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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.

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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 whitlockite. 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

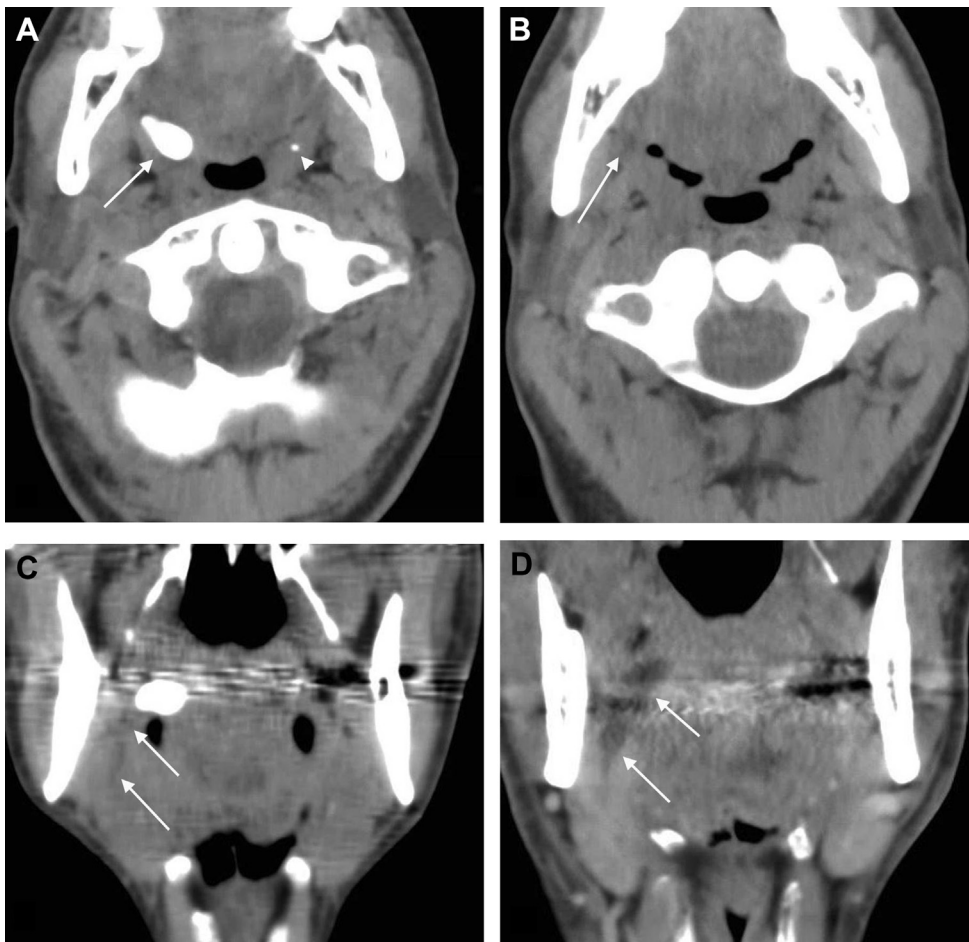


Fig. 1 – Axial noncontrast neck computed tomography (CT) performed at an outside institution revealed a $17 \times 10 \times 12$ mm calcification in the right tonsillar fossa (A, arrow) and a punctate calcification on the left (A, arrowhead). On the right, ductal distention in the right submandibular 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.

submandibular sialolith that had migrated to the tonsillar fossa with a fistulous connection to the SMG, initially confounding the correct diagnosis. By the authors' investigation, this is the first report in the literature describing this particular course of sialolith migration. More often, SMG sialoliths are missed because of radiolucency (in fact, it is suggested that 20% of submandibular stones are missed on imaging) or spontaneous extrusion [4]. Much less often does a submandibular sialolith elude diagnosis because of fistulous migration to a separate deep neck space (eg, parapharyngeal space). This phenomenon warrants discussion to better anticipate this potential, albeit uncommon complication.

Sialo-oral, if not sialo-pharyngeal, fistulae are well-described in the literature. Analogous to cholecystocholedochal and ureterocolic fistulae in which stone formation is the culprit, chronic inflammation (sialadenitis) and pressure-induced necrosis/ischemia caused by the sialoliths lead to fistulous tracts. Kieliszak et al [5] describe 2 cases of sialo-oral fistulas with associated sialoliths resulting in Wharton's duct atrophy precluding endoscopic cannulation. Saluja et al [6] comment on SMG ductal giant sialoliths (>15 mm in any dimension) and their tendency to cause intraoral fistulae by direct extension or the creation of a suppurative tract, reporting specifically on a 2.1 × 1.4 cm sialolith. Stanford [7] reported a patient with recurrent spontaneous large stone extrusion, where a barium swallow obtained for evaluation of epigastric pain incidentally highlighted a posterior floor-of-mouth branching sialo-oral fistula.

