The Laryngoscope

Treatment Strategy of Hilar and Intraglandular Stones in Wharton's Duct: A 12-Year Experience

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Objectives/Hypothesis: To suggest a strategy for transoral removal of hilar and intraparenchymal submandibular stones.

Study Design: Retrospective case series.

Methods: Retrospective evaluation was performed for 514 consecutive patients with hilar and intraparenchymal submandibular stones treated via endoscopy-assisted surgery from January 2006 to June 2018. Three patients had bilateral stones. The stones were classified as: hilar (type I), posthilar (type II), intraparenchymal (type III), and multiple stones (type IV).

Results: The affected glands included 311 with type I, 84 with type II, 65 with type III, and 57 with type IV stones. Stones were successfully removed in 478 glands (92.5%, 478/517). Main treatment techniques included hilum ductotomy in 311 glands, intraparenchymal ductotomy in 68, submandibulotomy in 14, intraductal retrieval in 74, and hilum ductotomy accompanied by intraductal retrieval in 11. At a mean 40-months follow-up of 478 successful cases, clinical outcomes were good in 425, fair in 27, and poor in 26 glands. Postoperative sialograms in 75 stone-free patients were categorized as: type I, normal (n = 6); type II, ectasia or stenosis in the main duct and no persistent contrast on functional films (n = 44); type III, ectasia or stenosis in the main duct and mild contrast retention (n = 15); and type IV, poor shape of the main duct and evident contrast retention (n = 10). Postoperative sialometry of 32 patients revealed no significant differences of the gland function between the two sides.

Conclusions: Appropriate use of various endoscopy-assisted approaches helps preserve the gland and facilitates recovery of gland function in patients with different depths of hilo-parenchymal submandibular stones.

Key Words: Sialolithiasis, submandibular gland, sialendoscopy, stone removal, gland function.

Level of Evidence: 4

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INTRODUCTION

Approximately 80% to 90% of sialolithiasis occurs in the submandibular gland (SMG), and the most frequent sites of SMG stones are the distal duct and hilum.[1–3] It is not a major surgical problem to remove stones situated in the distal or middle part of Wharton's duct, but extracting stones located in the proximal ducts, especially in the deep hilum or intraglandular ducts, is more challenging.[1] Since the introduction of endoscopy, the deeply located stones can be successfully removed in 80% to 90% of cases.[4,5] Notwithstanding, there still exists a small group of patients with deep hilum or intraparenchymal stones that need to undergo submandibulectomy, due to the considerable difficulties of surgery, poor gland function after stone removal, or postoperative complications. [2] Up to date, our center has used sialendoscopy for >12 years and has treated >600 patients with deep hilo-parenchymal submandibular stones. The purpose of this study was to evaluate these patients who underwent endoscopy-assisted transoral removal of deep hilar and intraparenchymal SMG stones, so as to suggest a positioning and treatment strategy for these intractable stones.

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MATERIALS AND METHODS

Subjects

From January 2006 to June 2018, 514 consecutive patients with deep hilar and intraparenchymal stones in Wharton's duct (including three with bilateral stones) underwent endoscopy-assisted transoral removal at our center. The diagnosis was verified by one or a combination of ultrasonography (US), spiral computed tomography (CT), and sialography. Proximal stones that did not touch the submandibular gland were excluded. There were 269 males and 245 females. Their ages ranged from 9 to 86 years (median = 38 years). The clinical records relative to stones sites, operation methods, and complications were retrospectively reviewed.

Surgical Approaches

There were 509 patients operated on under local anesthesia on an outpatient basis, and the remaining five were operated on under general anesthesia. A Laduscope T Flex PD-HS-0250 endoscope (PolyDiagnost, Pfaffenhofen, Germany) was used.

