Sialoendoscopy-Assisted Sialolithectomy for Submandibular Hilar Calculi

Deng-Gao Liu, SMD, Lan Jiang, SMD,† Xiao-Yan Xie, SMD,‡*
Zu-Yan Zhang, DDS, PhD,§ Lei Zhang, SMD,‖ and
Guang-Yan Yu, PhD, DDS¶

Purpose: To assess the clinical effects of endoscopy-assisted sialolithectomy for submandibular hilar calculi.

Materials and Methods: The present study was undertaken in 70 patients with symptomatic stones in the hilum of submandibular glands who underwent endoscopy-assisted sialolithectomy from December 2005 through March 2011 in the Peking University School and Hospital of Stomatology. The operative data were analyzed retrospectively. All patients were followed periodically postoperatively. Submandibular gland function was investigated by postoperative symptoms, clinical examinations, sialography, and scintigraphy.

Results: Submandibular stones were successfully removed in 65 patients, with a success rate of 92.9%. Temporary lingual nerve injury occurred in 1 patient. Two patients developed ranulae and underwent an uneventful sublingual gland excision. During a mean follow-up of 23 months (range, 6 to 55 mo), 52 of 65 patients were symptom free, whereas 11 patients complained of occasional swelling of the affected gland at mealtimes and 2 patients developed a recurrent stone. Thirty patients underwent postoperative sialography. The sialographic appearances included 4 types: *1)* approximately normal; *2)* the main duct was significantly dilated at the hilum, but no persistent contrast was seen on the functional film; *3)* the main duct was significantly dilated in the hilar region, and persistent contrast was seen at the dilated hilum of the functional film; *4)* the main duct was dilated or strictured, and persistent contrast was seen on the functional film. Three of the 4 patients who underwent scintigraphy exhibited good function.

Conclusions: Sialoendoscopy-assisted sialolithectomy is a safe and effective gland-preservation technique for patients with hilar stones of the Wharton's duct.
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Sialolithiasis is the most frequent etiology of obstructive sialadenitis and occurs mainly within the ductal system of the submandibular glands. Submandibular stones in the distal portion of the duct can be removed by a straightforward intraoral procedure. However, it is difficult to remove large stones situated at the genu of the duct or within the gland transorally. Consequently, the affected gland is usually removed in its entirety together with the stones. To date, mounting evidence has shown that a significant percentage of affected submandibular glands has a normal histologic appearance, and the gland can regain its function after stones are removed.[1,2]

Sialoendoscopy is an innovative procedure that visualizes the lumen of the salivary ducts for the treatment of ductal diseases. With the development of minimally invasive techniques, small stones can be retrieved and removed directly under endoscopy.[1-3] In cases of larger and impacted stones that are located at the genu of the Wharton's duct, endoscopy-assisted transoral removal has been described by McGurk[4] Marchal,[5] and Nahlieli et al.[6] Since 2005, this tech-

Received from the Peking University School and Hospital of Stomatology, Beijing, China.

*Professor, Department of Oral Radiology.

†Resident, Department of Oral Radiology.

‡Resident, Department of Oral Radiology.

§Professor, Department of Oral Radiology.

‖Associate Professor, Department of Oral and Maxillofacial Surgery.

¶Professor, Department of Oral and Maxillofacial Surgery.

Address correspondence and reprint requests to Dr Liu: Department of Oral Radiology, Peking University School and Hospital of Stomatology, Beijing 100081, China; e-mail: kqldg@bjmu.edu.cn
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nique has been used in the authors' center for the removal of submandibular stones in the hilar region. This study aimed to assess the treatment effects, complications, and recovery of gland function in patients with submandibular hilar stones who underwent endoscopy-assisted sialolithectomy.

