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A retrospective case series illustrating a possible association between a widened hilum and sialolith formation in the submandibular gland[☆]

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ABSTRACT

Objective: The purpose of this study was to explore any association between anatomical variances in the ductal system and sialolith formation using sialoendoscopy and acrylic resin replication of the ductal system.

Methods: A retrospective study of 372 submandibular gland sialoendoscopies was performed to review the findings of the submandibular gland duct anatomy. Using sialoendoscopy and replicated casts, a high rate of hilar widening was noted in patients with submandibular sialolithiasis.

Results: Sialolithiasis was detected in 326 of the patients who presented with obstructive symptoms. Around 67% (285/426) of the stones were located in the distal third of the ducts or at the hilum of the submandibular gland. During the sialoendoscopic procedure, the anatomy of the ductal system was examined and 285/326 (87.4%) of the hilums were noted to be widened like a basin. The anatomy of the duct from the replicated casts demonstrated a treelike structure and the basin-like widening of the hilum was found in all the excised submandibular glands.

Conclusion: Using sialoendoscopy, a high number of patients presenting with sialolithiasis in the submandibular gland seem to have an anatomical variance in the hilar region. The reproduced ductal system from excised glands also demonstrated this abnormal widening of the hilum. Although further studies need to be performed, we try and explain why there is such a high prevalence of hilar widening in patients with submandibular sialolithiasis.

1. Introduction

Salivary gland diseases are relatively common (Kuczkowski et al., 2010; Yilmaz et al., 2010; Lukšić et al., 2011; Lukšić et al., 2012). The most frequent non-neoplastic salivary disorder is obstructive sialadenitis. Obstructive salivary disease may be caused by physical impediments or strictures in the ductal system and is commonly found in the submandibular or parotid gland. Sialolithiasis is the main cause of obstructive salivary disease. It accounts for 66% of obstructive cases and about 50% of all major salivary gland disease (Capaccio et al., 2007). The incidence of salivary calculi is 60 million cases per year, and it affects 1.2% of the general population as demonstrated by postmortem studies (Marchal et al., 2001; Nahlieli et al., 2006; Maresh et al., 2011). Other reports state that 1:10,000–30,000 are affected every year (Gillespie et al., 2011; Huoh and Eisele, 2011). The cause of sialolithiasis is not completely understood but it has been suggested that they occur as a result of deposition of calcium salts around a central nidus of desquamated epithelial cells, foreign body, products of bacterial decomposition, microorganisms, or mucous plugs (Ouellette and Slack, 2003; Kasaboglu et al., 2004; Al-Abri and Marchal, 2010; Jardim et al., 2011; Oteri et al., 2011).

The submandibular gland has a high rate of sialolith formation when compared to the other salivary glands (Kasaboglu et al., 2004; Emir et al., 2010; Jardim et al., 2011; Oteri et al., 2011). Some have attributed this higher rate to the distance the saliva has to travel from the gland to the duct orifice, the alkaline and viscous nature of

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the saliva, and angulations formed by the duct anatomy (Emir et al., 2010; Jardim et al., 2011).

The majority of calculi are located in the distal third of the duct or at the hilum of the gland; pure intraparenchymal stones are uncommon (Lustmann et al., 1990). The striking difference between the parotid and submandibular glands is the tortuous and the upward path the secretory products of the submandibular gland has to travel as well as the type of secretion (serous versus mucous) (Lustmann et al., 1990; Capaccio et al., 2007; Zhang et al., 2010).

A retrospective study was performed to review the findings of the submandibular gland duct anatomy involved in obstructive disease. Using sialoendoscopy and replicated casts, a high rate of hilar widening was noted in patients with submandibular sialolithiasis. We hypothesize that we will be able to find the relationship between a widened hilum and sialolith formation in the submandibular gland.

2. Materials and methods

2.1. Sialoendoscope procedure

During the previous 6 years, we performed 372 submandibular gland sialoendoscopies on 233 male patients and 139 female patients between 12 and 79 years of age who presented with obstructive symptoms. All patients underwent preoperative and postoperative screening using plain radiography, sialography, ultrasound, CT or MR sialography. Postoperatively, the patients were seen at 1 week and in 1-month intervals up to a year. The sialoendoscopies were performed under local anaesthesia, and the length of the procedure ranged from 30 to 90 min.