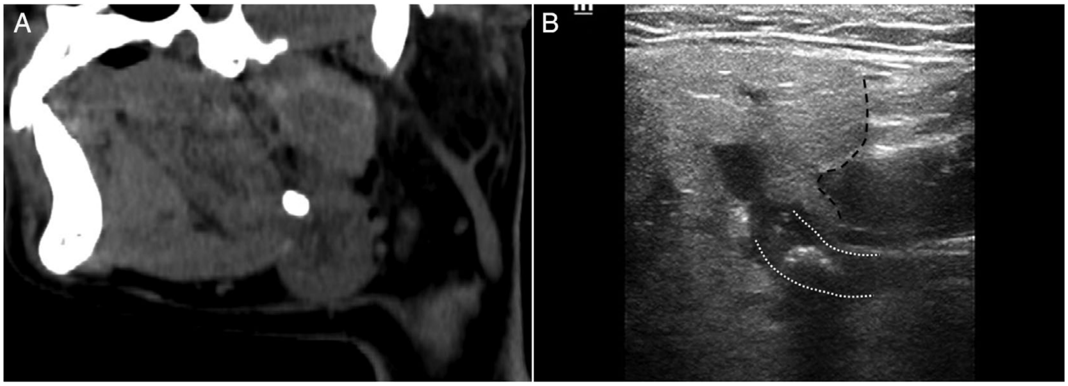

Fig. 1. (A) A hilar stone located just at the hilum of Wharton's duct. (B) The ultrasonography showed the stone at the hilum region (white dotted line) of the main duct. Note the antierior boundary of the gland (black dotted line) (type I).

The hilum was specified as the site where the duct entered the gland and coursed downward lateroposteriorly, which was initially detected by ultrasonography (US) or spiral CT, and was further verified by sialography if necessary. Based upon these imaging modalities, the stones were classified into four types: 1) hilar stone: at the hilum or proximally with a distance ≤0 mm from the hilum (Fig. 1), 2) post-hilar stone: intraglandular stone with a distance of 0 to 5 mm proximally from the hilum (Fig. 2), 3) intraparenchymal stone: intraglandular stone with a distance of ≥5 mm proximally from the hilum (Fig. 3), and 4) multiple stones: concomitant hilar and intraglandular stones.

The treatment methods included:

1. Hilum ductotomy: After the hilum stone was positioned by endoscopy and palpation, the oral floor was elevated by an assistant, and a 2- to 3-cm incision was made in the oral floor longitudinally along the proximal duct. Then, the hilum was incised at the precise site for stone removal. Particular care was taken to avoid injury to the lingual nerve and accompanying veins.
2. Intraparenchymal ductotomy: After exposure of the hilum and incision of the gland capsule, the intraglandular duct was traced proximally till the stone was detected, then the duct was incised to extract the stone.
3. Submandibulotomy: When the stone was located excessively far from the hilum but with a marked proximity to the gland capsule, the gland was incised accordingly to remove the stone.

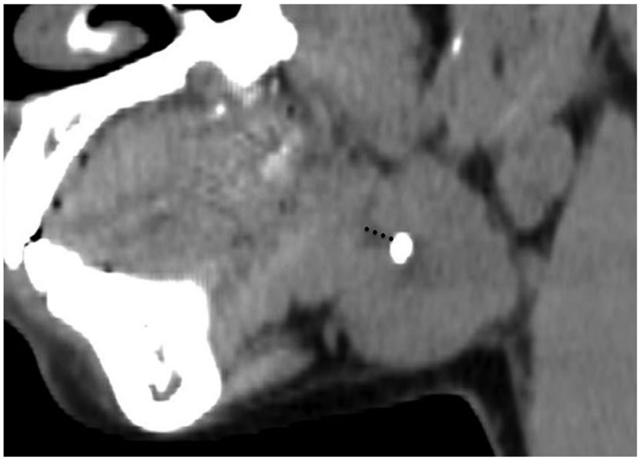

Fig. 2. A post-hilar stone impacted 4.8 mm (dotted line) proximal to the hilum (type II).

4. Basket retrieval: This method was used mainly for mobile small stones. It might be used as a primary approach for single stone cases, or as an assistance for multiple stone cases.

After removal of the main stone, the entire duct was reexplored for remnant stones or mucus plugs. The ductal incision at the hilum or intraglandular duct was routinely left open. A 4-Fr angiocatheter was used as a stent in cases where the proximal duct was severely damaged. Antibiotic treatment with amoxicillin or cefaclor was administered for 3 to 7 days. Postoperatively, patients were counseled to avoid sialogogues and spicy food. After removal of the sutures and stents, self-massage of the gland and sialogogues were advocated.