Materials and Methods

PATIENTS

From December 2005 through March 2011, 243 patients with submandibular gland calculus were treated at the Peking University School and Hospital of Stomatology using sialoendoscopy. Of these, 70 patients (38 men and 32 women) had large impacted calculi at the hilum of the Wharton duct and underwent endoscopy-assisted sialolithectomy. The clinical and operative data of these 70 patients were acquired and retrospectively analyzed. Their ages ranged from 13 to 67 years (mean, 34.4 yrs). Their main symptom was recurrent swelling of the affected gland at mealtimes, which persisted from 1 week to 9 years. The diagnosis was made by 1 or a combination of radiographic indicators, including cross-sectional mandibular occlusal films, lateral projections of the gland, and cone-bean computed tomography. The inclusion criterion was that the stones were situated at or proximal to the third mandibular molar region. Moreover, the stones were verified to be impacted at the hilum of the Wharton duct under endoscopic view. Cases with free hilar stones that were amenable to basket retrieval were excluded. The clinical outcomes were followed. Informed written consent was obtained from each patient. Approval for this study was obtained from the institutional ethics review board.

OPERATION PROCEDURES

Sixty-five endoscopic procedures were performed under local anesthesia on an outpatient basis and 5 were performed under general anesthesia. A Laduscope T Flex PD-HS-0250 endoscope (Polydiagnost, Pfaffenhofen, Germany) was used. This is a highly flexible, semirigid endoscope with a nitinol sheath. It is 80 mm long and has an outer diameter of 1.1 mm, a 0.4-mm working length channel, and a separate channel for irrigation. After the introduction of the endoscope, the main duct of the gland was explored under persistent irrigation. Small and mobile stones at the distal or middle part of the duct were removed by basket entrapment. Impacted hilar stones were then removed by an endoscopy-assisted sialolithectomy technique (Fig 1). After the stone was verified, a 2- to 3-cm incision was made in the floor mucosa according to the light transmitted from the endoscope. An assistant raised the floor of the mouth, with digital pressure in the submandibular triangle. The duct was isolated from the surrounding tissues, with particular care to avoid injury to the lingual nerve. Then, the hilum was incised at the precise location of the stone, and the stone was removed. Thereafter, the entire duct was re-explored for remnant stones or mucus plugs. The hilum was then sutured after a 4Fr angiocatheter had been inserted as a stent. This stent remained in situ for 1 to 2 weeks after surgery.

Antibiotic treatment with amoxicillin or cefaclor was administered for 7 days. Postoperatively, hydration was achieved by the patient drinking more than 2 L of water a day, and patients were counseled to avoid sialogogues and spicy food. After the stent and sutures were removed, frequent self-massaging of the gland and sialogogues were recommended.

FOLLOW-UP SCHEDULE

Patients who could not return to the clinic received telephone calls to assess symptom recurrence and changes in the size of the gland. In the clinic, patients underwent a clinical evaluation, which assessed the size and consistency of the affected gland, the presence of tenderness, the appearance of the ostium, and the amount and nature (clear or milky) of the salivary flow on massage. Sialography of the submandibular gland was performed with a water-soluble contrast agent, diatrizoate meglumine, using a closed intravenous catheter (22-gauge). After the catheter was introduced, 1.5 to 2 mL of contrast was injected carefully. Lateral views and a 5-minute emptying film were taken. The appearances of the main ducts, branch ducts, and parenchyma were analyzed. Two experienced oral-maxillofacial radiologists independently analyzed each case and reached a consensus by discussion.

Scintigraphic assessment was performed with the patient placed facing a digital gamma camera fitted with a high sensitivity collimator. Technetium pertechnetate (technetium 99m; 40 mBq; effective dose, 0.5 mSv) was injected intravenously and the uptake was measured by consecutive 1-minute anterior scans. Glandular secretion was measured from further scans 20 minutes after the patient had consumed citric acid, a sialogogue.

