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Pediatric Salivary Gland Obstructive Swelling: Sialendoscopic Approach

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Objective: To assess the efficacy of sialendoscopy as a diagnostic and interventional procedure for salivary ductal pathologies of children.

Design: Prospective case series study.

Setting: Tertiary care teaching hospitals.

Patients: Eight children were investigated under general anesthesia by sialendoscopy for recurring salivary gland swellings between 2003 to 2004 in two university centers.

Intervention: Diagnostic sialendoscopy was used for classifying ductal lesions as sialolithiasis or stenosis. Interventional sialendoscopy was used to treat these disorders. Different variables were analyzed: type of endoscope used, intraoperative findings, type of device used for sialoliths fragmentation or extraction, total number of procedures, and size and number of sialoliths removed.

Results: Five cases of parotid and three cases of submandibular gland recurring swellings were included in the present study. Diagnostic sialendoscopy was possible in all cases. Salivary stones were found in six patients and parotid ductal stenosis in the remaining two. Multiple stones were seen in two cases. Interventional sialendoscopy was also possible in all cases, allowing an intraductal retrieval of the stones in three cases, and a marsupialization of the duct in two cases. Two cases required laser fragmentation of the stone. No major complications occurred intraoperatively or during follow-up (mean 18 months).

Conclusion: Diagnostic sialendoscopy is a new technique allowing a reliable evaluation of salivary ductal disorders in children, with low morbidity. Interventional sialendoscopy allows early treatment of pediatric sialoliths and stenosis in most cases, avoiding classical open surgery.

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INTRODUCTION

Pediatric salivary gland swellings are relatively uncommon and when unilateral should raise the question of obstructive pathologies such as sialolithiasis and stenosis.

Sialolithiasis can occur at any age, but there is a peak incidence between the fourth and sixth decades.[1] Less than 5% of all sialolithiasis occur in childhood, and less than 100 pediatric cases of sialolithiasis have been described in the literature,[2–4] most often included in larger adult series.[5–7]

The diagnosis of salivary stones in children is difficult because the lower limit of radiologic detection of salivary stones is 2 mm in diameter,[8] a size sufficient to cause a ductal obstruction.

The small diameter of the child's salivary ductal system has made a sialendoscopic exploration more challenging than in adults. Therefore, only 15 sialendoscopies to treat pediatric sialolithiasis have been reported to date,[3] using a surgical unit of an outside diameter of 2 to 3 mm. We report on a new minimally invasive technique of salivary duct exploration and treatment of obstructive swelling under direct endoscopic control in children.

PATIENTS AND METHODS

Patients

Between January 2003 and June 2004, eight children presenting recurrent salivary gland swelling (5 with parotid and 3 with submandibular symptoms) were investigated and treated by sialendoscopy in two university centers (Geneva University Hospital, Switzerland and Edouard Herriot Hospital, Claude Bernard University, Lyon, France). There were three girls and five boys, with a mean age of 7 (range 3–12) years.

Patients were included according to clinical and radiologic criteria. All patients suffered at least three parotid or submandibular episodes during meals. All patients had prior mumps vaccination.

All patients were evaluated initially for unilateral salivary gland swellings. Most of them had a preoperative radiologic evaluation, including ultrasound and sialography performed in others centers. Sialography was performed in three cases and

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ultrasound in all the cases. Sialolithiasis was suspected in two cases by ultrasound coupled with sialography and in five cases by ultrasound alone. In one case, there was a suspicion of sialolithiasis on sialography with a normal ultrasound. This parotid case was initially suspected to be a recurrent juvenile parotitis.

All procedures were performed under general anesthesia. Diagnostic sialendoscopy was used for classifying ductal lesions as sialolithiasis or stenosis. During the same anesthesia, interventional sialendoscopy was used to treat these pathologies. A "sialendoscopic success" was defined as the clearance of the entire ductal lumen and its branches of any disease, as well as the resolution of symptoms. The mean follow-up of this population was 18 (range12–30) months.

The interventions were performed with the patient under oral antibiotic prophylaxis. Antibiotics (Amoxicilline-Clavulanic acid) and corticosteroids (Predsinolone 50 mg) were given for 48 hours.

Instruments

We used two of the last generation sialendoscopes developed in Geneva. The first system has a semirigid optic endoscopic device (reference 11576; Karl Storz Co., Tuttlingen, Germany), which gives the endoscope some flexibility. It has an internal diameter of 0.75 mm and two shafts: one for diagnostic purposes (external diameter of 1.1 mm) and one for interventional purposes, with a second shaft (working channel) of 1 mm of diameter, being 1.71 mm of total external diameter. The second system is the 1.3 mm "all in one" sialendoscope (Fig. 1) (reference 11575; Karl Storz Co.), with an external diameter of 1.3 mm, including a fiber optic, an operating channel (0.65 mm diameter), and an irrigation channel (0.25 mm diameter). This unit can be used either as a diagnostic or as an interventional sialendoscope. In both devices, it is possible to introduce baskets (reference 11575K/L; Karl Storz Co.) for retrieval of stones and laser fibers for in situ laser lithotripsy fragmentation.