Follow-up Schedule

Patients who could not return to the clinic were followed up through telephone calls or mailed questionnaires. Patients who returned to the clinic underwent a clinical evaluation, including clinical manifestation, sialography, and sialometry. As for sialography, appearance of the ductal system on filling films and 5-minute evacuation films was analyzed independently by two experienced oral radiologists who reached a consensus through discussion. Submandibular saliva was collected by a 6-gauge scalp needle coated with a plastic outer sheath, which was sucked onto the ductal orifice and connected to a saliva storage device. Resting and stimulated flow rates were measured respectively for 5 minutes. Total saliva flow in 10 minutes was calculated by adding the resting and stimulated saliva flows.

Clinical outcomes of success cases were scored as good (asymptomatic with clear saliva), fair (with occasionally mild symptoms that can be alleviated autonomously), and poor (with persistent symptoms, severe side effects, or gland atrophy).

Statistical Analysis

Statistical analyses were conducted using SPSS 25.0 (IBM, Armonk, NY). The quantitative parameters were expressed as mean ± standard deviation, and the Student t test was used to compare differences between the affected and unaffected control glands if the data coincided with the Gaussian distribution. Otherwise, the range and median of the values were provided, and Wilcoxon rank sum test was used for comparison.

The study design was approved by the institutional review board of our school (PKUSSIRB-201839154), and all participants signed an informed consent.

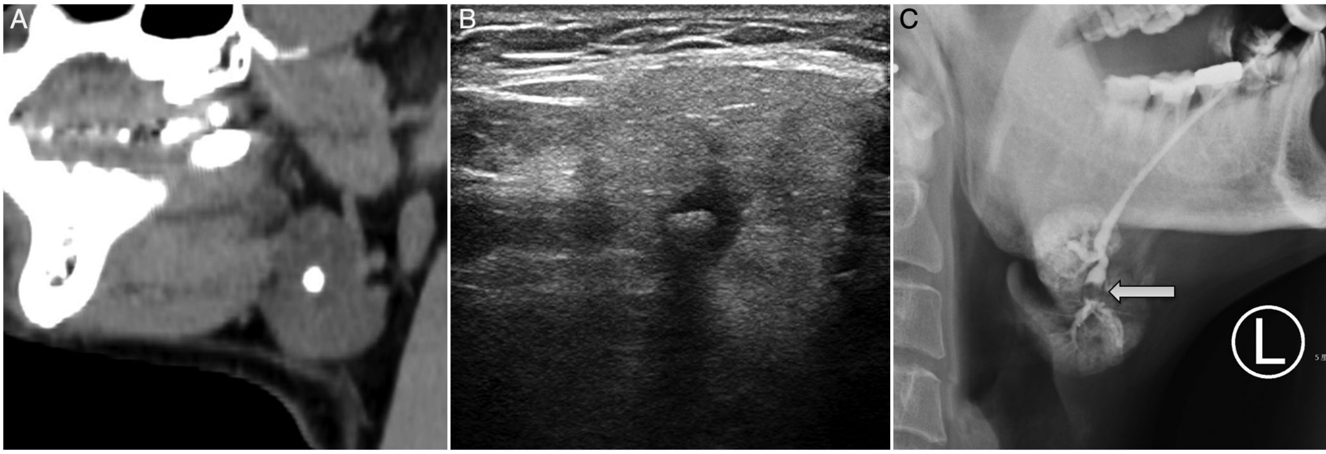

Fig. 3. (A) Sagital computed tomography view and (B) ultrasonography showed a intraparenchymal stone in Wharton's duct. (C) On sialogram of the left Wharton's duct (L), the stone appeared as a filling defect (white arrow) in the intraglandular duct (type III).

RESULTS

Treatment Results

The size of stones varied from 3 to 25 mm, with a mean of 7.4 mm. The 517 glands comprised 311 glands with type I, 84 glands with type II, 65 glands with type III, and 57 glands with type IV stones. The stones were successfully removed in 478 glands (92.5%, 478/517). Success rates varied significantly in different stone types as follows: 98.7% for type I, 95.2% for type II, 55.4% for type III, and 96.5% for type IV stones. In 39 glands, the removal of stones failed due to the patients' intolerance (n = 11) or excessively deep stones (n = 28) (Table I).

