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Transoral removal of hiloparenchymal submandibular calculi: a long-term clinical experience

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Abstract Traditional management of hiloparenchymal submandibular calculi is based on sialadenectomy. Recently, different minimally invasive and conservative techniques have been developed for the treatment of the submandibular calculi. We aimed to investigate the effectiveness of transoral surgical removal of large hiloparenchymal calculi by monitoring the trend for recurrence with clinical and ultrasonographic follow-up. A consecutive series of 84 patients with large (>7 mm) hilar or hiloparenchymal submandibular calculi underwent the transoral surgical removal under general anaesthesia. A video-assisted endoscopic procedure was performed in eight patients. All the patients underwent diagnostic ultrasonography and colour Doppler ultrasonography and clinical evaluation to define the exact location (hilar vs. parenchymal) and the diameter of the stone. The surgical procedure was successful in all but one of the patients. Stone recurrence was observed in 16 patients but obstructive symptoms were observed in only 12 patients during a median follow-up time of 52 months. The risk for recurrence was higher in patients who previously underwent extracorporeal shock-wave lithotripsy. Conservative transoral removal of large hiloparenchymal submandibular calculi is a safe and effective surgical procedure. Future studies with longer follow-up will confirm the risk for recurrence of calculi.

Keywords Salivary calculi · Transoral surgery · Ultrasonography · Sialoendoscopy

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Introduction

Sialolithiasis is one of the most common non-neoplastic diseases of the large salivary glands with an incidence in the autopsy population of 1.2% [1, 2]. Most stones are located mainly in the distal tract and hilum of the submandibular glands; intraparenchymal stones are less frequent (<10%) [2, 3].

Traditional management of proximal and hiloparenchymal submandibular stones is based on sialadenectomy [4] with its known risks of injury to the facial, lingual or hypoglossal nerves, Frey's syndrome and unaesthetic scars [5, 6]. In recent years, conservative techniques have been developed for the treatment of the submandibular calculi, such as extracorporeal shock wave lithotripsy [3, 7], sialoendoscopy [8–10], interventional radiology [11], transoral surgery [2, 14] and endoscopic-assisted transoral surgery [2, 12]. Clinical experience with extracorporeal shockwave lithotripsy has shown that only 30% of submandibular stones, in particular large stones (>7 mm) and those in the hiloparenchymal are not responsive to this modality of treatment [7]. The treatment of choice for large or fixed stones is endoscopic-assisted intra-oral removal of stones [12, 13]. The follow-up of patients undergoing conservative treatment for submandibular gland calculi may be assessed clinically, scintigraphically [15], or by ultrasound [16]. The

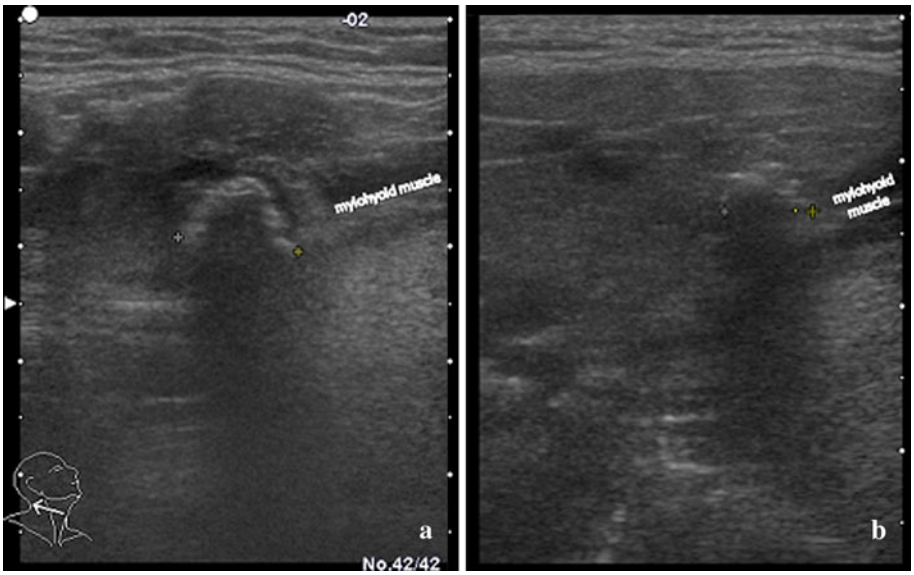

Fig. 1 Ultrasonographic identification of the location of the submandibular calculi: **a** an hilo-parenchymal calculus located at the level of the mylohyoid muscle; **b** a parenchymal calculus located above the level of the mylohyoid muscle

clinical outcome is difficult to standardize as criteria vary in the literature. Scintigraphic examination has demonstrated a variable improvement in glandular function in most patients after stone removal [15]. Ultrasonography (US) is also an amenable and simple method of assessing duct and parenchymal architecture.