Results

In 68 cases, the hilar stones were found wedged in the ductal wall, attached by mucus plugs. The mucosa lining the neighboring duct had a matted appearance, with ecchymosis and engorgement of the blood vessels. In the other 2 cases, the hilar stones were found displaced in the periductal space through a perforation of the duct. Forty-six patients had single stones,

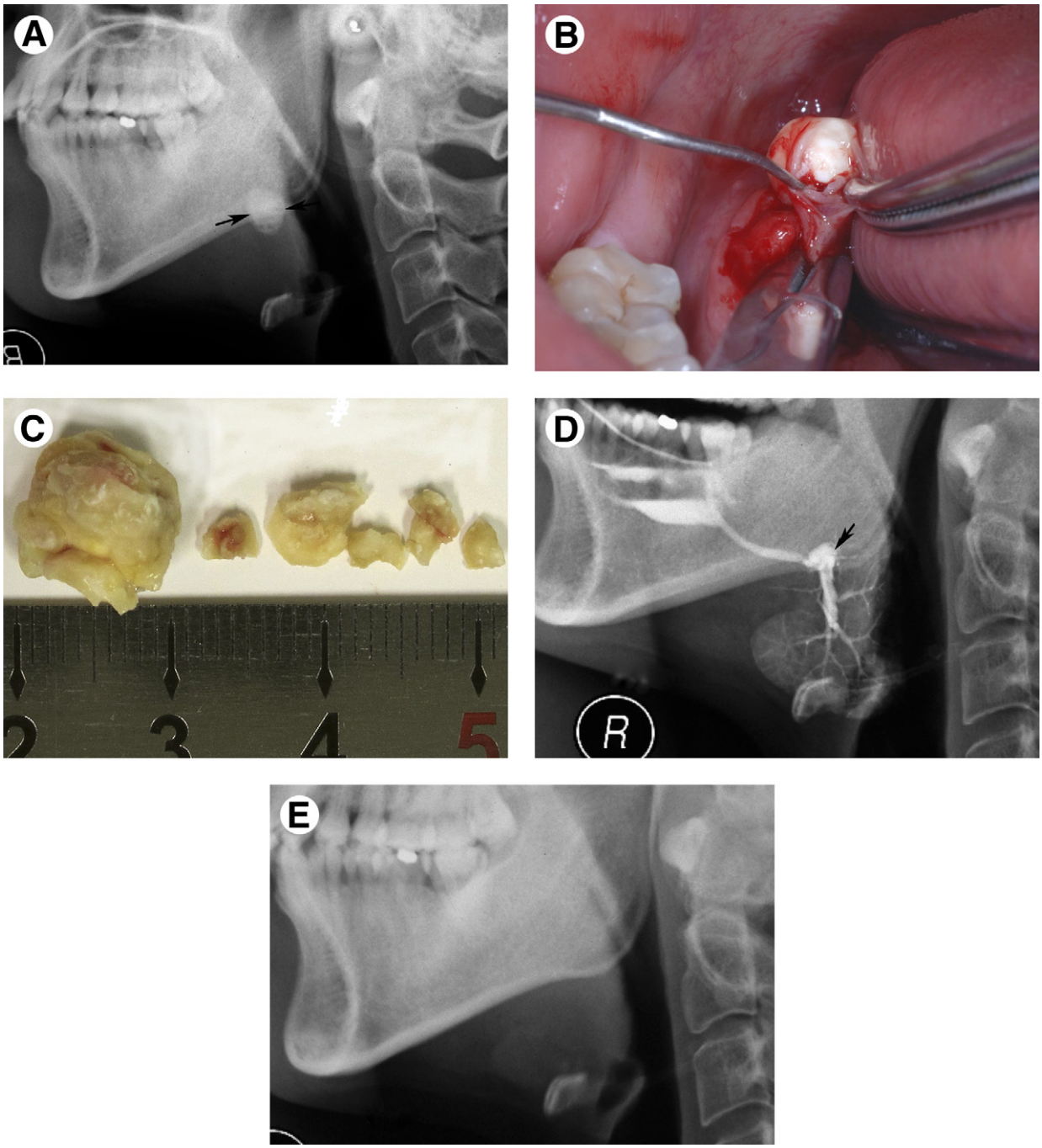

FIGURE 1. A 42-year-old woman with a 5-year history of sialolith in the right Wharton duct. *A,* Lateral view of the gland shows a large stone (*arrows*) superposed with the mandible. *B,* The stone was removed through an incision at the genu of the Wharton duct. *C,* Extracted stone fragments. Six-month follow-up sialograms show that the proximal duct exhibits significant dilatation (*arrow*) on the *D,* filling film, but no persistent contrast is opacified on the *E,* functional film.

