Interventional Sialoendoscopy for Treatment of Obstructive Sialadenitis

Christopher G. Pace, DMD, * *Kyung-Gyun Hwang, DDS, PhD,* † *Maria Papadaki, MD, DMD, PhD,* ‡ *and Maria J. Troulis, DDS, MSc* §

Purpose: This follow-up study documents the overall success rate of interventional sialoendoscopy; it is a novel, less invasive treatment for obstructive sialadenitis.

Patients and Methods: This was a retrospective follow-up study of 189 patients who underwent a sialoendoscopic procedure at Massachusetts General Hospital from 2004 through 2013. Included were patients who underwent sialoendoscopic treatment for symptoms and clinical findings consistent with obstructive sialadenitis. Four different interventional sialoendoscopic techniques were used: dilation of stricture and irrigation, stone retrieval by basket, stone fragmentation with lithotripsy or laser, and stone removal by endoscopic-assisted "cutdown" operation using the "modified McGurk" technique. The outcome assessed was whether the patient was asymptomatic at 6 months postoperatively.

Results: Interventional endoscopic navigation was accomplished in 164 of 189 patients (87%). In 17 cases, the duct orifice was inaccessible owing to scarring, so the duct could not be navigated. Symptomatic relief was achieved in 148 of 164 patients (90%). Dilatation and lavage for sialadenitis without a stone was accomplished in 52 of 189 patients (28%). Sialoliths were retrieved or fragmented in 137 of 164 cases (84%).

Conclusion: The results of this study show a high success rate in the treatment of obstructive sialadenitis using interventional sialoendoscopy.

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Recurrent salivary duct obstruction is a common disease of the major salivary glands. Recurrent swelling, pain, and discomfort are usually exacerbated during meals, when salivary flow is stimulated. Sialolithiasis is a primary cause obstructive salivary gland disease, with other etiologies including inflammation, strictures, kinks, foreign bodies, anatomic malformations, mucous plugs, tumors, or polyps.[1-4]

Traditional diagnostic imaging for obstructive salivary gland disease includes routine plain radiography, ultrasonography, sialography, scintigraphy, and computed tomographic (CT) scanning. Despite providing 2- or 3-dimensional images of the salivary gland ductal architecture, they may not identify the etiology and location of obstruction. They also do not allow direct visualization of the inner duct system.[1,5,6] Magnetic resonance sialoendoscopy is an alternative way to diagnose the obstruction. It provides presurgical and noninvasive diagnostic information, but does not permit the interventional treatment for duct obstruction.[7]

Traditional treatment options for obstructive salivary gland disease consist of conservative management of symptoms (gland massage, antibiotics, hyperhydration, and sialogogues) or surgical

*Instructor, Department of Oral and Maxillofacial Surgery, and AO/Synthes Fellow, Massachusetts General Hospital, Harvard School of Dental Medicine, Boston, MA.

†Professor, Division of Oral and Maxillofacial Surgery, Department of Dentistry, College of Medicine, Hanyang University, Seoul, South Korea.

‡Attending Oral and Maxillofacial Surgeon, University Hospital of Heraklio, Crete, Greece.

§Associate Professor, Harvard School of Dental Medicine, Boston, MA; Director of Residency Training Program, Department of Oral and Maxillofacial Surgery, Massachusetts General Hospital, Boston, MA.

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Address correspondence and reprint requests to Dr Pace: Department of Oral and Maxillofacial Surgery, Massachusetts General Hospital, Harvard School of Dental Medicine, Warren Suite 1201, 55 Fruit Street, Boston, MA 02114; e-mail: drcpace@yahoo.com

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intervention. The intraoral or extraoral surgical approach may result in sensory or motor nerve damage and facial scarring. Interventional sialoendoscopy is an efficient, minimally invasive, alternative technique for treatment of major salivary gland obstruction caused by mucous plugs, strictures, and sialoliths.[8-10]

After Konigsberger et al[11] first introduced the application of endoscopy for salivary gland stones in 1990, the field of sialoendoscopy has evolved with advancements in instruments and techniques forged by innovators such as Nahlieli et al,[6] Marchal et al,[12] Katz,[8] and McGurk et al.[13] The authors reported their early clinical results of 94 patients from 1999 to 2004 at Massachusetts General Hospital (MGH; Boston, MA) and Baptist Hospital (Miami, FL).[3] The aim of this retrospective follow-up study was to evaluate outcomes using a minimally invasive, alternative treatment for obstructive sialadenitis at the MGH from 2004 through 2013.

Patients and Methods

This is a retrospective case-series study of 189 patients with obstructive sialadenitis of the submandibular or parotid gland. The study was approved by the MGH institutional review boards. The study included patients who had obstructive sialadenitis secondary to sialolithiasis, mucous plugs, or strictures (including some systemic conditions associated with Sjögren syndrome or radioiodine treatment). The diagnosis of sialolithiasis was confirmed by clinical examination, plain radiographs, and CT scans (Fig 1). The same attending surgeon and a resident performed all cases using a Marchal 1.1-mm sialendoscope (Karl Storz, Tuttlingen, Germany).