The aim of this study was to determine the role of conservative transoral surgical removal in the management of large hiloparenchymal submandibular calculi by monitoring recovery with a combination of clinical and US assessment.

Methods

Eighty-four patients (51 M:33 F) with a mean age of 52 years (range 22–81 years) with symptomatic hilo-parenchymal stones of the submandibular glands underwent videoendoscopic-assisted transoral surgical removal of stones between April 2003 and April 2008. A small cohort of these patients ($n = 18$) had been unsuccessfully treated previously by extracorporeal shockwave lithotripsy. The study was approved by the local ethics committee and all the patients gave their informed consent to take part in the study.

All patients underwent ultrasound and Doppler sonographic assessment (Hitachi H21, 7.5 MHz, Hitachi High Technology Corporation Ltd., Tokyo, Japan) in conjunction with clinical evaluation to define location (hilar vs. parenchymal) (Fig. 1a, b) and size of stone (minimum diameter 8 mm). In addition, 23 patients underwent standard X-ray and sialographic assessment. The stone was clinically defined as hilar when at least two margins were detectable during bimanual palpation of the oral floor; the stone was defined as hiloparenchymal when only the distal margin was detectable during palpation and the remaining margins were covered by glandular tissue; the stone was defined as parenchymal when completely covered by glandular tissue. The exclusion criteria were a significant limitation in mouth opening and ductal atresia (diagnosed by US and MR-sialography).

Surgical procedure

Due to the proximal position of the stone, the procedure is performed under general anaesthesia with headlight illumination and loupe magnification. With the mouth held open by a small gag the tongue is retracted antero-medially, the floor of the mouth is infiltrated by 5 ml of Mepivacaine 25 mg/ml + Adrenaline 5 mcg/ml just below the oral mucosa. The duct is identified and cannulated with a salivary probe (Bowman probes, Karl Storz, Tuttlingen, Germany). An oblique incision is made near the papillar region of the Wharton's duct, along the floor of the mouth toward the second molar. Once the mucosa is parted, the loose areolar tissue is dissected, first using sharp-tipped scissors and then smooth-tipped scissors, medially to the internal edge of the sublingual gland, which is rotated laterally to expose Wharton's duct. The horseshoe-shaped lingual nerve is easily identified running obliquely from the tongue, passing under the duct then ascending medially through the tail of the sublingual gland over the Wharton's duct to run below the constrictor muscles on to the infra temporal fossa. The lingual nerve is mobilized from the duct and retracted medially to visualize the stone in the gland hilum. The hilum of the gland is moved upward with an external finger pressure to the submandibular gland area. An incision is made over the calculus and the stone delivered by a microelevator or a Freer elevator (Martin, Tuttlingen, Germany) (Fig. 2). A submandibulotomy is performed in the case of intraparenchymal stones. The cavity is then irrigated with saline to clear debris. Videosialoendoscopy (1.2 mm, Nahlieli