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whereas the remaining patients had at least 2 stones. The size of the stones ranged from 4 to 15 mm.

In 61 patients (61/70, 87.1%), complete stone removal was accomplished (total removal). In 4 patients (4/70, 5.8%), the large hilar stones were removed, with residual stone fragments that persisted in the deeper branches (subtotal removal). Therefore, the overall success rate was 65/70 (92.9%). In 5 patients (5/70, 7.1%), the removal of the stone failed and gland dissection was suggested.

In the period immediately after the procedure, significant swelling of the affected gland was noted in all

patients because of irrigation. The swelling subsided spontaneously after a few hours. One patient developed temporary lingual nerve hypoesthesia for 1 month, which was caused by the clamping of forceps; this resolved after the administration of prednisone and vitamins B_1 and B_{12}. Two patients developed ranula 3, and 4 months postoperatively, they underwent an uneventful sublingual gland excision. No other major complications were noted in the follow-up period.

Patients were followed for a mean period of 23 months (range, 6 to 55 mo) after surgery. Of the 65 patients who underwent successful sialoendoscopy-assisted sialolithectomy, 52 (80.0%) patients remained asymptomatic, and 13 (20.0%) had mild obstructive or infective symptoms. In the latter group, 2 patients developed new stones in the distal part of the duct 6 and 24 months postoperatively. These were removed endoscopically without complications.

During the follow-up period, 32 patients from the initial cohort of 70 patients underwent further evaluation of their gland function. Their ages ranged from 17 to 61 years (mean, 35.5 yrs). The size of their stones ranged from 4 to 15 mm (average, 7.4 mm), and 12 patients had multiple stones. In these patients, 29 were free of stones, 2 had recurrent stones (described earlier), and the remaining patient had a residual stone fragment. Nineteen of 32 patients (18 free of stones and 1 with a residual stone) were symptom free, and clinical examination showed that their affected submandibular glands had a normal size and consistency, with a patent ostium and clear saliva on massage. The other 13 patients (11 free of stones and 2 with recurrent stones) had intermittent swelling, and clinical examination showed that their affected glands were relatively larger, with flocculent saliva on massage. Of these 32 patients, 1 underwent diagnostic endoscopy for severe swelling of the submandibular gland, which showed complete occlusion of the hilar duct. This symptom subsided gradually over the next 2.5 years. One patient was excluded for fear of an allergic reaction to contrast, leaving 30 patients who underwent postoperative sialography.

The sialographic appearances of the 30 patients could be classified into 4 types (Table 1): *1)* relatively normal (n = 10; Fig 2); *2)* the proximal duct exhibited significant dilatation and/or stricture, but no persistent contrast was visible on the functional film (n = 9; Fig 1); *3)* the proximal duct exhibited significant dilatation and/or stricture, and persistent contrast was evident in the hilar area of the functional film (n = 3; Fig 3); and *4)* the proximal duct was dilated and/or strictured with significant contrast retention on the functional film (n = 8; Fig 4). In total, 10 patients had ductal strictures and 17 patients had ductal ectasia at the hilar part of the Wharton duct on sialogram.

Table 1. SIALOGRAPHIC APPEARANCES IN 30 PATIENTS

Type	Symptomatic	Stricture	Ectasia	Total
1	2	0	0	10
2	2	4	8	9
3	1	2	2	3
4	6	4	7	8
Total	11	10	17	30

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Of 11 patients with swelling symptoms, 3 accepted endoscopic dilatation 6 to 18 months after stone removal for the Wharton duct stricture. Their symptoms subsided after the procedure. The other 8 patients accepted the mild discomfort without requesting further treatment.

