ABSTRACT

Salivary duct obstruction is most commonly due to sialolithiasis, but this is less common in children. This presents with unilateral, episodic post-prandial swelling and pain in the affected parotid or submandibular gland.

Venous malformations located in the submandibular region can present with swelling, and initial radiologic workup with either CT scan or plain radiographs can reveal multiple calcified densities characteristic of phleboliths, which instead may be interpreted as sialoliths.

We present two patients with venous malformations who were referred with a presumed diagnosis of submandibular sialolithiasis.

CASE REPORTS

Patient 1: A 14 year old girl was referred for management of presumed submandibular gland sialolithiasis. She was initially evaluated for an asymptomatic right submandibular gland enlargement three years before presentation to us (Figure 1A). Prior workup had included an ultrasound and computed tomography (CT) (Figure 1B) which were interpreted as consistent with phleboliths located within the right submandibular gland. Fine needle aspiration cytology was inconclusive. Magnetic resonance imaging (MRI) one year prior to our evaluation was also interpreted as a sialolithiasis of the right submandibular gland. (Figure 1C, 1D)

She had no features of submandibular duct obstruction secondary to sialolithiasis, most notably no changes in swelling and no pain. Her right submandibular region, although minimally enlarged had remained stable in size and did not cause any pain. Review of her MRI showed a lobulated mass with increased signal on T2 weighted imaging, with multiple well defined hypodense areas consistent with phleboliths within a venous vascular malformation (Figure 1C, 1D). Since she had no symptoms at the time of our evaluation symptoms, observation was recommended.

Patient 2: A 7 year old girl was referred after excision of a non-tender left submandibular mass, with pathologic diagnosis as either a calcified fibrotic nodule or a salivary stone. After this surgery, she presented on two occasions to the emergency department at our institution for swelling in the left paramandibular area. CT demonstrated a left submandibular and paramandibular mass slightly hypodensitizing to muscle extending to the left masseter with a peripheral focus of calcification (Figure 2A, 2B). She was referred for evaluation of sialolithiasis. Her clinical history revealed intermittent, painless left submandibular swelling without pain, fever or eating difficulties. Examination was significant for non-tender, mild left submandibular and paramandibular fullness.

Due to her persistent swelling, an MRI was obtained in and this demonstrated a lobulated mass with hyperintense signal on T2 weighted images with a well defined hypodense area consistent with a phlebolith within a venous vascular malformation (Figure 2C, 2D). She subsequently underwent percutaneous sclerotherapy with ethanol (Figure 2E, 2F). She has required two additional sclerotherapy procedures to achieve reduction in the size of the malformation.

DISCUSSION

Three cases of vascular malformations mimicking submandibular gland obstruction have been reported, all in adults. In each of these cases, an initial diagnosis of submandibular gland sialolithiasis was made on the basis of plain radiographs or CT images. In two of these cases, surgical excision of the submandibular gland was performed, revealing a diagnosis of cavernous hemangioma containing multiple phleboliths. In the third case, sialendoscopy did not reveal any sialoliths, but instead showed prominent proliferation of vessels in the submandibular duct lining.

Pediatric vascular malformations are congenital lesions of aberrant vascular development, and are classified according to their predominant vessel type. They are present at birth and usually exhibit progressive, slow growth but may experience periods of rapid growth and infiltration into surrounding tissues. Venous malformations are a subtype of vascular malformations which were previously classified as cavernous hemangiomas. Physical examination usually reveals a compressible mass which can increase in size with Valsalva maneuvers. The overlying skin may have a bluish hue. MRI with contrast enhancement is the preferred imaging study for characterizing venous malformations, offering superior visualization of soft tissues, in addition to avoiding radiation exposure. Venous malformations have a lobulated ‘bunch of grapes’ appearance, are hyperintense on T2-weighted images, moderately intense on T1-weighted images and demonstrate moderate enhancement with contrast.

Newer contrast agents such as Ablavar® (gadofosveset trisodium) are useful in identifying the venous vascular channels within these malformations. Phleboliths are commonly visualized as rounded signal voids. The absence of linear flow voids distinguishes this lesion from arteriovenous malformations. Ultrasound is another useful imaging modality, demonstrating sluggish flow within multiple vascular spaces with heterogeneous hypoechogenic patterns.

Sialolithiasis is an uncommon occurrence in children as compared to adults, while the converse is true for vascular malformations. The symptoms of pediatric sialolithiasis are similar to that of adults, with recurrent postprandial swelling and pain most commonly reported. In children, however, salivary stones are usually smaller (<1cm) and more commonly located within the distal salivary ducts compared to adults, a feature which can be used to increase the index of suspicion for phleboliths.

CONCLUSIONS

Phleboliths are a common feature of venous malformations and may mimic sialolithiasis on plain radiographs and CT scan. A diagnosis of salivary duct obstruction secondary to sialolithiasis is unlikely in the absence of clinical features of post-prandial swelling and pain, particularly in children and sialoliths are overall less common in the pediatric population. Careful history and physical examination with confirmatory MRI can diagnose venous malformations in such cases.

REFERENCES