The main treatment methods applied included hilum ductotomy in 311 glands, intraparenchymal ductotomy in 68 glands, submandibulotomy in 14 glands, intraductal retrieval in 74 glands, and hilum ductotomy accompanied by intraductal retrieval in 11 glands (Table II).

Treatment success was achieved via one endoscopic procedure in 466 glands. In five glands, the first procedure failed due to the patients' discomfort or severe inflammation, and the stone was removed via a second procedure 1 to 20 months later. In the remaining seven glands, the stone moved distally after the first intervention and was removed 1 to 12 months later.

During surgery, ductal breakage occurred in two glands with huge stones, and the remnant proximal duct was ligated. After surgery, all patients complained of mild to moderate pain with a duration of 3 to 7 days. Eleven patients developed temporal lingual nerve injury, which was relieved within 1 month.

Follow-up Results

There were 514 patients followed up for an average period of 40 months (range = 3 to 120 months) after surgery. Among the 478 successful cases, four had swelling symptoms and accepted a second endoscopy for ductal dilatation. Two glands that had experienced ductal ligation were asymptomatic during the follow-up, and US confirmed gland atrophy. Seven developed ranula 1 to 22 months (mean = 8 months) postoperatively and underwent an uneventful sublingual gland excision. Thirteen patients had recurrent stones 6 to 58 months (mean = 25 months) postoperatively and experienced a second lithectomy. Twenty-seven patients had occasionally mild symptoms. The remaining 425 glands were asymptomatic with favorable function. Overall, the clinical outcomes were good in 425 (88.9%, 425/478) of the glands, fair in 27 glands (5.6%, 27/478), and poor in 26 glands (5.4%, 26/478). Among the 39 failed cases, six were asymptomatic, five underwent submandibulectomy, and the remaining 28 had occasionally mild symptoms.

TABLE I.
Surgical Treatment Results of 517 Glands Relative to Four Different Stones Types.

	Hilar	Post-hilar	Intraparenchymal	Multiple	Total
Success	307	80	36	55	478
Failure	4	4	29	2	39
Total	311	84	65	57	517
Success rate	98.7%	95.2%	55.4%	96.5%	92.5%

TABLE II.
Different Surgical Methods Applied for the 478 Successful Cases.

	Method 1	Method 2	Method 3	Method 4	Method 1 + 4	Total
Hilar	272	0	0	35	0	307
Post-hilar	0	55	8	17	0	80
Intraparenchymal	0	13	6	17	0	36
Multiple	39	0	0	5	11	55
Total	311	68	14	74	11	478

Objective Tests of Submandibular Gland Function for Stone-Free Patients

The 26 patients with poor outcomes were excluded from objective tests, and 377 asymptomatic patients did not return to the clinic. The remaining 75 patients (33 males and 42 females) underwent sialography 3 to 84 months (mean = 12 months) after surgery. The stone size averaged 7.7 mm. Forty-eight patients were asymptomatic with clear saliva flow of the affected glands. The remaining 27 patients had occasional swelling. Postoperative sialographic appearances were categorized into four types: type I, normal (n = 6); type II, ectasia or stenosis in the main duct and no persistent contrast on functional films (n = 44); type III, ectasia or stenosis in the main duct and mild contrast retention (n = 15), and type IV, poor shape of the main duct and evident contrast retention (n = 10). The 27 symptomatic patients included one with type I, 15 with type II, five with type III, and six with type IV sialograms.

Among the 75 patients, 32 asymptomatic patients (14 males and 18 females) underwent sialometry examination at the meantime. They included three with type I, 19 with type II, four with type III, and six with type IV sialograms. Follow-up duration of them ranged from 3 to 84 months (mean = 13 months). The stone size averaged 7.1 mm. Wilcoxon rank sum test did not show significant differences for the following values: 5-minute resting saliva flow between the affected gland (0.00–1.75 g, median = 0.15 g) and the control gland (0.01–1.39 g, median = 0.30 g; $z = -1.141$, $P = .254$); 5-minute stimulated flow between the affected gland (0.01–3.48 g, median = 0.64 g) and the control gland (0.05– 2.96 g, median = 0.84 g; $z = -1.440$, $P = .150$); 10-minute total flow between the affected gland (0.02–5.23 g, median = 0.67 g) and the control gland (0.08–4.35 g; median = 1.06 g; $z = -1.589$, $P = .112$). However, for the affected gland, the 5-minute stimulated flow was significantly higher than the 5-minute resting flow ($z = -4.413$, $P = .000$).