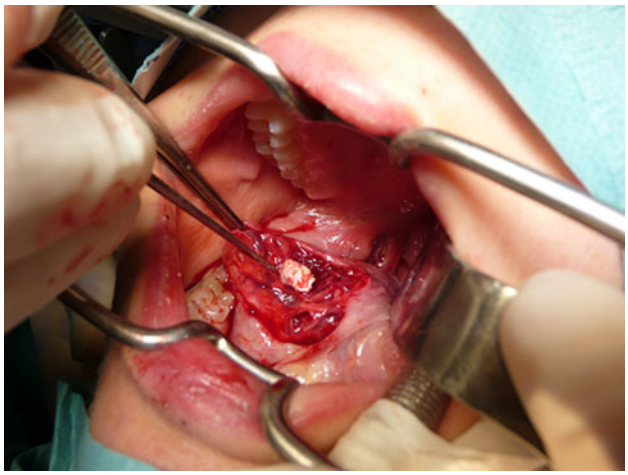

Fig. 2 Removal of the stone in the hilar region

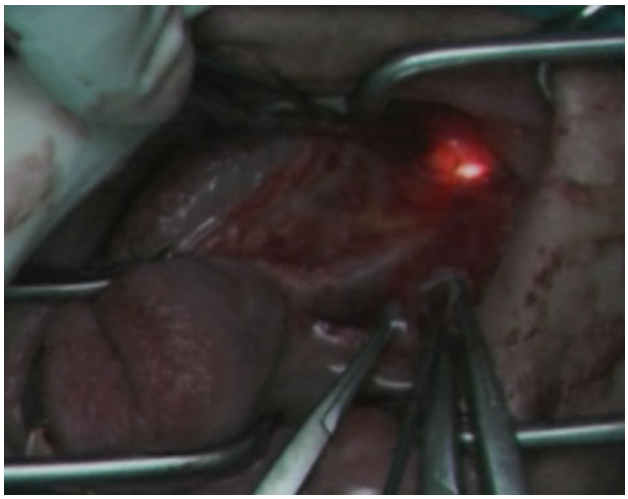

Fig. 3 Sialoendoscopic lighting as a guide during transoral removal of the submandibular calculi

sialoendoscope, Karl Storz Co., GmbH, Tuttlingen, Germany) may be performed for two purposes: to better locate the position of the stone in the hiloparenchimal area before incision (Fig. 3) or to check for any residual intraparenchymal calculi through the hilar surgical incision. Finally, a net of haemostatic and antimicrobial fibrillar surgical (Tabotamp, Johnson & Johnson Medical Limited, Gargrave, Skipton, UK) is positioned over the hilar opening to avoid the risk of stricture or stenosis. In the case of ostial stenosis, the distal third of the duct can be rehabilitated by making an axial incision and inserting a 6F salivary polymeric stent (Optimed, Ettlingen, Germany) or a 14–20 G Venflon tube (Artsana, Grandate, Italy) attached to the oral floor with a resorbable suture. The stent is usually removed after 2 weeks. The wound is irrigated with antibiotic solution (rifampicin) and the oral floor sutured using resorbable stitches (3.0 Vicryl). All the patients received antibiotic therapy (ceftriaxone) for 1 week after the operation; steroids were also administered in the case of oedema of the oral floor.

Outcome

All the patient underwent a postoperative clinical evaluation 1 week, 3 weeks and 1 year after the surgical procedure. Early and late postoperative complications were evaluated (Table 1). All the patients were telephonically interviewed to evaluate the outcome and in addition US evaluation was proposed to all patients with a minimum follow-up time of 36 months. The US features assessed were the presence of residual stone, gland size, duct dilatation and the vascularization of the parenchyma.

Statistical analysis

Association between recurrence of stones and previous treatment was tested by Fisher's exact test, while symptom-free survival curves were drawn according to Kaplan–Meier methodology. Difference between two groups (patients with and without previous treatment) was tested with the log-rank test. *P* value of 0.05 was considered significant. Data processing and all statistical analyses were performed with SAS software (version 9.1; SAS Institute, Cary, North Carolina).

Results

The size of the stones, as evidenced by US, ranged between 7 and 25 mm (mean 11, median 10 mm). Forty-six patients had a hilar stone (mean 9.8 mm, median 10 mm), 34 a hiloparenchymal stone (mean 12.6 mm, median 12 mm) and 4 an intraparenchymal stone (mean 10.8 mm, median 11 mm). The stone was located in the right submandibular gland in 41 patients, and in the left submandibular gland in 43 patients. The duration of the obstructive symptoms before surgery ranged between 4 and 30 years (mean 51, median 20 months). During this period about 70% of patients showed from 1 to 6 infective episodes (76% between 2 and 4).

The calculus was successfully removed in all but one of the patients; the only failure was related to an intraparenchymal stone. A videoendoscopic-assisted procedure was necessary in only eight patients to check, through the hilar opening, for any residual parenchymal calculi (6 patients) or to better locate, through the papillar ostium, an intraparenchymal calculi (2 patients).