Four asymptomatic patients underwent scintigraphic assessment (6 mo postoperatively for 3 patients and 18 mo postoperatively for the other). A time-activity curve showed that the bilateral glands had comparable function in 3 patients, whereas the fourth patient exhibited a decrease of contrast uptake and secretory function in the affected gland.

Discussion

Sialolithiasis is the most common cause of acute and chronic salivary gland infections. The most common site is the submandibular gland, where 80% to 90% of calculi are found; 5% to 10% of stones are found in the parotid gland; and a small number is found in the sublingual gland. The typical presentation is a painful swelling of the gland at mealtimes. The incidence of symptomatic salivary calculi has been estimated at 59 million/year and the prevalence is 0.45%.[1]

Calculi in the proximal duct or within the gland have always posed a therapeutic challenge. The traditional choice of sialadenectomy carries attendant risks, including injury to adjacent nerves and facial scarring, and is an invasive procedure. With the development of sialoendoscopy, several minimally invasive techniques have been reported, including extracorporeal and intracorporeal lithotripsy, basket retrieval, and transoral removal of the calculus.[7] An important principle that has encouraged these developments is that the secretory function of the gland can recover after stone removal. A histopathologic study has reported that a significant percentage of submandibular glands that were removed with a calculus had a normal histologic appearance.[1]

Despite their desirable results, these minimally invasive techniques have their limitations. The success rate of extracorporeal lithotripsy ranges from 40% in

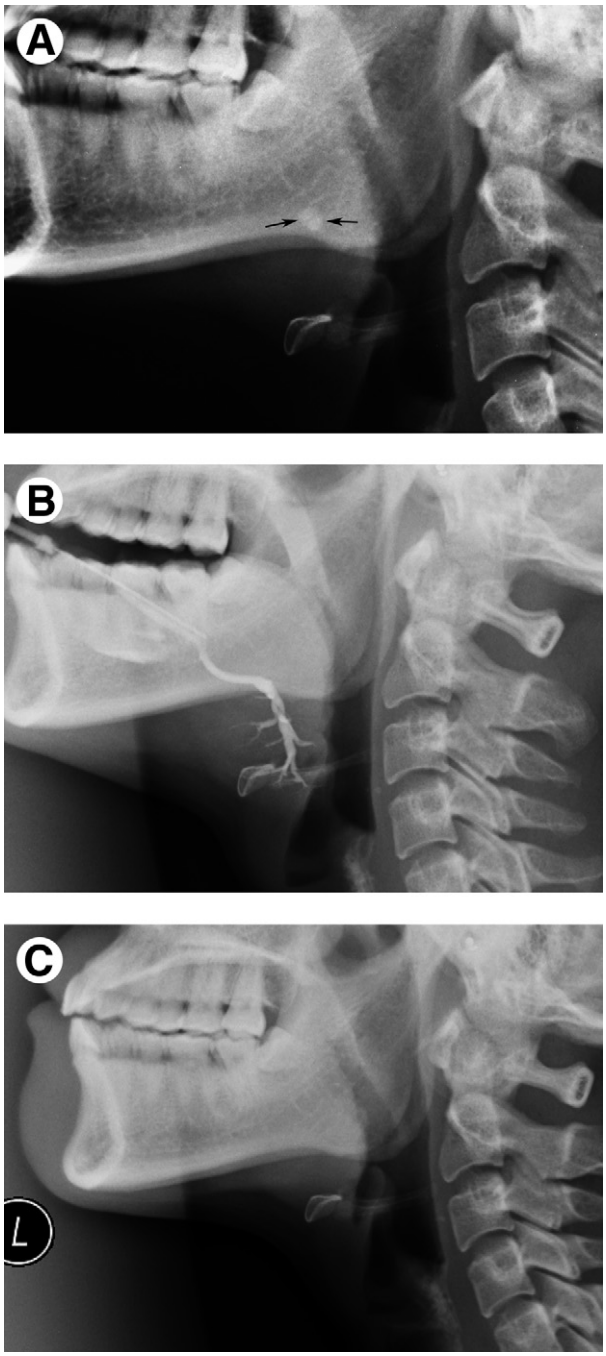

FIGURE 2. A 30-year-old woman with a stone in the left Wharton's duct. *A,* Lateral view of the gland shows a hilar stone (*arrow*). *B,* The filling film of the 7-month follow-up sialogram exhibits an approximately normal appearance of the ducts. *C,* No persistent contrast is opacified on the functional film.