Data were collected from hospital and clinical charts and videograms of the endoscopic procedures (by C.G.P. and K.-G.H.). Data included gender, age, clinical and surgical information, and date of surgery. Clinical findings included involved gland and recurrent perioperative symptoms (swelling and pain). Surgical information included success of duct navigation and dilation, stone size, retrieval or fragmentation of stone, and presence of strictures or mucous plugs. One of 4 different interventional techniques was used, depending on the characteristic findings of the duct and stone: dilation of stricture and irrigation, stone retrieval by basket, stone fragmentation with lithotripsy or laser, and stone removal by endoscopic-assisted operation (Table 1).

For sialadenitis not associated with a stone, diagnostic endoscopy was performed through the duct system with the aid of irrigation. When a duct stricture with diffuse mucosal thickening was encountered, the narrow duct was negotiated using dilation with copious irrigation. The mucous-like material was lavaged from the duct with saline and suction. Intraduct steroid was administered after copious irrigation of the duct structure.

Mobile stones measuring 2 to 7 mm were retrieved by interventional endoscopy using a 3-prong basket (Karl Storz). The natural duct orifice was dilated, allowing insertion of a 14-gauge polyethylene angiocatheter. The angiocatheter facilitated insertion of the sialendoscope and prevented ductal spasm. Injection of lidocaine through the catheter promoted further dilation of the duct. Smaller mobile stones were retrieved using a basket (Fig 2), whereas a small papillotomy incision was necessary to facilitate the delivery of large stones (Fig 3).

Large stones attached to the duct lumen were fragmented using lithotripsy (Calcustat; Karl Storz) or holmium laser (Fig 4). After fragmentation, the small stone particles were retrieved with the basket (Figs 5, 6) or lavaged from the duct with copious irrigation. If fragmented stone particles were irretrievable during the operation, the remaining small pieces of stone were evacuated through the stent for 1 to 2 months postoperatively.

Large stones that adhered to the ductal inner wall and blocked the duct system were retrieved by transoral and intramucosal dissection under the guidance of endoscopic light ("modified McGurk technique"; Fig 7).[13] The stones were located on camera with the sialendoscope, and transmucosal illumination from the camera light was used to identify the location of the intraoral incision. An incision was placed over the mucosa, and blunt dissection to the stone was performed. A small incision was placed in the identified salivary duct to facilitate stone removal (Fig 8). The stent that had been inserted into the duct was advanced to extend proximally to the new duct incision and sutured into place. The duct was copiously irrigated.

Patients were followed immediately postoperatively and at 1, 3, 6, and 12 months. A successful outcome was assessed as clinically symptom free for 6 months after treatment.

Results

From 2004 through 2013, 189 patients (107 women and 82 men; age range, 8 to 81 yr) underwent an interventional sialoendoscopic procedure for obstructive salivary gland disease. The involved glands were 110 submandibular glands and 79 parotid glands. Sialadenitis not associated with a salivary stone was diagnosed in 52 of 189 patients, whereas sialadenitis with sialoliths was diagnosed in 137 of 189 patients. Interventional endoscopic navigation was successfully accomplished in 164 of 189 patients (87%). In 19 of 25 patients, the duct was inaccessible owing to orifice

