



## Pediatric sialendoscopy in Asians: A preliminary report<sup>☆,☆☆</sup>



Chin-Hui Su<sup>a,b</sup>, Kuo-Sheng Lee<sup>a</sup>, Jui-Hsien Hsu<sup>a</sup>, Fei-Peng Lee<sup>b,c</sup>, Hsiang-Yu Lin<sup>d,e,f</sup>,  
Shuan-Pei Lin<sup>d,e,f</sup>, Shih-Han Hung<sup>b,g,\*</sup>

<sup>a</sup> Department of Otorhinolaryngology, Mackay Memorial Hospital

<sup>b</sup> Department of Otolaryngology, School of Medicine, Taipei Medical University, Taipei, Taiwan

<sup>c</sup> Department of Otolaryngology, Shuang-Ho Hospital, Taipei Medical University

<sup>d</sup> Department of Pediatrics, Mackay Memorial Hospital, Taipei, Taiwan

<sup>e</sup> Mackay Medicine, Nursing and Management College, Taipei, Taiwan

<sup>f</sup> Department of Medicine, Mackay Medical College, New Taipei City, Taiwan

<sup>g</sup> Department of Otolaryngology, Taipei Medical University Hospital

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### ABSTRACT

**Background/purpose:** The sialendoscopic approach in treating pediatric salivary gland disorders has been reported with great success through the years. Whereas this success has been widely reported in Caucasian populations, relatively little has been reported regarding the use of this procedure in pediatric patients in Asian countries. The purpose of this study is to report our preliminary experience in pediatric sialendoscopy.

**Methods:** The data from 20 patients (<18 years old), who underwent sialendoscopy for obstructive sialoadenitis in the Department of Otorhinolaryngology of Mackay Memorial Hospital between October 2013 and November 2015, were reviewed.

**Results:** Twelve of our 20 patients (60%) were diagnosed with sialolithiasis and 8 of our 20 patients (40%) presented with non-lithiasis obstructive sialoadenitis. Ductal stenosis was found in 13 patients, and 18 patients had debris/mucous plug formation. The overall success rate was 95% (19/20) in our series, and 85% (17/20) of the patients had achieved a complete remission after a single sialendoscopy procedure.

**Conclusions:** Sialendoscopy is an ideal treatment in the management of obstructive sialoadenitis in Asian pediatric patients. If necessary, Holmium:YAG laser lithotripsy and sialostent placement could be applied, and both procedures are well tolerated in pediatric patients.

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Sialoadenitis accounts for up to 10% of salivary gland diseases in the pediatric population, with viral parotitis and juvenile recurrent parotitis (JRP) being the two most common causes [1]. Obstructive sialoadenitis is another frequently encountered condition and is common in patients with non-neoplastic salivary gland disorders [2]. Whereas stones are responsible for most of the cases of obstructive sialoadenitis in the adult population, sialolithiasis in children is relatively uncommon, with few case series reported in the literature [3–6]. In children, most calculi have been reported to be relatively small (<1 cm, 93.1%) and located in the distal duct (62.0%) [4]. However, other etiologies, including strictures, mucoid plugs, and anatomic ductal abnormalities have been observed, as well [7].

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\* Corresponding author at: Department of Otolaryngology, Taipei Medical University Hospital, No.252, Wu-Hsing Street, Taipei City, 110, Taiwan. Tel.: +886 2 2737 2181; fax: +886 2 28369275.

E-mail address: [seedturtle@gmail.com](mailto:seedturtle@gmail.com) (S.-H. Hung).

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For pediatric patients, conservative treatment with medication is typically considered first, but for stone cases, early planning of surgical remove might be standard [8].

In cases in which conservative management with medication fails to control the disease, surgery, including salivary gland excision procedures, is considered, particularly if a structural obstruction of the salivary duct is confirmed (e.g., stones) [9]. The complications of open surgery are related to bleeding, wound infections, permanent scar formation, and damage to the facial nerve during salivary gland surgery, which is probably the most significant surgical complication [10].

Sialendoscopy was introduced more than two decades ago and has become the leading diagnostic technique and interventional tool for pediatric sialoadenitis [11]. In addition to sialolithiasis, sialendoscopy is increasingly used in patients with chronic sialoadenitis, such as Sjogren's syndrome, radioiodine sialoadenitis, and juvenile recurrent parotitis [12–15]. This procedure could be applied to pediatric patients, who have been hypothesized to have relatively smaller salivary ducts. Great success has been attributed to sialendoscopic approach in treating pediatric salivary gland disorders through the years [16–19]. Whereas this success has been widely reported in Caucasian populations, relatively little has been written regarding the use of this procedure in

pediatric patients in Asian countries. In this study, we report our preliminary experience in pediatric sialendoscopy in Taiwan.

## 1. Methods

The Institutional Review Board of Mackay Memorial Hospital approved the study protocol. We reviewed the data of 20 patients (<18 years old) who underwent sialendoscopy for obstructive sialoadenitis in the Department of Otorhinolaryngology of Mackay Memorial Hospital between October 2013 and November 2015. The procedures were performed under general anesthesia by the same surgeon. We compared the clinical information of the patients, including patient data, CT and sialendoscopic findings, surgical time, and clinical outcomes.

The procedures were performed using a Miniature Straight Forward Telescope with a direct view at 0° (All in One Karl Storz®, Germany). The semi-rigid scope had a working length of 12–16 cm and an outer diameter of 1–1.3 mm (models 11,575 A and 11577KE). A 0.65-mm working channel and an irrigation channel with a diameter of 0.25 mm were attached. An operation sheath with a 1.15-mm working channel and a telescope channel of the same diameter were further attached. Laser lithotripsy was performed with a Holmium:YAG laser generated through the VersaPulse® PowerSuite™ system (Lumenis®, Israel) and was used during the sialendoscopy for stone fragmentation. The Holmium:YAG laser was used with a power of 3–6 W, a rate of 5–10 Hz, and energy delivery of 0.6 J.