Papilla dilatation was achieved with a set of customized dilators (reference 745910 and 745845-744856; Karl Storz Co.). Fragmentation of larger stones was achieved with a 0.4-mm holmium laser probe (Coherent, Versa pulse Select, Santa Clara, CA). Dilation of stenosis was performed either with the interventional endoscope itself, or with balloon catheters in case of localized stenosis (reference 28635P; Karl Storz Co.)

Surgical Technique

The first step consists of the dilation of the papilla with salivary probes of increasing diameter, followed by the use of the papilla dilator. Papillotomy is avoided if possible. The second step is the diagnostic sialendoscopy. The cleansing of the endoscope tip and slight dilation of the duct to obtain a clear vision are achieved by an intermittent rinsing through the endoscope with a local anesthetic solution (half xylocaine 2% and half NaCl 0.9%). Diagnostic sialendoscopy allows a minimally invasive and complete exploration of the ductal system. When stones or stenosis or other ductal disorders are identified, an interventional sialendoscopy is performed during the same stage. For stones smaller than 2 to 3 mm in diameter, the customized wire basket is introduced behind the stone, spread out and tightened around the stone against the tip of the endoscope, trapping the stone, before the removal of the entire sialendoscope (Fig. 2). For larger stones, fragmentation is required before extraction. Laser sialolithotripsy is performed under direct visual control and retrieval of stone fragment is achieved with the grasping wire basket. After the last stone is removed, the endoscope is introduced again to rinse the duct, and verify its integrity and the absence of disease behind the removed stone. When a stenosis is located, a balloon catheter is passed through the stenosis and inflated to dilate the stenosis under visual control.

The variables analyzed include the number of diagnostic and interventional procedures, the type of ductal pathology (stones, stenosis), the number of stones or stenosis, and the fragmentation and extraction device used.

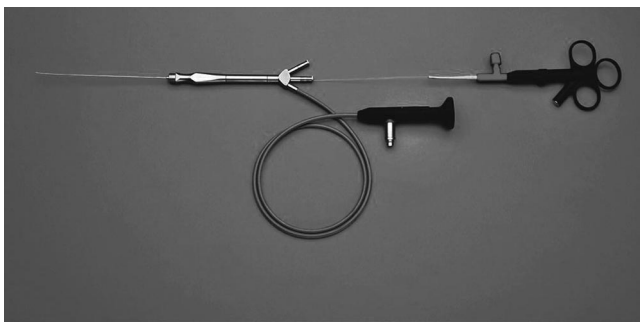

Fig. 1. Marchal sialendoscopes, 1.3-mm diameter.

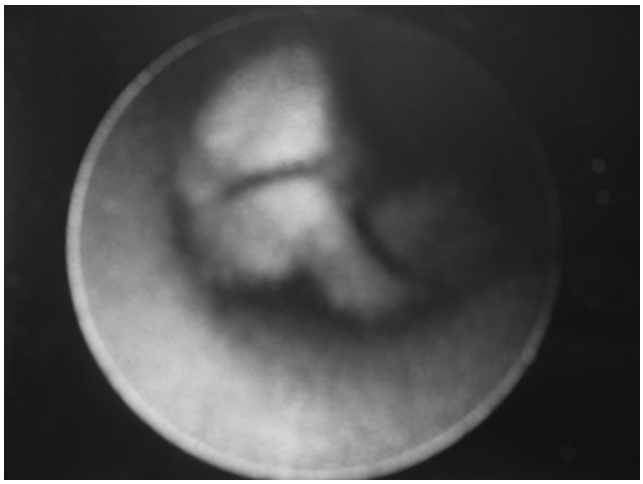

Fig. 2. Stone trapped under vision endoscopic control with wire basket and retrieval by the papilla.

RESULTS

Eight children presenting recurrent salivary gland swelling (5 with parotid and 3 with submandibular symptoms) were included in the present study. There were three girls and five boys, with a mean age of 7 (range 3–12) years. Most of them had prior radiologic evaluation, with a suspected diagnosis of sialolithiasis in five cases (Table I).

TABLE I.
Results.

	Parotid	Submandibular
Diagnostic sialendoscopy	5/5	3/3
Stenosis	2/5	0/3
Mucous plug	3/5	2/3
Intraductal retrieval	2/5	2/3
Large marsupialization	1/5	1/3
Laser fragmentation	1/5	1/3

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Diagnostic sialendoscopy was possible in all cases. The cause of ductal obstruction was identified as being salivary stones in six cases and ductal stenosis in two cases. Mucous plugs were also found in three cases. All submandibular glands investigated presented sialoliths, including one case with multiple stones. Three cases of sialolithiasis and two cases of ductal stenosis were identified in the parotid glands. These two stenoses had not been detected by preoperative ultrasonography.