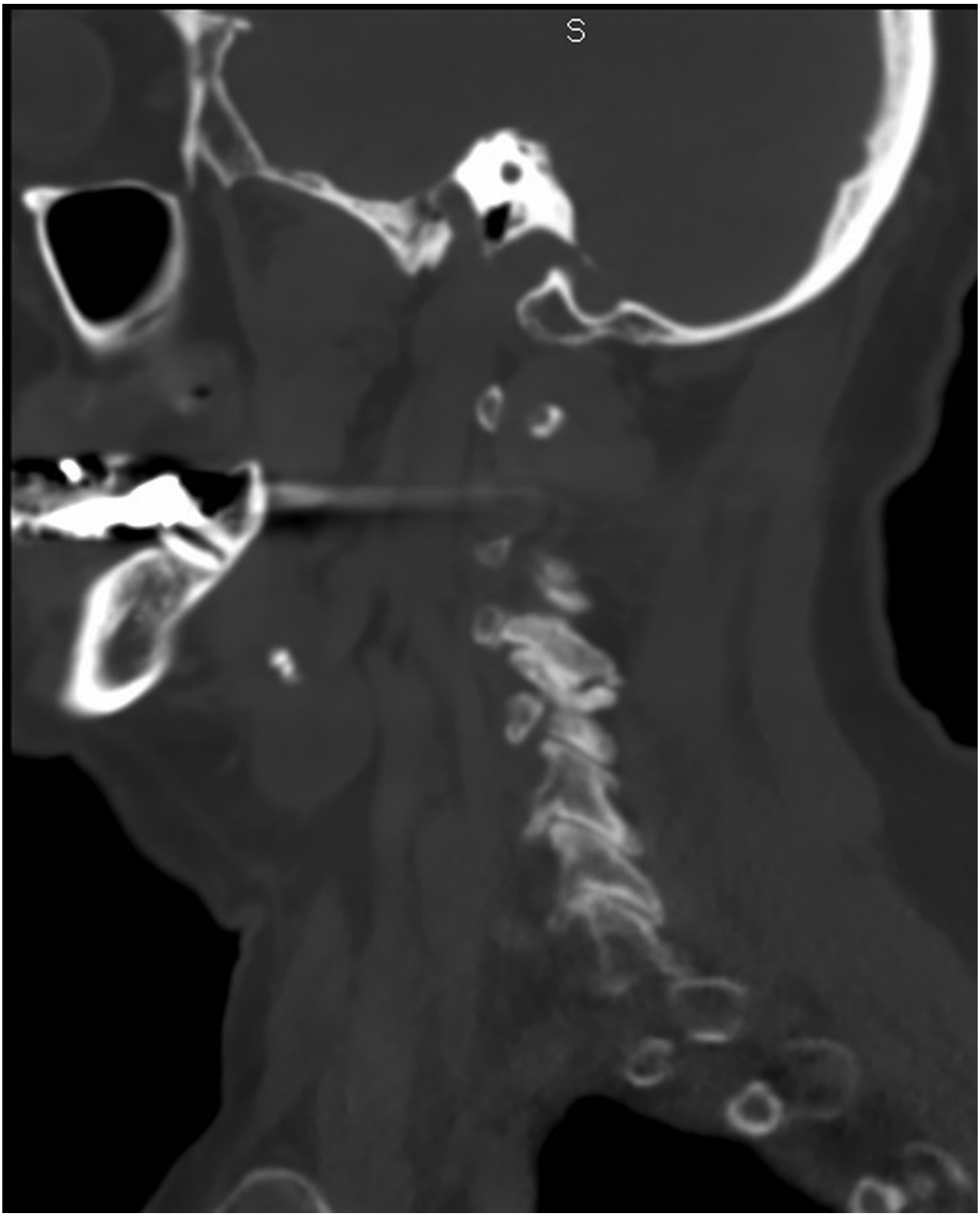

FIGURE 1. Saggital computed tomographic view of stone.

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scarring. In 6 of 25 patients, the stone was located in the gland, beyond the workable distance of the endoscope (Table 2).

Of the 164 successful navigations, 148 patients (90%) were asymptomatic for 6 months after the endoscopic procedure. In 5 of these 148 patients, the

Table 1. FOUR DIFFERENT INTERVENTIONAL SIALOENDOSCOPIC TECHNIQUES

Endoscopic Procedure	Total Patients Successfully Navigated (n = 164)	Stone	Procedure
Dilatation and lavage	52	no	Whenever the duct was very narrow and there was stricture with a diffuse thickening and white tissue appearing around the lumen, the narrow and stricture duct was negotiated with copious irrigation. Visualized diffuse mucus-like tissue was flushed out and suctioned with saline.
Basket retrieval	53	<5 mm	After dilatation of the duct through the natural orifice was accomplished with dilators, small stones were retrieved using a grasper or a basket. A small papillotomy incision was carried out to facilitate the retrieval of large stones.
Combination with lithectomy	51	>5 mm	Large stones that were stuck to inner duct wall and blocked the duct system were retrieved by intramucosal dissection under guidance of endoscopic light.
Lithotripsy and laser	8	>5 mm	Immobilized large stones attached to the duct lumen were fragmented using lithotripsy or laser. After fragmentation, small stone particles were retrieved with a basket or with copious irrigation.

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obstructive symptom recurred 9 to 12 months postoperatively. Two of these 5 patients were free of symptoms after a second endoscopic procedure. Of the 41 patients (189 minus 148) who were symptomatic after 6 months, 2 were asymptomatic after a second and a third procedure to remove the stone. In total, the endoscopic success rate was 90% (148 of 164 patients) after short-term follow-up, and 87% (143 of 164 patients) were asymptomatic after long-term follow-up.

Procedures and Treatment Results

SIALADENITIS NOT ASSOCIATED WITH STONE

In 48 of 52 patients (92%), the ductal stricture was dilated and the mucous-like materials were flushed out. After the first endoscopic procedure, 42 of 48 patients (88%) were asymptomatic from the obstructive gland symptom at their follow-up appointments. In 5 of 42 patients (12%), the symptoms recurred after approximately 1 year. Of the 10 patients who were still symptomatic after the procedure, all but 2 (80%) had involvement of the parotid gland. Thirty-seven of 52 patients (71%) were completely asymptomatic after 1 year.

