Sialolithiasis, or stones within the salivary gland ducts or parenchyma, is somewhat common in the adult population—with 12 per 100 000 people affected—and is characterized by symptoms of recurrent glandular swelling, pain, and trismus. Traditional medical management consists of treatment with antibiotics and anti-inflammatory drugs, with hopes of spontaneous expression of the stone through the normal papilla. Surgical management is reserved for stones located in the proximal duct, and glandular resection is reserved for deeply embedded stones.

Originally introduced in the 1990s, sialendoscopy is a minimally invasive diagnostic and treatment modality for inflammatory diseases of the salivary glands. It is an alternative to traditional surgery for salivary gland disease and is associated with decreased morbidity. Sialendoscopy allows both direct visualization of the major salivary gland ductal system, as well as facilitating endoscopic intervention and treatment of chronic sialadenitis and sialolithiasis. It is indicated in cases of intermittent salivary gland swelling of unclear origin, and it allows for differentiation and treatment of stenoses and mucus plugs in addition to sialoliths. Therapeutic indications include treatment of sialolithiasis, dilation of strictures, and treatment of radioiodine-induced sialadenitis.

The only contraindication to sialendoscopy is acute sialadenitis, in that the semirigid endoscope may cause perforation in these cases, and irrigation may propagate infection. Whereas there are added risks of glandular perforation, damage to vasculature and nerve tissue, duct avulsion, and infection, these are not widely reported.

The utility of sialendoscopy is limited by certain sialolith characteristics, namely, size, location, and mobility; anatomical factors such as a narrow papilla; and access challenges such as in patients with microstomia, trismus, or substantial mandibular tori.

Case 1
A man in his 20s presented with a history of 4 weeks of recurrent swelling of the left submandibular gland, predominantly while eating. Intraoral examination demonstrated normal floor of the mouth with normal-appearing overlying mucosa. External palpation demonstrated a soft, mobile, and nontender submandibular gland. A computed tomographic (CT) scan corroborated swelling of the left submandibular gland, but no stones were appreciated. In-office diagnostic ultrasound showed a submandibular gland of normal size, echotexture,
and vascular pattern, lacking evidence of ductal dilation, stenosis, or calculus.

Office sialendoscopy was attempted, but the location of the ductal orifice could not be confirmed with lacrimal probe, wire guides, or other techniques. Despite administration of citric acid, no salivation was noted from the distal aspect of the submandibular duct.

At that point, a surgical exploration of the floor of the mouth was performed in the office. A linear mucosal incision from the region of the papilla to the level of the first molar was made along the expected course of the submandibular duct. Medial and lateral mucosal flaps were elevated. The lingual nerve and branches to the tongue were directly visualized into the posterior floor of the mouth. There was no obvious duct found in the expected region of the submandibular papilla or in the mid-floor of the mouth.

External pressure was then placed on the left submandibular gland, at which point a submucosal papilla became apparent at the posterior floor of the mouth (Figure 1). With additional pressure on the gland, clear saliva was seen emanating from this papilla. A salivary fistula into the posterior floor of the mouth was suspected, and the floor of mouth exploration was terminated.

Because the patient remained symptomatic with submandibular swelling, he was advised that submandibular gland excision through an external approach under general anesthesia was the only feasible option for definitive treatment. The patient went on to undergo a submandibular gland resection in the operating room without complications, with complete resolution of symptoms.

Case 2

A man in his 20s presented with a several-week history of submandibular swelling on the right side. A CT scan was performed, which revealed proximal ductal dilation likely secondary to a complex of ductal stones ranging from 1.8 to 2.6 mm in diameter. In-office diagnostic ultrasound demonstrated that these sialoliths were superficial to the mylohyoid muscle at its posterior border.

In-office sialendoscopy was attempted but failed as a result of inability to cannulate the region of the papilla. At the same time in the office, a mucosal incision was made over the distal aspect of the submandibular duct. The duct could not be identified. The mucosal incision was carried posteriorly into the floor of the mouth in the region of the second premolar. Medial and lateral mucosal flaps were elevated and the floor of the mouth was thoroughly explored, with the lingual nerve and the branches to the ventral aspect of the tongue in view. Again, the Wharton duct could not be identified. A submandibular ductal fistula was also suspected in this case, and the floor of mouth exploration in the office was terminated.

The patient was taken to the operating room for right transoral submandibular sialolithotomy, with possible submandibular gland excision. As the floor of the mouth was examined again, there appeared to be an ostium adjacent to the second molar, which, with external pressure on the submandibular gland, was easily demonstrable and drained clear saliva. This ostium could not be cannulated using lacrimal probes.

The decision was then made to perform an open submandibular gland excision because the stones were not easily palpable. Although transoral sialolithotomy could have been performed, the patient desired definitive therapy and did not want to assume the risks of restenosis if transoral sialolithotomy failed.

The submandibular gland was exposed using standard techniques. After division of the nerves distal to the submandibular ganglion and preservation of the lingual nerve, the duct was sought. Exploration revealed 2 tracts emanating from the hilar region of the gland—one coursing superiorly and posteriorly to the mylohyoid muscle, and the second atrophic one coursing deep to the mylohyoid muscle (Figure 2). The more superiorly coursing tract was followed into the floor of mouth region until the oral cavity was entered just adjacent to the second molar.