Interventional sialendoscopy was also possible in all cases. In seven of eight cases, only one procedure was performed to eliminate the cause of obstruction. The last case that presented a 6-mm parotid stone required three procedures and was complicated by a ductal perforation and spontaneous expulsion through the cheek. Intraductal retrieval of the stones was performed in three cases. One of them required prior laser fragmentation of the stone, followed by a mini marsupialization of 2 to 3 mm. A larger marsupialization of 1 cm was necessary in two cases because of bigger stones. One child, who had prior laser fragmentation of a parotid sialolith, had experienced ductal perforation during intraoperative attempts of sialolith retrieval.

No relapse and no postoperative complications occurred during follow-up (mean: 18 months, range: 12–30 months). No major complications such as facial palsy or bleeding were encountered. No open salivary gland resection was necessary.

DISCUSSION

The incidence of parotid involvement is still debated in literature, ranging from 10% to 40% of cases.[1,2] The incidence of pediatric salivary duct stenosis is not mentioned in the literature, probably because conventional radiology and sialography are not accurate enough to detect this disorder.

Unilateral pediatric obstructive swellings may be divided into two main disorders: sialolithiasis and stenosis.[3–9]

Pediatric sialolithiasis represent 5% of all cases of lithiasis. Etiology of salivary stones is still unknown. Previously, authors suspected that foreign bodies were one of the causes of lithiasis.[3] We could not confirm this theory in our limited series.

The frequent involvement of the parotid ductal system (5 of 8 cases, including 3 sialoliths in the present series) is not found in the literature,[3] probably because of our tertiary recruitment and the use of a thin endoscope of less than 1.3 mm in external diameter. According to the literature, the submandibular gland seems to be more frequently involved than does the parotid.[4] Such was not the case in our series, probably because our patients were referred to a tertiary care center. Moreover, the use of a thin endoscope allowed a more accurate diagnosis of lithiasis.

We believe that the popularization of parotid gland sialendoscopy will result in an increased recognition of parotid gland obstructive diseases.

An interesting preliminary observation is that the size of the salivary gland ductal system in children treated for salivary stones seems to be not much smaller than in adults. The available literature on the size of the ductal system is limited to adults.[10] The evolution in endoscopic technology has permitted the development of sialendoscopes of 0.9 mm, which allow a deeper and more complete exploration of the ductal system, as well as the treatment of even distal stenosis or sialolithiasis.

Although we did not report in this article our experience with recurrent juvenile parotitis cases, one of the presented cases was initially diagnosed as a juvenile parotitis. A stenosis was diagnosed and successfully treated. One possible explanation for these juvenile cases could also be the presence of small sialoliths. Indeed, none of the two stenoses was diagnosed by conventional radiologic methods.

Of the six cases of sialolithiasis, only four had been diagnosed with previous radiologic methods. The two undiagnosed stones were of 2 mm in diameter. The diagnosis with classical radiologic methods depends on the size of the stone, with 2 mm being the limitations inferior limit of detection of all radiologic methods (ultrasound, computed tomography scan, magnetic resonance sialography).[9] Because some of these investigations require general anesthesia, we emphasize the role of sialendoscopy as a diagnostic procedure.

Most authors[2,4] report cases of pediatric submandibular stones treated by the classical approach: stimulation of salivary secretion caused by induced spontaneous passage of the duct, sialolithotomy, or sialadenectomy.

Shock wave extracorporeal lithotripsy has a global success rate of 70%.[11] However, there is no specific data about pediatric sialolithiasis lithotripsy.

None of the eight cases could evacuate a stone after stimulation of salivary secretion.

Sialolithotomy of the duct is possible for submandibular stones when it can be touched by bidigital palpation. In case of sialoliths of the posterior part of the gland, sialadenectomy is the common treatment with a risk of damage to the lingual nerve of 1%.[12]

Sialendoscopy also proved to be a valuable therapeutic method for pediatric ductal obstructive pathologies. Interventional sialendoscopy allowed us to retrieve five of six stones and dilate two stenoses under endoscopic control. However, it is important to differentiate the sialendoscopic procedures described here from the blind retrieval of sialoliths.[10,13]

Possible complications of diagnostic or interventional sialendoscopy are hemorrhage and perforation. In our series, one perforation occurred, and the stone was extruded through the ductal orifice causing, thankfully, no consequence. Careful manipulation of the endoscopes and the baskets and forceps might avoid these complications and potentially more significant consequences, especially in submandibular cases.

CONCLUSION

Diagnostic sialendoscopy is a new technique for a reliable evaluation of salivary ductal disorders in children, associated with a low morbidity. Interventional sialendoscopy allows early treatment of pediatric sialolithiasis and stenosis in most cases, and avoids classical open surgery approaches. This conservative approach is particularly important in children.

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