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CASE REPORT

An unusual case of bilateral submandibular sialolithiasis in a young female patient

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Summary Salivary calculi in the pediatric population comprise only 3% of all cases of sialolithiasis. In addition, the presentation of bilateral calculi in children is rare. The formation of stones in the salivary glands has been attributed to slow salivary flow, salivary stagnation and unknown metabolic events. There is no report in the literature of an association between dietary calcium intake and calculi formation. We describe the clinical findings and management in a patient with bilateral sialolithiasis thought to be linked to high dietary calcium intake and review the literature.
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1. Introduction

While the incidence of sialolithiasis peaks in middle-aged adults, it is a rare condition among children [1—3]. Furthermore, bilateral sialolithiasis in the pediatric population is extremely uncommon. A review of all reports of sialolithiasis in children and adults from 1913 to 1989, reported bilateral sialolithiasis in 0.5—2.2% of all salivary stones [2]. The precise etiology is unknown, however, it is thought that stagnation of calcium-rich saliva combined with unknown metabolic events contribute to calculi formation [4]. To date, a link between dietary calcium and sialolithiasis has not been established. A trial of conservative therapy usually precedes surgical intervention. Novel modalities of treatment are also emerging.

We describe the clinical findings and management in a patient with bilateral sialolithiasis potentially associated with high dietary calcium intake and review the literature.

2. Case report

An 11-year-old girl was referred to the Hospital for Sick Children in Toronto for the management of sialoadenitis of her submandibular glands.

She presented to her family physician with two episodes of peripheral limb fractures sustained from traumatic injuries. Suspecting osteogenesis imperfect or osteoporosis, a regimen of calcium supplementation, including calcium carbonate, Vitamin D and increased dietary intake of calcium-rich products, such as milk, cheese and yogurt was recommended. Two months later, the girl returned with new onset abdominal pain related to her calcium-rich diet and was advised to consume up to one

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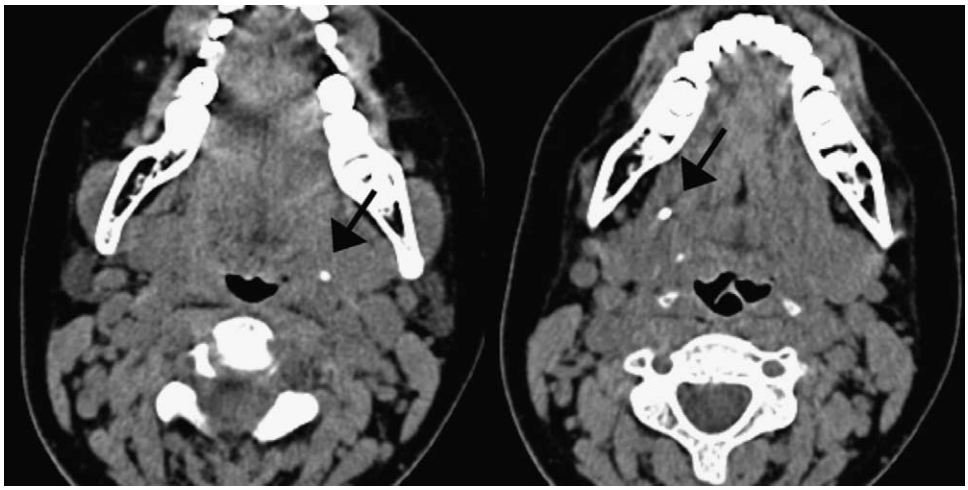

Fig. 1 Axial computed tomography scans demonstrating bilateral calcified densities (calculi) in the submandibular glands (arrows).

bottle of regular strength TUMS®, containing 500 mg calcium carbonate per tablet, daily. She presented once more within weeks complaining of pain and diffuse swelling in her left and right submandibular regions. CT scan (Fig. 1) performed at that time revealed bilateral 2—3 mm calcified masses in the left and right submandibular ducts. The patient was referred to an endocrinologist who noted that calcium metabolism studies, and serum and urinary calcium were normal. There was no evidence of kidney or urinary tract stones. She was also seen by a general otolaryngologist, who made the diagnosis of bilateral submandibular sialolithiasis, recommending conservative treatment and referred her for further evaluation.

The girl was asymptomatic when she presented to our clinic. Intraoral examination did not reveal any submandibular swelling or tenderness. Both, Wharton's and Stensen's ducts were normal and patent with clear salivary flow produced on gentle palpation. There was no erythema, edema or tenderness of the orifices of either duct bilaterally and no calculi were palpable. The remainder of the head and neck exam was unremarkable.

The diagnosis of bilateral submandibular sialolithiasis was made and we concurred with the referring otolaryngologist's conservative approach given the child's lack of symptoms. Massage of the submandibular gland and increased hydration to stimulate salivary flow was recommended.

3. Discussion

Though incidence of sialolithiasis peaks in the third to sixth decades of life, only 3% of all cases, occur in children and sialolithiasis remains a rare cause of salivary dysfunction in pediatric populations [1—3]. Children with sialolithiasis also show a strong male preponderance, and on average are 10 years of age [1—3].

Despite high calcium content of sialoliths, serum calcium or phosphate levels are not believed to be associated with stone formation. Calcium phosphate and carbonate, in the form of hydroxyapatite, constitute the major elements of salivary stones, along with other salts (Mg, Zn, NH_3) and organic material [3]. Morphoanatomic factors (i.e. salivary duct stenosis, ductal epithelial inflammation or injury, foreign body impaction) producing stagnation of calcium-rich saliva and foci for calcium deposition, and poorly understood metabolic events that increase salivary bicarbonate content, decrease calcium phosphate solubility and favor precipitation of calcium and phosphate, are believed to contribute to stone generation [3].

Sialoliths most commonly occur in the submandibular glands (80—92%), but are also found in the parotid glands (6—20%), and the sublingual and minor salivary glands (1—2%) [5]. Several reasons favor submandibular over parotid gland stones: (1) longer duct, (2) slower, anti-gravity salivary flow, (3) higher salivary calcium and (4) more alkaline and viscous secretions [3]. Submandibular stones are consistently found in Wharton's duct (75—85%), while parotid stones occur equally in the parenchyma and duct [2,3]. Bilateral sialolithiasis is rare. One of the largest series on pediatric sialolithiasis reported only a single case of bilateral sialolithiasis over 13 years [6]. A review of all reports with 50 patients or more on sialolithiasis in children and adults from 1913 to 1989, reported bilateral sialolithiasis in 0.5—2.2% of all salivary stones [2].

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