The gland was then excised en bloc (Figure 3) and the calculi were noted to be proximal to the takeoff of the more superiorly oriented tract, which extended into the floor of the mouth. The floor of mouth mucosa was then closed with an invertcon Connell suture of 4-0 Vicryl, and a suction drain was placed. The patient had an uneventful recovery.
Discussion

Salivary gland ductal abnormalities have been reported in past literature. In 1994, Gaur et al. described a submandibular gland with 3 ducts and suggested that such an anatomic variation presents a challenge during sialography. In addition, a retrospective study on Wharton duct abnormalities described ductal evagination and suggested that this structural anomaly could be the result of several factors, one of which includes sialoliths inciting focal weakness on ductal walls. Whereas sialooral fistula has been described in the literature as a complication of sialolithiasis, the pathogenesis remains unclear.

Ductal obstruction and inciting damage have been studied in other organ systems. For instance, cholecystocholedochal fistula and the Mirizzi syndrome have been described as manifestations of a process that is initiated by impaction of gallstones in the bile duct. Over time, the gallstones induce pressure necrosis, leading to inflammation and adhesions that facilitate tissue erosion and the ultimate formation of fistula. A fistulous tract can develop in other areas affected by chronic inflammation, necrosis, or ischemia, suggesting that this may be a generalized pathogenesis of fistula formation. For example, Lee et al. reported that an impacted ureteral stone with urosepsis can lead to necrosis of the adjacent ureteral wall, leading to periureteral abscess, which can then perforate into adjacent bowel loops, resulting in ureterocolic fistula secondary to calculous pyohydroureteronephrosis.

There have been other reports that describe salivary glands with multiple ducts. This situation can be seen in several separate conditions. First, pressure-induced necrosis could form a fistula into the posterior floor of the mouth with resultant atrophy of the distal duct. Second, there could be congenital ductal duplication, which would result in multiple ducts. In the third scenario, which is similar to the first, a hypoplastic duct could predispose to forming a stone, which could eventually result in chronic obstruction and fistula formation. In the third scenario, the inciting problem is ductal hypoplasia, whereas in the first, the inciting factor is the stone itself.

Our second patient (in case 2) appeared to have 2 separate ductal tracts, of which 1 was atrophic. In this particular case, the patient may have had pressure-induced necrosis of the duct with a fistula formation into the posterior floor of the mouth. Because there was ultrasonographic evidence of sialolithiasis, chronic ductal obstruction was likely integral to fistula formation. It is uncertain whether the pathophysiologic cause of the fistula was due to a process in the salivary gland analogous to Mirizzi syndrome, involving pressure necrosis and extrusion, vs a similar pathophysiological scenario. It is possible that cases 1 and 2 initially had hypoplastic duct variants, which could potentially impair salivary drainage and ultimately lead to sialolith formation over time. Once formed, the stone would further contribute to continued pressure necrosis and fistula formation. It is our opinion that a process analogous to the Mirizzi syndrome is more than likely responsible for fistula formation. We believe that in both cases, sialoliths caused pressure necrosis of the duct and surrounding tissue, with resulting tissue loss of the embryological or native duct and fistula formation elsewhere.

The patient in case 2 had a clearly identifiable fistula that was posterior to the normal location of the submandibular duct opening in the floor of the mouth. This served as the functional secretory tract for saliva. Interestingly, the patient developed calculi proximal to the fistula takeoff, causing recurrent periprandial submandibular swelling.

In case 1, there was no evidence of sialolithiasis, and ductal obstruction due to sialolithiasis may have contributed to fistula formation at one time. Because no main submandibular ductal remnant was identifiable during in-office surgical exploration, it is possible that in this case, the condition may have been long-standing, and the original duct had atrophied substantially.

Histological examination of a suspected fistula can reinforce the clinical diagnosis of ductal fistula. A true fistula...
would histopathologically reveal stratified nonkeratinizing squamous epithelium, differentiating it from an anomalous duct, which would be expected to have normal duct histological characteristics. Unfortunately, the histopathological description of the ductal samples was not available in our specimens and could not be determined from the permanent tissue blocks.

In cases in which the main submandibular duct cannot be immediately located, it is important to include fistulization as a diagnostic possibility. Surgical exploration of the floor of the mouth may be indicated, and repeated imaging with ultrasound or CT scan may be helpful for guiding treatment. In the event that a sialolith is visible on ultrasound and/or CT and palpable, traditional transoral sialolithotomy may be indicated, even without identification of the duct. In cases in which no calculi can be identified and sialendoscopy is not technically possible, complete excision of the submandibular gland may be appropriate.

Conclusions
Submandibular fistula is a rare complication of chronic submandibular sialolithiasis. The precise etiology and pathogenesis of sialo-oral fistula formation are currently unknown but could be extrapolated from previously described syndromes involving other regions of the body. There is opportunity for advancement of diagnostic theory in this area. When a fistula is present, alternative approaches to diagnosis and treatment are necessary.