As expected, early sequelae were observed in 63 patients (75%) and consisted of a mild and transitory swelling of the gland (23 patients), variable gland swelling and oedema of the floor of mouth (23 patients), tingling of the tip of the tongue (16 patients) and lingual nerve injury (1 patient)

Table 1 Surgical and post-surgical parameters of patients who underwent transoral removal of hilo-parenchimal submandibular stones

	n	%
Gender		
Male	51	60.7
Female	33	39.3
Previous treatments		
No	66	78.6
Shockwave lithotripsy	18	21.4
Side		
Right	41	48.8
Left	43	51.2
Site		
Hilar	46	54.8
Hilo-parenchymal	34	40.5
Intraparenchymal	4	4.8
Results		
Success	83	98.8
Failure	1	1.2
Early complications		
No	21	25.0
Swelling only	23	27.4
Swelling and		
Oedema	23	26.2
Tingling	16	19.1
Lingual nerve injury	1	1.2
Late complications		
No	75	89.3
Oral mycosis	4	4.8
Tingling	3	3.6
Ranula	1	1.2
Hilar stenosis	1	1.2
Subjective evaluation		
No symptoms	71	84.5
Recurred obstructive symptoms	12	14.3
Recurred infections	1	1.2
Residual stones		
No	68	81.0
Yes	16	19.0
Post treatments		
No	74	88.1
Endoscopy	6	7.1
Endoscopy and		
Other transoral removal	1	1.2
Sialoadenectomy	1	1.2
Shock wave lithotripsy	1	1.2
Shock wave lithotripsy	1	1.2

(Table 1). Late complications were observed in nine patients (11%) and were represented by oral mycosis (4 cases), persistence of tingling of the tip of the tongue (3 cases),

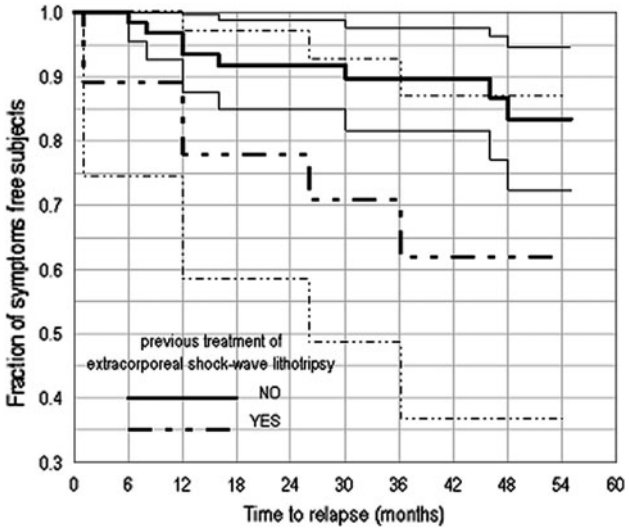

Fig. 4 Fraction of symptoms free subjects as a function of time to relapse (Kaplan–Meier survival curve with 95% CI)

hilar stenosis (1 case) and ranula (1 case). None of the patients had a permanent lingual nerve injury.

The patients were followed up for 12–74 months (mean 45.7, median 52): 71 were symptom-free, 12 had recurrent obstructive symptoms and signs, and one had recurrent infections without evidence of any obstructive cause. Overall, on US examination, a recurrent stone was observed in 16 patients. The size of the recurred stones varied from 2 to 6 mm and they were located in the main duct (6 patients), in the hilum (8 patients) and in the parenchyma (2 patients). The four asymptomatic patients and two symptomatic patients with residual calculus did not undergo any further procedure; of the remainder nine patients had the stone successfully removed thanks to combined modalities (sialoendoscopy, extracorporeal shock wave lithotripsy, further transoral removal) and one patient had his gland removed (Table 1).

At 12 months from the operation 90% (CI 95%: 84–97%) of the patients were symptom-free, this percentage decreased to 79% (CI 95%: 68–89%) at 48 months from the operation. The symptom-free time distribution was statistically different (Log-rank test: $p = 0.0101$) in patients with and without previous treatment of extracorporeal shock-wave lithotripsy (Fig. 4). Also, the presence of previous treatment was more frequently associated with recurrence of stones (Fisher test: $p = 0.0092$). No other statistical association was found between recurrence of the stone and any of the variables evaluated and shown in Table 1.

53 out of 84 patients had a minimum follow-up period of 3 years: 37 were clinically and ultrasonographically assessed, 16 answered to telephonic interview. 29 of the 37 (78%) patients were clinically and US essentially normal. The US features of the patients are described in Table 2; in particular

Table 2 Ultrasonographic features at 3 years follow up evaluation

	n	%
Residual stones		
No	29	78
Yes	8	22
Gland dimension		
Normal	27	72
Increased	5	14
Decreased	5	14
Ductal dilatation		
No	24	65
Hilar	7	19
Intraductal	6	16
Vascularisation		
Normal	29	78
Decreased	7	19
Increased	1	3

a normal vascularization was observed in 29 patients, a decreased vascularization in seven patients and an increased vascularization in one patient.

