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American Journal of Otolaryngology–Head and Neck Medicine and Surgery 32 (2011) 85–87
American Journal of OTOLARYNGOLOGY
www.elsevier.com/locate/amjoto

Bilateral giant submandibular sialoliths and the role for salivary endoscopy

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Received 19 September 2009

Abstract Salivary stones larger than 15 mm are classified as giant sialoliths. They are uncommon in the practice of otolaryngology, and their management has always been a therapeutic challenge. Traditionally, when they cannot be retrieved by marsupialization, removal of the salivary gland has been advocated. Sialendoscopy and the recent development of combined endoscopic and external approaches for extraction of large stones with preservation of the major salivary glands are promising. We present the first case of simultaneous bilateral giant sialoliths, and the first report that associates giant sialoliths and the use of salivary endoscopy. In this case, both giant stones were removed with the assistance of a salivary endoscope and without removing the submandibular glands.

1. Introduction

Sialolithiasis is the most common disease of the salivary glands, with an incidence of approximately 1.2% in the adult population [1,2]. The average stone size is 3.2 mm for the parotid gland and 4.9 mm for the submandibular gland [3]. Sialoliths that exceed 15 mm in any one dimension or 1 g in weight have been classified as giant [4,5]. In the normal gland, the diameter of the Wharton and Stensen ducts is approximately 3 and 4 mm, respectively [3].

Some authors have analyzed the characteristics of different giant sialoliths reported in the literature [2,5]. Bodner [5] found only 14 well-described cases in his review published in 2002. We found that all giant sialoliths documented in the English language have been unilateral [2,5] and present the first case, to the best of our knowledge, of simultaneous bilateral giant sialoliths. Both giant stones were removed with the assistance of a salivary endoscope and without removing the glands. This is also the first report that associates giant sialoliths and the use of salivary endoscopy.

2. Case report

A 69-year-old man presented to our office complaining of chronic unrelenting purulent discharge from bilateral submandibular ducts despite appropriate medical therapy. A computed tomographic scan revealed large bilateral radiopaque masses at the hilum of each submandibular gland, consistent with salivary stones (Fig. 1). The masses were palpable intraorally. A staged transoral sialolithotomy approach was planned and performed bilaterally. After the stones were removed, a salivary endoscope was used to explore the main duct, the stone cavity, and the distal ductal system. Exploration was made through the main duct via a transpapillary approach and also through the intraoral sialolithotomy opening. Small pieces of the larger stones and/or smaller stones were found lodged in the intraglandular ducts immediately distal to the cavity, which were removed under direct visualization. Once the calculi were removed, the salivary duct and soft tissues were closed in layers at the end of each procedure. The patient tolerated both outpatient operations without complications and is currently asymptomatic 1 year after his last intervention. The right and left stone measured 2 × 1.5 × 1.5 and 2.3 × 1.6 × 1.3 mm, respectively (Fig. 2).

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doi:10.1016/j.amjoto.2009.09.011

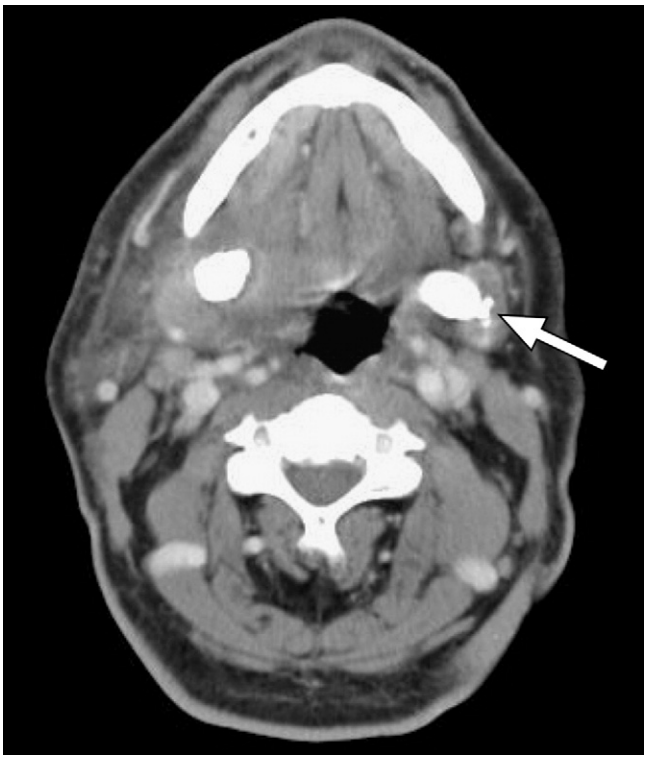

Fig. 1. Computed tomographic scan—axial view. Large bilateral radiopaque masses are seen at the hilum of each submandibular gland, consistent with salivary stones. Please note the oval shape and projections into the ductal system bifurcation that are seen on the stone of the left side (white arrow).