MOBILE STONE RETRIEVED BY BASKET

In 53 of 137 patients (32 submandibular glands and 21 parotid glands), stones were retrieved by basket or washed out with copious irrigation. Retrieved stones varied from 1 to 9 mm. After the endoscopic procedure, 50 of 53 patients (94%) were asymptomatic for 6 months. Two of 50 patients had recurrence of symptoms after 9 months. The final success rate of endoscopic stone retrieval by basket was 91% after 9 months.

IMMOBILIZED LARGE STONE FRAGMENTED BY LITHOTRIPSY OR LASER

In 8 of 137 patients (5 submandibular glands and 3 parotid glands), large stones were fragmented by intracorporeal shockwave lithotripsy or laser-assisted sialoendoscopy. Fragmented stone particles were retrieved by basket and irrigation. Remnant particles were passively evacuated through a stent for 1 to 2 months after the procedure. In 5 of 8 patients (63%), the stones were effectively removed, and patients were asymptomatic. In 2 patients who had involvement of the submandibular gland, the large stone was not fragmented into small pieces effectively during the operation. In 1 patient who had involvement of the parotid gland, the stone was fragmented to small pieces and retrieved by basket and irrigation. However, symptoms recurred after 3 months.

IMMOBILIZED LARGE STONE RETRIEVED WITH ENDOSCOPIC ASSISTED OPERATION

In 51 of 137 patients, large stones were retrieved using transoral and intramucosal dissection with the