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the submandibular gland to 75% in the parotid glands, with poorer results in cases of large stones. Intraductal laser fragmentation and basket extraction of stones are possible in only 80% of cases. The remaining 20% fail because of large stones and stenotic ducts, which prevent retrieval even if these stones can be captured.[2]

The technical details for the transoral removal of submandibular hilar calculi vary among investigators. In the approach described by Zenk et al,[8] the duct is

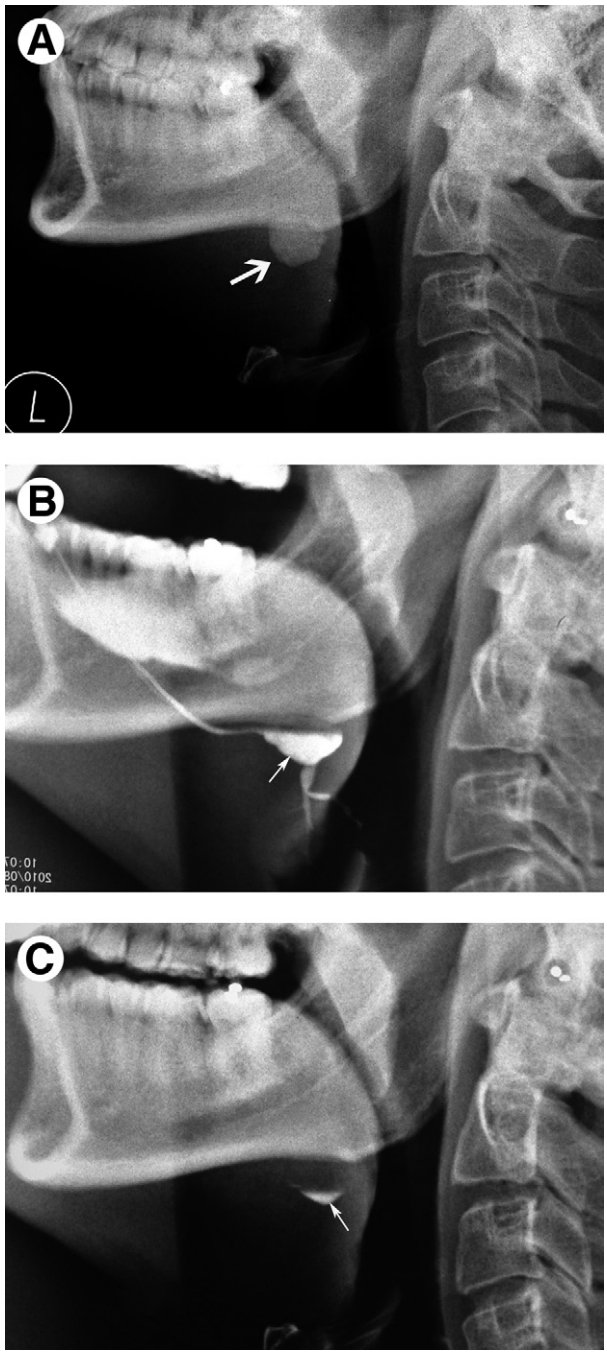

FIGURE 3. A 35-year-old man with a large hilar stone in the left Wharton's duct. *A,* Lateral film shows the hilar stone (*arrow*), which is approximately 1.5 cm. *B,* The filling film of the 18-month follow-up sialogram shows significant dilatation (*arrow*) at the hilar part of the duct. *C,* The emptying film shows mild retention of contrast (*arrow*) at the dilated part of the duct.

