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Case Report

Chronic sialadenitis with sialolithiasis associated with parapharyngeal fistula and tonsillolith

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ABSTRACT

Sialolithiasis is a common salivary pathology, suggested to affect over 1% of the population by postmortem studies. An uncommon complication of sialadenitis and sialolithiasis is the formation of fistulous tracts to other cervicofacial compartments. Submandibular gland sialocutaneous and sialo-oral fistulae have been sparsely described, but a sialo-pharyngeal fistula manifesting as a tonsillolith has yet to be described. We present an unusual case of a 35-year-old male presenting with recalcitrant neck pain and a presumed tonsillolith in the background of chronic submandibular sialadenitis, subsequently demonstrating a salivary fistula through the parapharyngeal space. We offer a thorough review of the literature to highlight the possibility of migratory sialolithiasis and its complications.

Introduction

Sialolithiasis is common, affecting approximately 1% of the population. The submandibular gland (SMG) is most frequently affected, as 80%-90% of stones precipitate in Wharton's duct [1]. Although sialolith composition varies by gland, concretions contain both inorganic and organic material. Most submandibular sialoliths are composed of primarily inorganic components including hydroxyapatite, brushite, octacalcium phosphate, or whitlocke. Stone formation is a multifactorial process influenced by salivary stasis, changes in saliva biochemistry and aggregation of calcium phosphate microsialoliths [2]. Stasis has also been associated with the evolution of more mucoid saliva, forming a lattice for calcium salt deposition. No relationship between serum concentrations of calcium or phosphorus and susceptibility to sialolithiasis has been identified. Sialolithiasis may be entirely asymptomatic. Chronic and acute suppurative sialadenitis are distinct entities. In fact, approximately one-third of patients with chronic sialadenitis will have no discernible sialoliths on endoscopy or imaging [3].

We describe an unusual case of a patient with chronic SMG sialadenitis, ultimately discovered to have a sialolith in the tonsillar fossa resembling a native tonsillolith. This report reviews key imaging and clinical features to alert clinicians to

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the entity of migratory sialolithiasis, a consideration rarely discussed in the literature.

Case report

We present the case of a 35-year-old male who initially presented to our department with recurrent bouts of right neck pain and submandibular swelling. The neck computed tomography scan revealed a 1.7 × 1.0 cm dense opacity in the right tonsillar fossa suggestive of a large tonsillolith (Fig. 1A-C). Flexible endoscopy demonstrated submucosal swelling in the right tonsil with partial exposure of an underlying stone. Of note, there was no radiographic evidence of sialadenitis or intraparenchymal sialolithiasis. His symptoms were attributed to the large tonsillolith and the decision was made to proceed with tonsillectomy. The patient had an unremarkable initial recovery. However, about 1 month post-tonsillectomy, he again experienced right neck pain and submandibular swelling. Low-grade symptoms were constant but significantly exacerbated by eating. On examination, no purulence was expressed from the sublingual caruncle with SMG massage and no stone was palpable in the floor of mouth. The patient's odynophagia and edema did not resolve with conservative measures, and a repeat computed tomography scan was obtained. This revealed a 3-cm fistulous tract extending from the right SMG to the right parapharyngeal space adjacent to the tonsillectomy site (Fig. 1D). The patient subsequently underwent SMG excision. Intraoperatively, the gland was noted to be indurated. During dissection, a small amount of turbid drainage was expressed in the region between the gland and the parapharyngeal space, distinct from the submandibular duct which had been identified and isolated. The final pathologic diagnosis described chronic inflammatory changes and atrophy consistent with chronic sialadenitis. The patient reported complete resolution of his neck pain at the 1 month postoperative follow-up visit.

Discussion

Although sialoliths are normally confined to the intra- or extraparenchymal salivary ductal system, our case describes

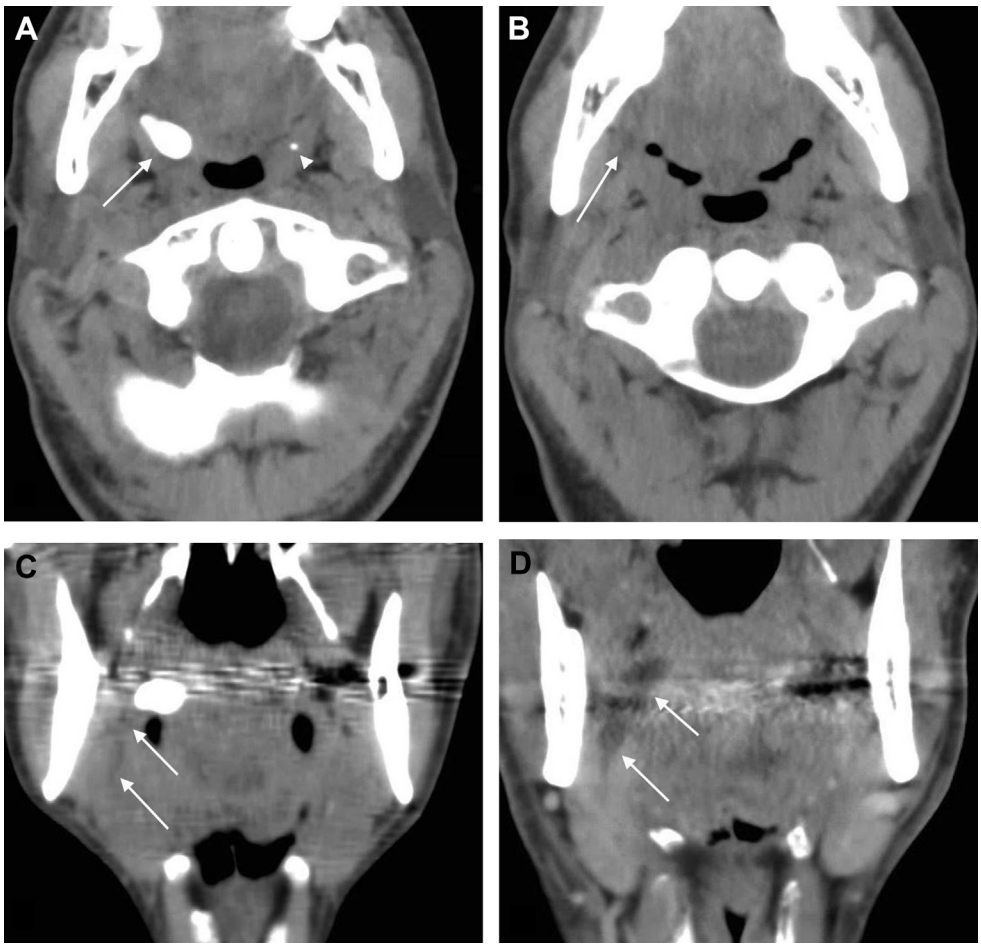

Fig. 1 – Axial noncontrast neck computed tomography (CT) performed at an outside institution revealed a 17 × 10 × 12 mm calcification in the right tonsillar fossa (A, arrow) and a punctate calcification on the left (A, arrowhead). On the right, ductal distention in the right submandibular gland was not prospectively recognized to extend to the calcification (B and C, arrow). After right tonsillectomy and stone removal, the patient had continuing right submandibular swelling. Subsequent neck CT with contrast revealed increased size of the fluid tract from the right submandibular gland to the right parapharyngeal space (D, arrow), which was then excised.