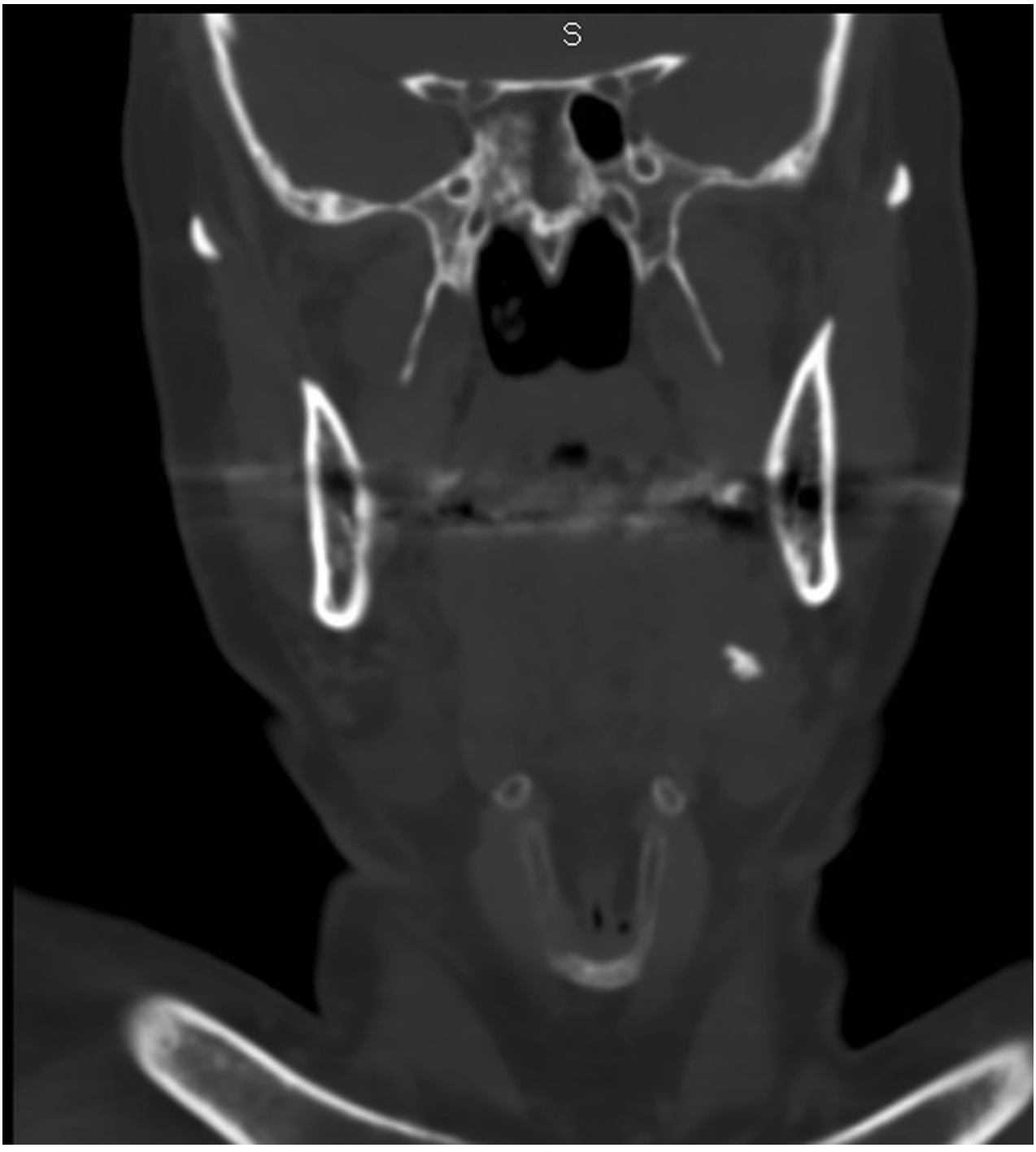

FIGURE 2. Coronal computed tomographic view of stone.

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guidance of endoscopic light. The submandibular gland was affected in 46 patients, and 5 patients had involvement of the parotid gland. After stone removal, 49 of 51 patients (97%) were asymptomatic for 6 months postoperatively. Four patients had recurrence of symptoms after 1 year. One patient required a second operation, and 2 patients required a third operation and were subsequently asymptomatic for 2 years. The other patient had no further treatment. The final success rate of this technique was 92% (47 of 51 patients).

Discussion

Recurrent salivary duct obstruction is a common disease of the major salivary glands. Persistent

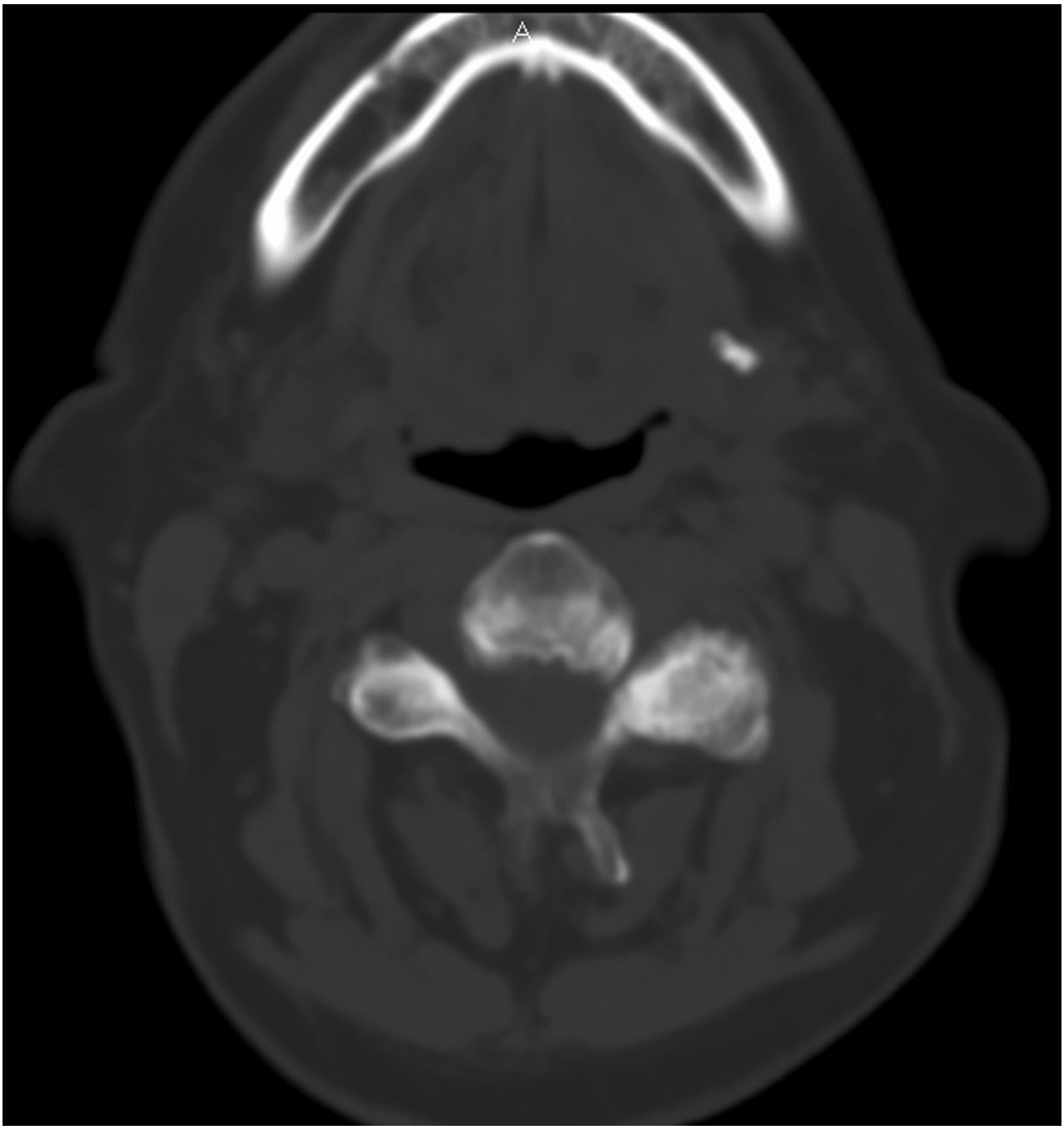

FIGURE 3. Axial computed tomographic view of stone.

