INTRODUCTION

Sialolithiasis is the most common disease of the salivary glands with an incidence of approximately 1.2% in the adult population. The literature traditionally documents that more than 80% of stones are located in the submandibular gland or its duct, 6-15% in the parotid and approximately 2% in the sublingual and minor salivary glands. However, some authors have more recently reported that up to 40% of salivary calculi, referred for treatment to a sub-specialty center, can occur in the parotid gland. Salivary stones are composed of organic and inorganic substances, such as microcrystalline apatite or whitlockite. The average size is 3.2 mm for the parotid gland and 4.9 mm for the submandibular gland. Sialoliths that exceed 15 mm in any one dimension or 1 gram in weight have been classified as giant. In the normal gland, the diameter of the Wharton’s and Stensen’s ducts are approximately 3 mm and 4 mm respectively.

Recent reviews have examined the characteristics of different giant sialoliths reported in the literature. Bodner et al. found only 14 well described cases in his review published in 2002. All giant sialoliths documented in the English literature have been unilateral. We 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.

CASE REPORT

A 69 year old patient was referred for evaluation of a chronic unremitting purulent discharge from bilateral submandibular ducts despite appropriate medical therapy. A computerized tomography (CT) scan showed large bilateral radiopaque masses at the hilum of each submandibular gland, consistent with salivary stones (figure 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, through the main duct via its papilla, and through the sialolithotomy opening as well. Small pieces of the larger stones and/or smaller stones were found lodged in the intra-glandular ducts immediately distal to the cavity, and were removed under direct visualization. The salivary duct and soft tissues were closed in layers at the end of each procedure. The patient tolerated the out patient procedure without complications. He is currently asymptomatic six months after his last procedure. The right and left stones measured 2.6 x 1.6 x 1.4 and 2.3 x 1.6 x 1.3 respectively.

DISCUSSION

Despite the very extensive literature, the pathogenesis of salivary calculi still remains poorly understood. Giant sialoliths likely share the same pathogenesis and are uncommon in the practice of otolaryngology. It can be assumed that the calculus in this patient had grown for many years, as it is believed that salivary stones grow at a rate of 1 to 1.5 mm per year. 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 was mandatory.

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. 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.

It has been demonstrated that results of interventional sialoendoscopy 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. According to Marchal, 93% of calculi in the parotid can be removed with wire baskets if smaller than 3 mm, however, the removal rate for sialoliths larger than 3 mm in the parotid is 35%. For stones up to 8 mm, fragmentation with laser or other methods and subsequent basket extraction is recommended, but for giant stones, a combined approach has been advocated. A combined approach consists in the combination of classic non-invasive sialoendoscopy plus an open sialolithotomy. The patient presented in this case underwent bilateral combined approaches. 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%. 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, since 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, which could have been potentially fragmented and left inside the ducts during a forceful extraction. In this patient, the salivary endoscope permitted the exploration of 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.

CONCLUSIONS

Salivary endoscopy is useful in the treatment of large salivary stones, as it allows duct exploration and retrieval of smaller intra-ductal fragments during combined approaches. This is the first case reported of bilateral giant sialoliths in the literature.

REFERENCES