Indeed, floor of mouth sialo-oral fistulae are not particularly remarkable given the location of the gland and Wharton's duct. Much less common are submandibular sialolith-associated fistulae to other cervicofacial compartments. Kishore Kumar et al [8] identified 200 patients evaluated over 6 years for cervicofacial fistulae, and found only one case (0.5%) was surely attributable to SMG sialolithiasis. Drage et al [9] report on 2 patients with chronic submandibular sialadenitis presenting with transcervical sialocutaneous fistulae, wherein a sialolith was identified fluoroscopically and removed from superficial soft tissues at the level of the thyroid cartilage. Paul and Chauhan [10] recount the case of a 4.5 cm SMG sialolith that caused a sialo-oral fistula in the anterior floor-of-mouth, while also being associated with a draining, cervical cutaneous tract (ie, a sialo-oro-cutaneous fistula). Singh et al [11] describe their management of a patient in whom an x-ray fistulogram revealed a tract extending from skin in the submandibular region to a sialolith. The mechanism of sialo-cutaneous and sialo-oral fistulae formation are likely similar. However, the proximity of the floor of mouth mucosa and Wharton's duct, in addition to the presence of the mylohyoid muscular sling intervening between Wharton's duct and cervical skin, makes the floor of mouth a much more likely egress point for submandibular sialoliths.

The simple coincidence of salivary and tonsillar stones is described in the literature. Giudice et al [12] describe the case of a 1.0 cm tonsillolith in a patient with a known history of chronic sialadenitis, but do not mention the presence of a corresponding fistula. Suarez-Cunquero et al [13] detail the incidental radiographic finding of multiple ipsilateral small tonsillar stones in a patient with symptomatic submandibular sialolithiasis, remarking on the potential similarity in the pathogenesis of tonsillo- and sialolithiasis.

Table 1 – Literature summary of cervicofacial salivary fistulae.

Author	Dates	Type of article	No. of patients	No. of stones	Age, y	Sialolith dimensions, cm	Greatest dimension, cm	Fistula	Imaging modality
Jayachandran [15]	2011	Case report	1	1	52	2.6 × 2.1 × 2.0	2.6	Sialo-oro-cutaneous	Radiography, CT, MRI
Drage [9]	2004	Case series	3	3	52.3	2	2.0	Sialocutaneous	Case 1—CT, Case 2—radiography, Case 3—sialogram
Kurtoglu [16]	2015	Case report	1	1	52	2.5 × 0.8 × 0.6	2.5	Sialo-oral	None described
Rauso [17]	2012	Case report	1	1	56	5.6	5.6	Sialo-oral	None described
Singh [18]	2015	Case report	1	1	53	1.0 × 1.0	1.0	Sialocutaneous	Fistulography, CT
Rangappa [19]	2014	Case report	1	1	55	1.0 × 1.0	1.0	Sialocutaneous	Ultrasound, radiography
Sutay [20]	2003	Case report	1	1	22	3.7 × 0.7	3.7	Sialo-oral	Radiography
Almasri [21]	2005	Case report	1	1	70	2.3 × 1.7	2.3	Sialocutaneous	Radiography, CT
Kasat [6]	2012	Case report	1	1	65	2.1 × 1.4	2.1	Sialo-oral	Radiography
Panuganti (current case)	2017	Case report	1	1	35	1.7 × 1.0	1.7	Sialo-oro-pharyngeal	CT
Pirkil [14]	2015	Case report	1	1	32	3.1 × 2.6	3.1	Sialo-oro-pharyngeal	CT
Paul and Chauhan [10]	1995	Case report	1	1	45	4.5 × 2.0	4.5	Sialo-oro-cutaneous	Radiography
Parkar [22]	2012	Case report	1	1	84	1.6 × 1.0	1.6	Sialo-oral	Radiography
Karenger [23]	1998	Case report	1	1	64	0.4 × 0.5	0.5	Sialocutaneous	Unspecified

CT, computed tomography; MRI, magnetic resonance imaging.

Our patient had a sialolith that we believe extruded from the SMG, migrating to the tonsillar fossa, creating a fistulous tract between the 2 regions. This process has not previously been attributed to inflammatory sialadenitis. Pirkel et al [14] recount the case of a symptomatic tonsillolith that required a tonsillectomy, during which a salivary “fistula” was identified as a discrete entity in the inferior pole of the tonsillar fossa. The authors posit that the fistula was in fact a large accessory SMG duct, a congenital phenomenon that is also uncommonly described in the literature, and that the tonsillolith was salivary in origin. In that case, tonsillar inflammation is the proposed etiology of minor salivary gland outflow tract occlusion resulting in tonsillolithiasis. Our patient failed tonsillectomy as a primary intervention and had a constellation of initial and recalcitrant symptoms more suggestive of chronic sialadenitis. Moreover, we were unable to identify a well-defined epithelial-lined duct between the SMG and parapharyngeal space at the time of SMG excision that would be consistent with the congenital accessory duct. Accordingly, we favor chronic inflammation and pressure as the etiology of the sialo-oropharyngeal fistula in our patient. Kieliszak et al [5] also highlight the two possibilities, congenital ductal duplication and chronic inflammation, as alternative pathogeneses of sialo-oral fistulae.

It is worth noting that the average greatest dimension of sialoliths associated with fistulae in our literature review (Table 1) was 2.4 cm (range 0.5–5.6 cm). Sigismund et al [24] review the characteristics of over 2300 submandibular sialoliths, indicating that the average stone diameter in symptomatic patients was approximately 0.83 cm. It is likely that this population contained at least a small cohort with salivary fistulas, but it is reasonable to assert that sialofistulae are more likely to be associated with larger sialoliths.

In summary, a salivary fistula is an uncommon but noteworthy complication of SMG sialolithiasis and sialadenitis. Understanding the anatomical relationship between the submandibular space and other cervicofacial compartments, clinicians can anticipate fistulae and appropriately direct treatment toward the underlying salivary gland pathology.

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