For every sialendoscopic procedure, papilla identification and dilatation were performed for the entry route of the sialendoscope. Retro-papillar incisions into the salivary duct were performed if there was difficulty in identifying or dilating the papilla. When palpable mouth floor stones could not be smoothly removed with sialendoscopy, a combined approach was performed, with additional incisions in the salivary duct and subsequent removal of the immobile stone. If the confined stones were not palpable trans-orally, or were located deep inside the floor of the mouth, the Holmium:YAG laser was used for stone fragmentation, with subsequent removal by sialendoscopy.

If a patient with obstructive sialoadenitis was found to have sialolithiasis with/or a stenosis in the salivary duct, a stent was placed inside the duct after the sialendoscopic procedures were completed. The length was measured by the scope from the papilla to the first bifurcation, and the stent was 0.5–1 cm longer than the main duct (the papilla to the first bifurcation). The guide wire was placed via the working channel to confirm the intraductal position. After proper dilatation, the stent was placed into the duct without resistance, before the guide wire was removed.

All of the patients were administered an intraductal steroid instillation (40 mg of Solu-Medrol) after the sialendoscopy. Prophylactic antibiotics and analgesics were administered post-operatively for seven days. The patients were followed-up for at least one month after the stent removal, and their outcomes were recorded according to the post-operative follow-up medical records. The LSD classification was recorded according to the criteria reported by Marchal et al. [20]. A good outcome with complete remission was defined as the patient being completely symptom-free without a relapse for at least three months after the procedure.

## 2. Results

In total, 20 pediatric patients underwent a sialendoscopy, including 13 males and seven females. The age ranged from 2 to 17, with an average of 11.2 years of age. The affected glands included 16 submandibular glands (11 left; 5 right), three left parotid gland, and three right parotid glands (Table 1). Twelve of our 20 patients (60%) were diagnosed with sialolithiasis with a stone size ranging from 2 to 18 mm. Nine patients presented with a solitary stone and three children had multiple stones. Seven children received Holmium:YAG laser lithotripsy for the

fragmentation of large or stacked stones. Three children received a sialendoscopy combined with an intraoral sialolithotomy. One patient was treated by simple stone removal and one patient was managed by conventional open sialolithotomy.

Seven of our 20 patients (35%) presented with non-lithiasis obstructive sialoadenitis. Four of these patients were suffering from recurrent parotid gland swelling every month, with a confirmed diagnosis of juvenile recurrent parotitis (JRP). The other three children presented with a few episodes that included signs of submandibular gland obstruction; we were unable to identify the exact causes in those cases. Ductal stenosis was found in 13 patients, and 17 patients had debris/mucous plug formation. Typical pale ductal mucosa and washed out mucous plugs were found in three of the patients presented with JRP (Fig. 1).

The mean surgical time was 62.2 min for the patients who only underwent diagnostic sialendoscopy. In cases in which additional procedures, such as diagnostic endoscopy with a retro-papillar incision approach and laser lithotripsy were performed, the mean surgical time was extended to 91.3 and 157.7 min, respectively (Fig. 2). There were no significant differences in the comparison with adult patients who underwent diagnostic sialendoscopy and laser lithotripsy procedures during the same data collection period (unpublished data).

The overall success rate was 95% (19/20) in our series, and 85% (17/20) of the patients achieved a complete remission after a single sialendoscopy procedure. Two patients with sialolithiasis and ductal stenosis required treatment by another sialendoscopic procedure after Holmium:YAG lithotripsy because of persistent ductal stenosis. We were not able to identify the stone through sialendoscopy on one 12-year old boy who was found to have a 4.68 mm calcification via computed tomography. The patient subsequently received a conventional open sialolithotomy because of persistent signs of obstruction.

Sialostents were placed in twelve of the 20 children who had ductal stenosis or underwent potential duct damaging procedures, e.g. laser lithotripsy and/or retro-papilla ductal incision. The minimum sialostent placement duration was two weeks, and the stents were removed during follow-up visits.

## 3. Discussion

This study reports that sialendoscopy can be safely and effectively performed on Asian pediatric patients. The operation time of the sialendoscopic procedure in pediatric patients did not differ from that in adults. Our data support the notion that the use of sialendoscopy in Asian pediatric patients might be as effective as in adults.

Interventional sialendoscopy has become more accepted since its introduction by Prof. Marchal more than a decade ago [21,22]. During this time, the procedure has evolved from its first use for simple diagnostic observation to the integration of various treatment modalities. In addition to using traditional forceps or wire-basket instruments, the sialendoscope instrument can be easily integrated with the Holmium:YAG laser device without compromising the performance of standard sialendoscopy [23,24].

The first report specifically concerning pediatric sialendoscopy was by Nahlieli et al., who reported the use of sialendoscopy in 15 children [25]. As sialendoscopic procedures rely on instruments passing through the salivary orifice and working inside the salivary duct, it is reasonable to suggest that the procedure might be more easily performed on larger individuals, such as adult patients. However, there does not appear to be any evidence discouraging the use of sialendoscopic procedures in pediatric patients. Faure et al. reported a preliminary observation that the size of the salivary gland ductal system in children treated for salivary stones does not appear to be not much smaller than that in adults [16,26]. The procedure appears to have shown itself to be as effective in managing pediatric patients as adults. The authors, later on, published their five-year experience in pediatric sialendoscopy and concluded that sialendoscopy is a pertinent technique for the diagnosis and treatment of salivary gland disorders in children and allows for

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