. Sialendoscope-assisted transoral removal of hilo-parenchymal sub-mandibular stones: surgical results and subjective scores. Acta Otorhinolaryngol Ital 2017; 37: 122-127.

[2] Moradi Farsani D, Shetabi H, Rafiee Zadeh A and Saffari Rad N. Comparison of Tranexamic acid, Remifentanil, and Hydralazine on the bleeding volume during Dacryocystorhinostomy surgery. Int J Physiol Pathophysiol Pharmacol 2022; 14: 177-186.

[3] Schapher M, Mantsopoulos K, Messbacher ME, Iro H and Koch M. Transoral submandibulotomy for deep hilar submandibular gland sialolithiasis. Laryngoscope 2017; 127: 2038-2044.

[4] Burian E, Probst FA, Weidlich D, Cornelius CP, Maier L, Robl T, Zimmer C, Karampinos DC, Ritschl LM and Probst M. MRI of the inferior alveolar nerve and lingual nerve-anatomical variation and morphometric benchmark values of nerve diameters in healthy subjects. Clin Oral Investig 2020; 24: 2625-34.

[5] Guenzel T, Hoch S, Heinze N, Wilhelm T, Gueldner C, Franzen A, Coordes A, Lieder A and Wiegand S. Sialendoscopy plus laser lithotripsy in sialolithiasis of the submandibular gland in 64 patients: a simple and safe procedure. Auris Nasus Larynx 2019; 46: 797-802.

[6] McCain JP and Montero J. Surgical retrieval of submandibular stones. Atlas Oral Maxillofac Surg Clin North Am 2018; 26: 111-117.

[7] Xiao JQ, Sun HJ, Qiao QH, Bao X, Wu CB and Zhou Q. Advantages of submandibular gland preservation surgery over submandibular gland resection for proximal submandibular stones. Oral Surg Oral Med Oral Pathol Oral Radiol 2018; 125: e113-e117.

[8] Saga-Gutierrez C, Chiesa-Estomba CM, Larruscain E, González-García JÁ, Sistiaga JA and Altuna X. Transoral sialolitectomy as an alternative to submaxilectomy in the treatment of submaxillary sialolithiasis. Ear Nose Throat J 2019; 98: 287-290.

[9] Yesilyurt M. Evaluation of patients using numeric pain-rating scales. Int J Caring Sci 2021; 14: 890-7.

[10] Foletti JM, Graillon N, Avignon S, Guyot L and Chossegros C. Salivary calculi removal by minimally invasive techniques: a decision tree based on the diameter of the calculi and their position in the excretory duct. J Oral Maxillofac Surg 2018; 76: 112-118.

[11] Burghartz M, Hackenberg S, Sittel C and Hagen R. Surgery of the major salivary glands and its impact on salivary flow-A review. Laryngoscope 2019; 129: 2053-2058.

[12] Sproll C, Naujoks C, Holtmann H, Kübler NR, Singh DD, Rana M and Lommen J. Removal of stones from the superficial lobe of the submandibular gland (SMG) via an intraoral endoscopy-assisted sialolithotomy. Clin Oral Investig 2019; 23: 4145-4156.

[13] Goh LC, Chitra BK, Shaariyah MM and Ng WS. Transcervical approach to the removal of a deep-seated giant submandibular calculus and the submandibular gland. BMJ Case Rep 2016; 2016: bcr2016217514.

[14] Delsing CPA, Bekkers S, van Hulst K, Erasmus CE and van den Hoogen FJA. Unsuccessful submandibular duct surgery for anterior drooling: surgical failure or parotid gland salivation? Int J Pediatr Otorhinolaryngol 2019; 123: 132-137.

[15] Li J, Xu XY, Lu ZW, Guan QB and Chen JF. Sialendoscopy-assisted intraoral incision approach for the treatment of posterior Wharton's duct stones: our experience and outcomes. Wideochir Inne Tech Maloinwazyjne 2021; 16: 249-255.

[16] Capaccio P, Montevecchi F, Meccariello G, D'Agostino G, Cammaroto G, Pelucchi S and Vicini C. Transoral robotic surgery for hilo-parenchymal submandibular stones: step-by-step description and reasoned approach. Int J Oral Maxillofac Surg 2019; 48: 1520-1524.

[17] Koch M, Schapher M, Mantsopoulos K, Goncalves M and Iro H. Intraductal pneumatic lithotripsy after extended transoral duct surgery in submandibular sialolithiasis. Otolaryngol Head Neck Surg 2019; 160: 63-69.

[18] Vergez S, Cheval M and Chabrillac E. Transoral robotic removal of submandibular sialolith combined with sialendoscopic assistance. Eur Ann Otorhinolaryngol Head Neck Dis 2021; 138 Suppl 2: 65-66.

[19] Kumar S, Kumar S, Arya V, Bootwala F, Ranganathan V, Thakker R, Hameed A and Saxena S. Bilateral hilar sialoliths in a child: a rare occur-

rence. J Oral Maxillofac Pathol 2022; 26 Suppl 1: S143-S145.
[20] Kondo N, Yoshihara T, Yamamura Y, Kusama K, Sakitani E, Seo Y, Tachikawa M, Kujirai K, Ono E, Maeda Y, Nojima T, Tamiya A, Sato E and Nonaka M. Treatment outcomes of sialendoscopy for submandibular gland sialolithiasis: the minor axis of the sialolith is a regulative factor for the removal of sialoliths in the hilum of the submandibular gland using sialendoscopy alone. Auris Nasus Larynx 2018; 45: 772-776.
[21] Lafont J, Graillon N, Hadj Saïd M, Tardivo D, Foletti JM and Chossegros C. Extracorporeal lithotripsy of salivary gland stone: a 55 patients study. J Stomatol Oral Maxillofac Surg 2018; 119: 375-378.
[22] Ozdemir S. Outcomes of pneumatic lithotripsy versus holmium laser-assisted lithotripsy with sialendoscopy in management of submandibular sialolithiasis. J Craniofac Surg 2020; 31: 1974-1977.
[23] Sionis S, Caria R, Trucas M, Brennan P and Puxeddu R. Sialoendoscopy with and without holmium: YAG laser-assisted lithotripsy in the management of obstructive sialadenitis of major salivary glands. Br J Oral Maxillofac Surg 2014; 52: 58-62.
[24] Su CH, Lee KS, Tseng TM and Hung SH. Endoscopic holmium:YAG laser-assisted lithotripsy: a preliminary report. B-ENT 2015; 11: 57-61.
[25] Koch M, Mantsopoulos K, Schapher M, von Scotti F and Iro H. Intraductal pneumatic lithotripsy for salivary stones with the StoneBreaker: preliminary experience. Laryngoscope 2016; 126: 1545-1550.
[26] Capaccio P, Di Pasquale D, Bresciani L, Torretta S and Pignataro L. 3D video-assisted transoral removal of deep hilo-parenchymal submandibular stones. Acta Otorhinolaryngol Ital 2019; 39: 367-373.
[27] Schapher M, Mantsopoulos K, Messbacher ME, Iro H and Koch M. Transoral submandibulotomy for deep hilar submandibular gland sialolithiasis. Laryngoscope 2017; 127: 2038-44.