3. Discussion

The etiology of salivary calculi still remains poorly understood. They are composed of organic and inorganic substances, such as microcrystalline apatite or whitlockite [1]; and having multiple in a single glandular system is not uncommon [6]. Giant sialoliths likely share the same pathogenesis as its smaller counterparts and are uncommon in the practice of otolaryngology. It is probable that the calculi in this patient had grown for many years, as it is believed that salivary stones grow at a rate of 1 [3] to1.5 mm per year [4]. The literature traditionally reveals that more than 80% of salivary stones are located in the submandibular gland or its duct, 6% to 15% in the parotid, and approximately 2% in the sublingual and minor salivary glands [1]. However, some authors have more recently reported that up to 40% of salivary calculi referred for treatment to a subspecialty center occurred in the parotid gland [3].

The management of large salivary gland calculi has always been a therapeutic challenge. Traditionally, when they cannot be retrieved by marsupialization, removal of the gland has been advocated [7]. Sialendoscopy and the recent development of combined endoscopic and external approaches for extraction of large stones with preservation of the major salivary glands are promising [7]. Sialendoscopy allows the endoscopic intraluminal visualization of major salivary glands, and offers an opportunity to diagnose and treat inflammatory and obstructive pathology related to the ductal system. Under direct visualization, small stones can be retrieved endoscopically using wire baskets; and ductal stenosis can be dilated with balloons. In a large patient series, this technique has been validated for its utility and safety for diagnosis and treatment of salivary gland ductal pathology [8].

It has been demonstrated that successful results of interventional sialendoscopy are related to the size of the stones and ducts in both submandibular and parotid glands. Size is probably the most determinant factor in predicting success [3]. According to Marchal [3], 93% of calculi in the parotid can be removed with wire baskets if smaller than 3 mm; however, the success rate for extraction decreases to 35% for larger stones. Fragmentation with laser or other methods and subsequent basket extraction are recommended for calculi up to 8 mm; but for giant stones, a combined approach has been advocated [3]. A combined approach consists of the combination of classic noninvasive sialendoscopy plus open sialolithotomy. The patient presented in this case underwent bilateral combined approaches and did not require removal of the submandibular glands.

The salivary endoscope has been a major advancement in our approach to sialoliths. In a review finished in 2007, our overall success rate for endoscopic stone removal was 74% [8]. One of the advantages of the endoscope is the easier localization of salivary stones. However, in this case, the salivary endoscope was not used to locate the stones because the sialoliths were palpable and too large to be delivered endoscopically. One of the stones found in this patient had projections into the ductal system (not described before for giant sialoliths), which could have been potentially fragmented during extraction and left inside the ducts. In this patient, the salivary endoscope permitted the exploration of

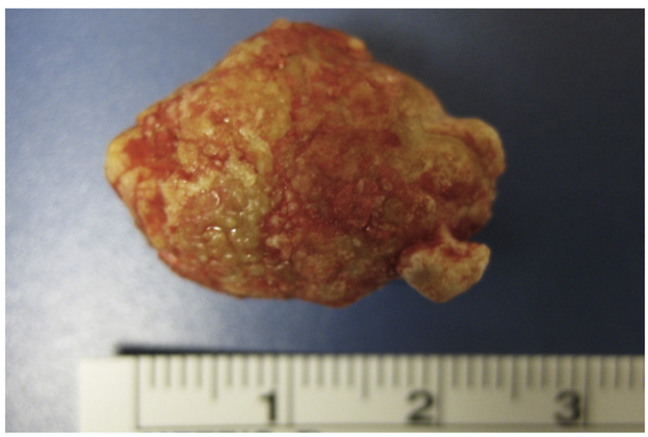

Fig. 2. Left giant submandibular sialolith. 2.3 × 1.6 × 1.6 cm in greatest dimension. Projections of the stone correlate with the computed tomographic scan (Fig. 1).

the ducts and the stone cavity both proximally and distally, which guided a more precise retrieval of fragments and/or additional smaller stones, which otherwise would not have been easily identified.

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