DISCUSSION

Animal studies showed that submandibular glands could regenerate following severe atrophy and secrete normal amounts of saliva.[6,7] Clinically, submandibular gland function affected by sialolithiasis had been proven to improve greatly after surgical sialolithectomy.[8] Submandibular gland resection nearly halved the unstimulated salivary flow.[9] Therefore, the submandibular gland should be preserved to the largest extent, even for those with deep hilo-parenchymal stones.

Site, shape, size, number, and quality of materials dictated treatment options. Moreover, the treatment results were influenced by the patients' potential of toleration and cooperation in the situation of local anesthesia. Capaccio et al. classified hilo-parenchymal submandibular stones into three types: hilar (at least two margins were detectable during palpation), hilo-parenchymal (only the distal margin was detectable during palpation and the remaining margins were covered by glandular tissue) and intraparenchymal.[4] In view of the uncertainty and confusability of clinical palpation, the hilar and intraparenchymal stones in the present study were classified into four subtypes, according to the stone site relative to the hilum. The hilar subtype might include the first two types described by Capaccio et al.[4] Primarily, hilum ductotomy was applied for extracting these stones, gaining a success rate of 98.7%, which was comparable to 98.5% in Capaccio et al.'s report of 479 patients.[3] In the report of Schapher et al., deep hilar stones involved stones in the hilar region or the adjacent parenchyma of the gland; however, further divisions of the intraparenchymal stones were not suggested.[9]

In previous reports, submandibulotomy was performed for intraparenchymal stones, which could not be exposed after duct incision to the hilum but were palpable within the adjacent gland.[2,4,9] In the present study, the intraparenchymal stones were classified into two subtypes: post-hilar and intraglandular stones, and both could not be palpated manually and were removed mainly by method 2 and 3. Method 2 and 3 could be regarded as two different variations of submandibulotomy. The success rates of the post-hilar and intraglandular stones were 95.2% and 55.4%, respectively. It was obvious that deeper stones gained lower success rate. Method 2 applied in the present study was similar to extended duct slitting described by Schapher et al., and could only be performed under good positioning of stones, favorable vision control, and cooperation among the operator, assistant, and patient.[9] Method 3 was selected when the stone was sited far from the hilum but with a marked proximity to the gland capsule, and the hilum duct was left intact. These two methods were confirmed to be reliable treatment options for 63 type II and 19 type III cases, which was to date the largest patient group with submandibular deep hilar calculi. For smaller hilar or intraglandular stones, basket retrieval was attempted. Ductal dilation was performed for cases with ductal stricture distal to the stone. Surprisingly, this method was confirmed to be effective in 35 type I, 17 type II, and 17 type III cases.

As for the glands with multiple stones, the success rate reached 96.5%. The relatively higher success rate could be explained by the fact that the large-sized stones located in the hilum might frequently dilate the intraglandular duct; hence, the relatively smaller intraparenchymal stones might discharge spontaneously or with the help of basket retrieval.

The significant anatomical structures complicating surgery included Wharton's duct and its accompanying veins, lingual nerve, posterior margin of mylohyoid, sublingual gland, hilar ductal branches, and accessory gland of the main gland. In Park et al.'s report, a mucosal incision was made along the lateral border of the sublingual gland, and the lingual nerve was retracted to the medial side. Postoperatively, all patients had mild paresthesia of the tongue for 3 months.[10] In the technique described by Combes et al., the oral floor incision was made along the medial border of the sublingual gland, and the sublingual gland was retracted laterally after identification of the duct and lingual nerve. Consequently, 6% of patients were left with persistent modest tingling sensation of the tongue.[11] Our technique was similar to that of Combes et al., and the percentage (2%) of temporary tongue numbness was significantly lower than the former two reports. This can be explained by the following factors. First, the lingual nerve was identified and retracted only in some of these cases. Second, meticulous hemostasis was accomplished by digital pressure and suturing, whereas electrocoagulation was reserved only for cases with severe hemorrhage. All these helped minimize injury of the nerve and duct.