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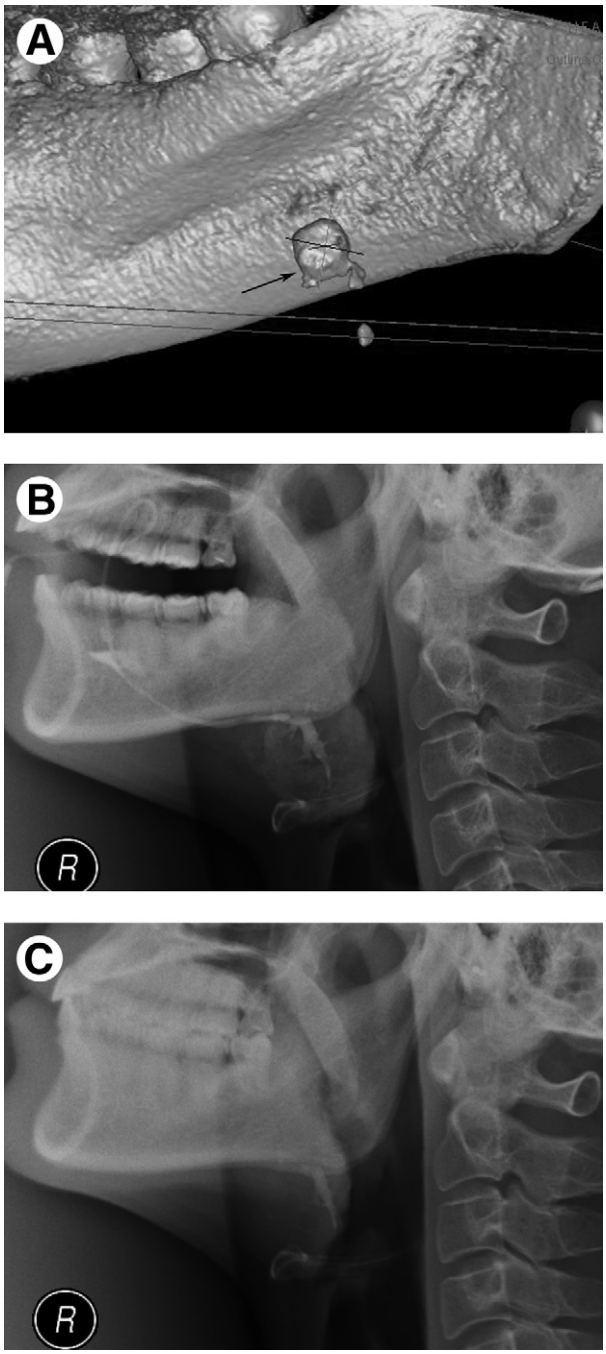

FIGURE 4. A 33-year-old man with a 1.5-year history of a stone in the right Wharton's duct. *A,* Three-dimensional cone-beam computed tomographic image shows multiple stones (*arrow*) at the hilum and deeper branches of the duct. *B,* The filling film 9 months postoperatively shows mild dilatation at the hilum of the duct. *C,* The emptying film shows significant contrast retention.

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slit, opened lengthwise, and sutured to the floor of the mouth after release of the stone. In the approach described by McGurk et al,[4] the duct is exposed, opened over the stone, and then resutured. In 2007, Nahlieli et al[6] reported a ductal stretching technique for the removal of submandibular hilar calculi under endoscopic guidance. In their approach, the duct was isolated up to the first molar and then stretched forward. After stone removal through a ductal incision above the stone, the distal 5 mm was excised and sutured to the orifice region. A sialo drain was inserted and sutured to the floor of the mouth to create a new orifice. In this approach, Nahlieli et al stressed that the endoscope served 2 functions: *1)* to pinpoint the exact location of the stone; and *2)* to explore the gland for additional stones and strictures after stone removal. The insertion of the sialo drain helped drain the saliva from the gland into the oral cavity and prevent future strictures. In the technique described by Marchal,[5] the oral floor incision was guided by the location of the illuminated area and was usually approximately 2 cm in length. After the duct was identified, it was dissected from the lingual nerve. After stone removal, the posterior opening of the floor mucosa was sutured with a stent through the Wharton papilla. Marchal[5] emphasized that the minimal posterior incision of the oral mucosa and duct decreased the risk of further ductal stenosis and provided a more rapid recovery for the patient. The guidewire technique may be used to position the stent accurately in the distal part of the duct. The present technique was similar to that of Marchal.[5] With the guidance of endoscopic lighting, the authors' experience showed that the lingual nerve did not need to be isolated from the genu of the duct, thus decreasing the risk of nerve injury except when the stone was located deep in the intraglandular duct tissues. Only 1 patient in the present series developed lingual nerve hypoesthesia, which resolved completely.