Sialoendoscopic procedures were performed through the main duct. A 1 mm sialoendoscope was used for all procedures (Karl Storz GmbH, Tuttlingen, Germany). An exploration unit with a 1.3 mm outer sleeve, and a surgical unit with a 1.1×1.3 mm outer sleeve with three channels were also used for this procedure. The channels were utilized to introduce surgical instrumentation and an irrigation port device to perform necessary operations during the sialoendoscopic procedure. Details of the surgical technique have been previously described (Nahlieli et al., 2003; Yu et al., 2010). Special care was taken to avoid trauma to the ductal walls and perforation. Some of the surgical instrumentation utilized included a basket for sialolith retrieval and forceps. In cases of obstruction due to a sialolith, removal was first attempted without fragmentation using the basket, irrigation as a mobilization technique, or forceps. If a stone remained wedged within the duct or was too large to be removed with the aforementioned techniques, mechanical pulverization using forceps was performed followed by removal of the fragments. During the procedure, the hilum of the submandibular gland in all the patients was examined and photographed digitally.

2.2. In vitro replication of the submandibular glands and its ducts

In 10 patients, the sialoliths was found in the hilum or in the parenchyma. These patients underwent excision of the submandibular gland. After obtaining informed consent from the patient and approval from the local ethics committee, the submandibular ductal systems of these patients were replicated using acrylic resin for examination. The submandibular glands were collected from 2001 to 2006 at the Department of Oral Maxillofacial Surgery, Shanghai Ninth Hospital. Five were from male patients and five were female patients with a mean age of 45 years.

To replicate the ducts, a cannula with normal saline was inserted into the ducts. Then, a 33% methacrylate solution was perfused throughout the Wharton ducts to prepare the duct casts. The perfused glands were then placed at room temperature for 48 h to allow for sufficient hardening time. They were then placed in 40% HCl for 1 week and rinsed in distilled water to remove the nonmethacrylate-perfused soft tissues of the gland and air-dried to form the final casts. These casts were analyzed for the branching patterns, lobule number, and the shape of the hilum.

3. Results

Of the 372 submandibular gland sialoendoscopies performed, the main cause of obstruction was sialolithiasis. Calcified stones were found in 326 submandibular glands (87.6%). A total of 426 calculi were removed from the ductal system. A single calculus was found in 262 (80.4%) submandibular glands, and multiple stones within Wharton's duct were found in 64 cases (19.6%). Two hundred eighty-five stones (66.9%) were located in the distal third of the ducts or at the hilum, 101 stones (23.7%) were located in the middle-third of the ducts, and 40 stones (9.4%) were located in the proximal third of the ducts. In 64 cases (19.6%) occult radiolucent calculi were found in the hilum of the submandibular gland.

The hilum of the gland was also examined in all the patients using sialoendoscopes. The sialoendoscopic exam revealed that, in 285 cases (87.4%) the hilum was a widened basin-like structure as opposed to a bifurcation or a trifurcation.

Twenty-eight patients who had undetectable calculi on plain radiography underwent MR sialography prior to sialoendoscopy. Of 28 patients, the radiolucent calculi were detected in 12 cases, and the basin-like structure of the hilum was found in 13 glands. The widening of ducts was also seen during conventional sialography (Fig. 1).

The resin replication of the ducts revealed a treelike structure. The main duct originated from the hilum and separated into several main branches, which entered the lobules through the lobular hilum. At various levels, all the replicated ducts showed similar features in all 10 duct casts. The diameters of the ducts decreased gradually from the main branches to the terminal branches. The basin-like structure of the hilum was also noted in all the replicated ductal systems (Fig. 2).

4. Discussion

The exact pathogenesis of sialolithiasis remains unclear, and various hypotheses have been proposed. They all propose that there is an initial organic nidus that progressively increases in size by deposition of inorganic and organic substances, occurring in two phases (Marchal et al., 2001). Organic substances include glycoproteins, mucopolysaccharides, and cellular debris. The inorganic components are comprised of calcium carbonates and calcium phosphates (LeGeros, 1991). Other factors, such as changes in pH, reduced salivary flow, or increases in calcium concentrations, are seen as contributing factors to the precipitation of calcium (Marchal et al., 2001).

The majority of calculi are located in the distal third of the duct or at the hilum of the gland; pure intraparenchymal stones are infrequent (Capaccio et al., 2007). Our investigation had similar findings: roughly 67% (285/426) of the stones were located in the distal third of the ducts or at the hilum of the submandibular gland. Other studies also found that 56% of the (35/64) sialoliths were located in the hilum area of submandibular gland (Iro et al., 2009; Su et al., 2010; Luers et al., 2011). The sialoliths in the hilum area were also noted to be larger than the sialoliths in the main duct (Iro et al., 2009; Su et al., 2010; Luers et al., 2011).