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swelling, pain, and discomfort can be aggravated while eating, when salivary flow is stimulated. Major salivary gland sialolithiasis is a primary cause of obstructive salivary gland disease and can be found in 1.2% of the general population.[14] The salivary duct stone is composed of organic and inorganic substances. The exact pathogenesis of salivary stone formation remains unclear; however, intracellular microcalculi, mucous plug, and bacteria within oral cavity have been implicated in the formation of the sialolith.[15] Other known causes of salivary duct obstruction are inflammation, strictures and kinks, foreign bodies, anatomic malformations, mucous plugs, and polyps.[1-4] Radioiodine treatment for thyroid disease and Sjögren syndrome also can cause recurrent obstructive symptoms in major salivary glands.[16,17]

Konigsberger et al[11] in 1990 and Katz[8] in 1991 reported their experience of endoscopic diagnosis of

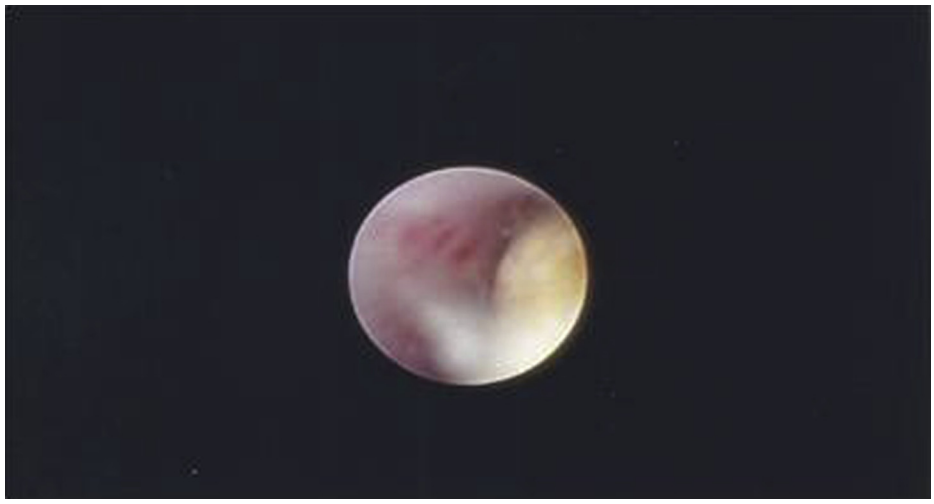

FIGURE 4. Approaching stone in the duct.

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salivary gland ducts. Since then, there have been instrument innovations and technique refinements allowing the transformation of sialoendoscopy from solely a diagnostic tool to an interventional instrument. In the present study, 4 different methods of interventional sialoendoscopy were used. When the stone was 3 to 5 mm in the parotid gland duct and 4 to 9 mm in the submandibular gland duct, the small stone was retrieved by a basket. These small stones were not attached to the duct wall, appearing buoyant in the duct lumen. When the duct orifice prevented delivery of a stone larger than 2 mm, a small papillotomy incision facilitated removal. In this study, 50 of 53 patients (94%) had 2- to 9-mm stones removed, and 48 of 53 patients (91%) were asymptomatic after 1-year follow-up. Marchal et al[9] reported that 97% of 36 patients who had stones smaller than 3 mm could undergo retrieval by basket without the need for fragmentation. Nahlieli et al[10] reported an 87% retrieval rate in 217 patients with stones smaller than 5 mm in the Stensen duct and 7 mm in the Wharton duct. Iro et al[18] reported 91.5% successful removal of stones smaller than 5 mm from the Wharton duct in 1,522 patients. Brown et al[19] reported the use of radiologically guided intervention for stones smaller than 7 mm, which resulted in 82% symptom relief.