In the present series, the success rate of sialoendoscopy-assisted removal of the submandibular hilar calculi was 92.9%, which is similar to previously reported success rates.[1,4,5] In the 5 cases that failed, the stones were found impacted in the main or branch ducts deep in the gland, and further attempts to remove them were considered hazardous to the lingual nerve and the parenchyma of the gland. These patients accepted the modest residual discomfort without requesting further treatment. Of the 65 successful cases, 2 patients developed ranula during the follow-up period. Because these 2 cases developed in the early phase of the authors' experience, they might be explained by the anteriorly extended incision injuring the sublingual gland. In the later cases, the intraoral incisions were more proximal and no further ranula occurred. Among the successful cases, 80% of patients remained asymptomatic, whereas 20% of patients had mild obstructive symptoms during the follow-up period. In what would be considered a poor-prognosis group, the salivary gland was preserved in most patients.[9]

Objective modalities for the evaluation of gland function include sialometry, sialography, and scintigraphy. Su et al[10] evaluated glandular function quantitatively using sialometry and scintigraphy in 17 patients who underwent sialoendoscopic procedures for stone removal or stenosis dilatation. Their results showed a significant increase in glandular function in the affected glands postoperatively. The authors believe that the introduction of a polyethylene catheter into the Wharton's duct might interfere with the natural secretion of saliva and the measurement error might outweigh the natural difference. Scintigraphic assessments have been used by several investigators.[1,2,10] Makdissi et al[1] measured submandibular gland function scintigraphically in 37 patients with a hilar calculus and concluded that glandular function improved to varying degrees in most patients after stone removal.

Conventional sialography was first described by Carpy in 1904.[11] Despite the wide use of endoscopy, sialography remains a simple technique and an important tool for the assessment of salivary gland obstruction in patients who present with sialadenitis and sialolithiasis.[11,12] It is a minimally invasive and painless technique if performed carefully. Acute salivary gland infection and allergy to contrast material are contraindications for sialography.[12] In the present study, sialography was applied to evaluate the recovery of the ductal system after stone removal. Because the ostium of the Wharton's duct was slit in most cases, a 22-gauge intravenous catheter was used to guarantee complete filling of the ductal and acinar systems. The sialographic appearances were divided into 4 types. Types 1 and 2 represented good recovery of the ductal system, and only 4 of 19 cases had mild symptoms. Types 3 and 4 represented incomplete or poor recovery of gland function, and 7 of 11 of cases had obstructive symptoms. Ectasia of the hilum in 17 of 30 cases occurred where the stones were situated. Strictures can develop from stone formation and from surgery, which can also cause the recurrence of symptoms. These sialographic pathologies necessitate long-term postoperative follow-up, particularly for symptomatic patients. It should be pointed out that the sialographic appearance mainly reflects the morphologic basis of gland function. To date, the most reliable modality for the evaluation of gland function is scintigraphy. Because the authors do not have a scintigraphic machine in their hospital, only 4 of the present cases underwent scintigraphic evaluation. Hence, the relation between sialographic and scintigraphic assessments could not be obtained and should be examined in further studies.

In conclusion, sialoendoscopy-assisted intraoral removal is a safe and effective gland-preservation technique for patients with large calculi at the hilum of the Wharton's duct. Sialography can be used as a simple tool to evaluate the gland status postoperatively. Although sialographic appearances differ among patients, most patients have asymptomatic gland preservation.

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