The duration of the hospitalization was one night stay for 55 patients, two nights for 23 patients and three or more nights for 6 patients.

Discussion

The general public desire for minimally or less invasive techniques has favoured in recent years the development of conservative and gland-preserving techniques for the management of salivary gland calculi.

The present study specifically investigated the outcomes of transoral removal of large hiloparenchymal submandibular calculi in a series of 84 patients. Successful stone retrieval was achieved in all but one of the patients. This result is in line with other results [2, 12, 15, 17, 18]. The only failure was represented by intraparenchymal calculi, very adherent to gland tissue. It has been shown that risk of failure increases with non palpable (intra parenchyma) stones; in contrast palpable stones can be reliably removed by the intra-oral technique [14]. Preoperative assessment (manual palpation and US location of stone) is important in the context of informed consent.

A videoendoscopic-assisted inspection of the hilar cavity has been advocated [2, 12] and is performed if it is suspected some stone fragments have been retained after the removal of the main stone. These fragments could facilitate new stone formation if not removed. According to our experience, the presence of a sialoendoscopic unit is useful during this surgical procedure not only to check for residual calculi but also to help the surgeon in the removal of deep intraparenchymal calculi.

The postoperative sequelae were minor and transitory in nature. A limited number of patients (5.9%) had persistent complications such as tingling of the tip of the tongue (3 patients), hilar stenosis (1 patient) and ranula (1 patient) but is compensated by the retention of a functioning gland in 98% of cases. An adaption of the intra-oral technique has been proposed where a portion of the proximal sublingual gland is removed [19]. In our experience this increases the risk of ranula formation and is unnecessary.

Recurrence of stone was observed in 16/84 patients but was symptomatic (obstruction/infection) in only 13. In these patients, secondary procedures (minimally invasive) were successful in eliminating symptoms and preserving the affected gland. Traditional sialadenectomy was only required in one case (1/84). The recurrence occurred mainly in the first 12 months. It is possible that the recurrence of calculi in the brief period after surgery is due to the persistence of micro debris (not evidenced by US) in the gland parenchyma, and this condition was more commonly observed in patients who previously underwent shock wave lithotripsy. In fact, an increased risk for recurrence was observed in patients who were previously treated by extracorporeal shock wave lithotripsy compared with naïve patients. Once again this may relate to dispersal of micro calculi into the adjacent soft tissues. Consequently, the treatment of choice for large stones should be transoral removal.

US and colour US were performed in patients with a minimum postoperative follow-up of 36 months: the restoration of the normal appearance of the ductal system and gland parenchyma was observed in most of the cases examined, confirming the histopathological and scintigraphic assumption that obstructive sialadenitis is a reversible condition [15]. As US is unable to adequately evaluate microliths less than 1.5 mm [20] a postoperative videosialoendoscopic check of the treated gland should be done, but this is only sometimes possible due to patient, time and cost factors.

Conclusion

Transoral removal of large (>7 mm) hiloparenchymal submandibular calculi is a safe, effective, conservative surgical procedure; it is mainly performed as a one night hospital stay procedure with minor postoperative discomfort for the patient [21]. Videoendoscopic assistance is useful during this procedure as it may influence the success of the procedure in particular cases such as deep and residual intraparenchymal calculi. An adequate preoperative clinical and

ultrasonographic evaluation should be always done to exactly locate the stone and to minimize the failure risk. Transoral removal of calculi under US monitoring has been recently proposed [22], but in our experience this approach does not add substantial improvement in the management of such calculi as the sialoendoscopic check is more useful in guiding the hilar or parenchymal surgical incision. A long-term clinical and statistical study will confirm the real risk for recurrence of calculi after this procedure and the residual role of sialadenectomy in patients undergoing modern minimally invasive and gland preserving techniques for calculi [21].

Conflict of interest I declare that all the authors have played a role in data collection, analysis and writing of the paper; I also declare that all authors have read and approved the paper. I certify that all my affiliations with or financial involvement, within the past 5 years and foreseeable future (e.g., employment, consultancies, honoraria, speakers bureau, stock ownership or options, expert testimony, grants or patents received or pending, royalties, donation of medical equipment) with any organization or entity with a financial interest in or financial conflict with the subject matter or materials discussed in the manuscript are completely disclosed.

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