Larger stones located more proximal to the gland and attached to the lumen cannot be retrieved by simple endoscopy with a basket. Since Seward[20] first attempted to retrieve large salivary stones in 1968, several transoral techniques have been introduced. Transoral stone removal has been a successful

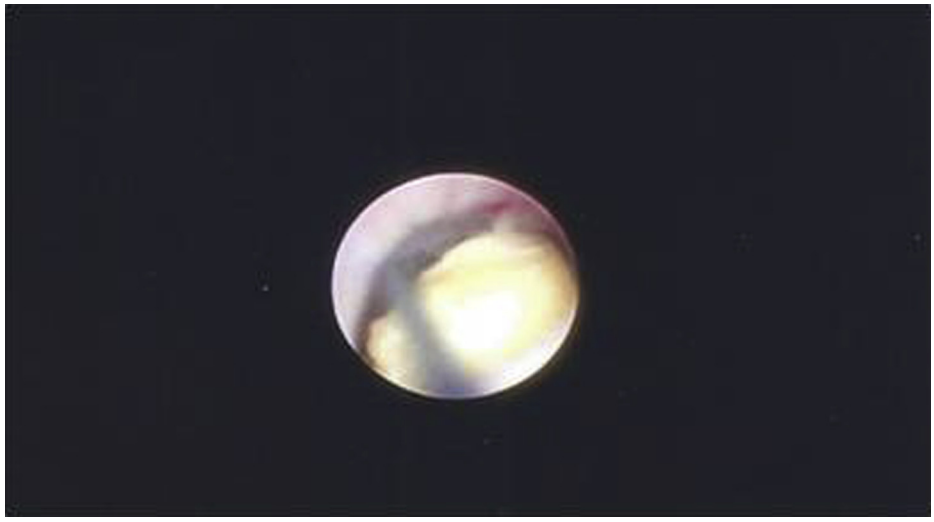

FIGURE 5. Basket around the stone.

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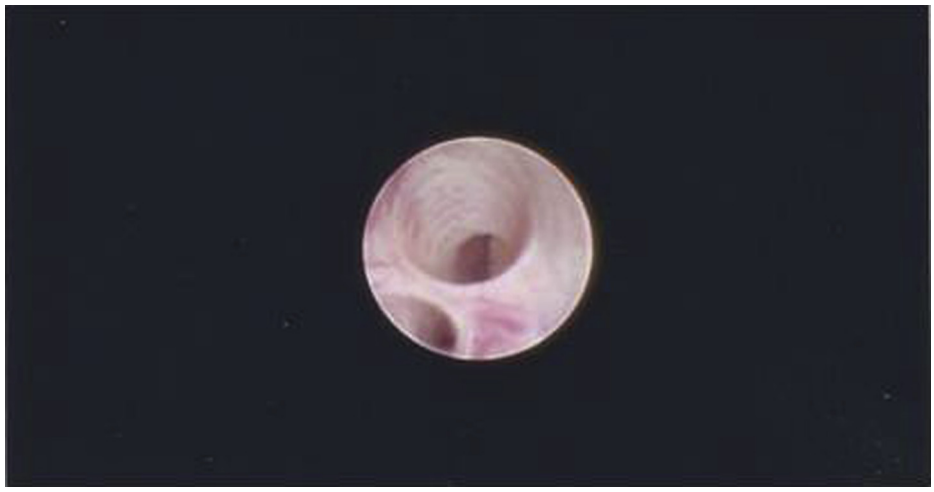

FIGURE 6. Bifurcation visualization after stone removal.

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treatment of choice for large, palpable, submandibular duct stones located in the perihilar area.[13,20,21] However, an extraoral approach is indicated for large stones when they are not palpable, are attached to the duct wall, or are deep in the parotid gland. Nahlieli et al[10] proposed endoscopically assisted removal for large parotid gland stones. These stones were retrieved by intramucosal dissection under guidance of endoscopic light. This endoscopically assisted stone removal has yielded a high success rate for stones in the large parotid and submandibular glands. Marchal[22] reported 92% success in 37 patients with parotid stones. Walvekar et al[23] claimed 90% success in 20 cases, and Liu et al[24] reported greater than 95% success in 34 patients. In the present study, 92% of patients had symptom relief after 1-year follow-up using a modified McGurk technique.[13]

The surgical approach to major salivary glands harbors the risk of injury to adjacent vital structures. The minimally invasive, endoscopic technique is an attractive alternative to mitigate the risks of traditional surgical approaches. Immobilized large stones attached to the duct lumen present a challenge, and improvements to the interventional sialoendoscopic technique are necessary for their retrieval. After Iro et al[25,26] attempted extracorporeal shockwave lithotripsy (ESWL) for treatment of salivary stones, several types of sialic lithotripsy have been introduced as