The techniques described by Schapher et al.[9] and Zenk et al.[2] involved incision of the submandibular duct and oral mucosa from the ostium to the stone site. Then, the incised duct was sutured to the floor of the mouth to create a proximal neo-ostium. Capaccio et al. incised the mucosa similarly, whereas a limited ductotomy was performed over the stone.[4] Undoubtedly, an excessive incision of the mucosa and the duct carried higher risks of postoperative discomforts and ranula formation. In our cases, a 2- to 3-cm incision immediately over the stone was used for exposure of the hilum and intraglandular ducts, and postoperative swelling and pain subsided within 3 to 7 days. The six cases that had ductal occlusion or stenosis and seven cases with ranula formation were primarily our early cases. The percentage of recurrent stones (2.7%) was lower than the data reported by Capaccio et al. and Schapher et al.[4,9] This might have contributed to the endoscopic inspection and dilatation of the proximal ductal system.[12] In addition, four hilar stones and three intraglandular residual stones that were not removed during the initial surgery migrated distally to the middle part of the duct during the follow-up period. It could presumably be stated that endoscopic ductal dilation was beneficial for distal migration of impacted stones. For those patients who could not tolerate surgery well, or those with severe inflammatory scarring in the surgical area, endoscopic procedures should be terminated properly, with the expectation that future surgeries could be accomplished with success.[3,9]

The use of extracorporeal and intraductal shock wave lithotripsy (ESWL and ISWL) was reported by several authors.[13–15] It was generally acknowledged that the ESWL technology was primarily applicable to stones with a diameter < 8 mm.[13,14] Lafont et al. suggested ESWL as the first-line treatment for all parotid stones and posterior or intragland submandibular stones with few side effects.[15] Intraductal laser lithotripsy included holmium laser, erbium laser, and thulium laser, and the reported success rate was estimated to be 80% to 100%.[16–18] Recently, intraductal pneumatic lithotripsy were introduced with gratifying results.[14] Nevertheless, this equipment was expensive and was not available in our center.

Objective evaluations of salivary gland function mainly included sialography, scintigraphy, and sialometry.[5,19–21] In the present study, postoperative sialograms showed that 67% of cases had good function. However, in 92% of the patients, the main duct had ectasia and/or stricture of the hilum, which was significantly higher than the data (21%) reported by Woo et al.[22] Abnormal ductal shape might develop from stone formation or surgical scarring, and might cause recurrence of symptoms. In the present study, sialometry was applied as a substitute for quantitative evaluation of gland function.[19] Despite the marked differences in individuals, statistical analysis showed no significant differences between the affected gland and control side. This confirmed that, even with poor ductal shape and longtime stone occlusion, gland function could return to a high level. Notably, sialogogue, self-massage, and periodical intraductal infusion were helpful for this process and should be advocated, especially for those large-stone cases.[8]

It should be pointed out that, although the stone classification system and surgical techniques in the present study were suggested, longer-term practices were imperative to potentiate the safety and efficiency. Furthermore, ESWL or ISWL might be a promising option for deeply situated stones (type II and III). Moreover, both sialography and sialometry had limitations for evaluating gland function. In our follow-up results, there was not strict correlation between the clinical complaints and sialographic types. No final conclusions concerning gland function could be drawn when only sialograms were considered. Furthermore, sialometry was deficient due to variations of ostium and saliva viscosity.

CONCLUSION

Hilar and intraglandular submandibular stones can be classified as hilar, post-hilar, intraparenchymal, and multiple stones, on the basis of US, spiral CT, or sialography. These deeply situated stones can be removed by the following endoscopy-assisted techniques: hilum ductotomy, intraparenchymal ductotomy, submandibulotomy, and intraductal retrieval. Appropriate use of these techniques and meticulous postoperative manipulations help preserve the gland and promote recovery of gland function.

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