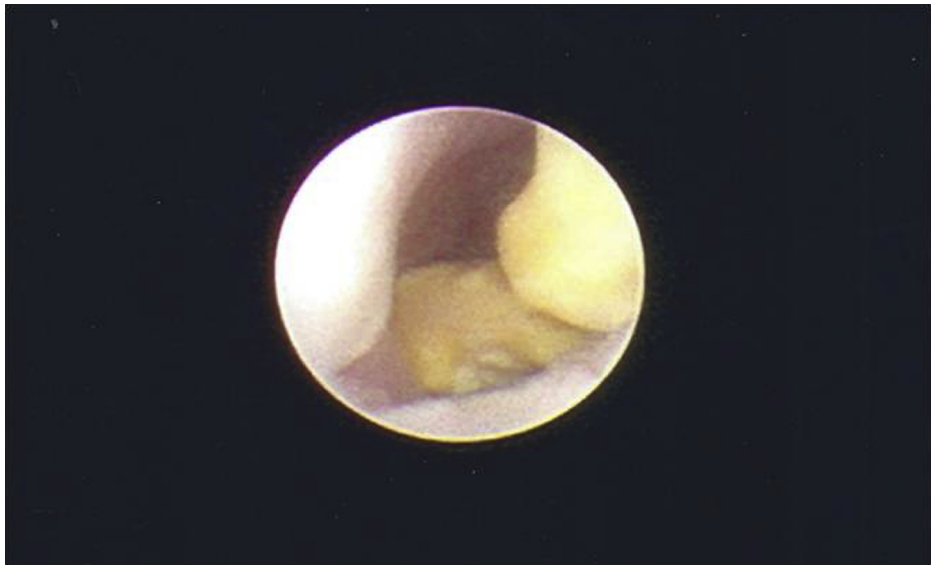

FIGURE 7. Encountering a larger stone.

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FIGURE 8. Complete removal of stone, basket, and catheter.

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a noninvasive method for the management of large salivary stones. However, ESWL has not resulted in an acceptable clinical outcome because of a success rate of only 30 to 70% and much postoperative discomfort (pain or swelling).[25-28] Intracorporeal lithotripsy and laser treatment have been used to manage large stones that are irretrievable with a basket alone. Using lithotripsy, large stones are fractured into smaller pieces, rendering them manageable with a basket or lavage. Any remaining particles are passively egressed through a stent for 1 to 2 months postoperatively. The authors' experience with lithotripsy and sialoendoscopy resulted in no symptoms in 8 patients (63%). Arzoz et al[2] reported that 15 of 18 patients treated for sialolithiasis with pneumobalistic lithotripsy were symptom free. Raif et al[29] reported that 15 of 21 patients were symptom free after erbium:YAG laser lithotripsy with interventional sialoendoscopy. Although several methods of lithotripsy have been used in salivary stone fragmentation, the reported clinical success rate and effectiveness of stone fragmentation are limited. More clinical research is necessary to evaluate the effectiveness of intracorporeal equipment for large stone fragmentation.

Table 2. IMMEDIATE FAILURE OF SIALOENDOSCOPY

	Patients Treated (N = 189)		
	Parotid (n = 79)	Submandibular (n = 110)	Total (N = 189)
No stone	3/35	1/17	4/52
Stone	9/44	12/93	21/137
Total	12/79	13/110	25/189

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Nonspecific sialadenitis is an obstructive salivary gland disorder without the presence of a duct stone. Some infectious diseases, autoimmune diseases, or radioiodine therapy can cause sialadenitis with stenosis of the salivary gland duct, but the etiology and mechanism are unclear.[30-32] Conservative treatment has limited success, and surgical removal of a gland exposes patients to risks of complications.[31,33-35] Endoscopic management of these glands can provide more information about the duct lumen pathology, such as duct strictures versus mucous plugs.[1] Interventional sialoendoscopic dilatation and lavage can provide minimally invasive management of obstructive salivary ducts. Although interventional sialoendoscopic management has been the treatment of choice for duct stricture, the clinical success rate has not been as successful as sialoendoscopic management for sialoliths. In the present study, the success rate was 81% (42 of 52 patients) in short-term follow-up and 71% (37 of 52 patients) after 1 year. Nahlieli et al[4] reported a similar success rate (80%; 20 of 25 patients) with endoscopic management of duct stenosis. Koch et al[36] reported a 75% success rate in the management of Stensen duct strictures.

Advances in the technology used to fabricate endoscopic equipment might facilitate more flexible and navigable scopes, smaller manipulation instruments, higher-definition video, and more effective evacuation techniques used in interventional sialoendoscopy. These advances will contribute to refinements in the procedures used to remove salivary duct stones by allowing deeper gland navigation, improved fragmentation of stones, and stone removal from duct lumens. More diagnostic information also could be gathered during the procedures that would ultimately minimize morbidity and improve treatment outcomes.

The endoscopic approach to salivary gland duct disorders provides an effective and minimally invasive option for diagnosis and treatment. In cases with sialolithiasis, interventional sialoendoscopy with basket retrieval of mobile stones and endoscopically assisted sialolithectomy of larger stones can yield a high rate of symptomatic relief. However, the applications of lithotripsy and laser in the management of large stones attached to the duct lumen require further technical refinements and innovative instrument development. In addition, further research of the mechanism and etiologic factors associated with non-stone obstructive gland disease is needed to develop improved treatment options.

Press Release

This article's Press Release can be found, in the online version, at http://dx.doi.org/10.1016/j.joms.2014.06.438.

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