Introduction:
Submandibular gland mucoceles are a rare entity. Most salivary gland mucoceles occur in the minor salivary glands of the lip or on the floor of mouth, the latter termed a ranula. These mucoceles rarely involve the major salivary glands. The number of documented cases of submandibular mucoceles remain small, with only about a dozen previous cases of submandibular mucoceles documented in the English literature. Of these, there is only one case where a patient presented with an ascending ranula that extended superiorly through the parapharyngeal space. We report a case of a left sided ascending submandibular mucocele in a pediatric patient who had a previous history of an untreated ranula.

Case Report:
The patient is a 12 year old male with no significant past medical history who was diagnosed with a ranula of floor of mouth one year prior to presentation. At that time, surgical excision of the ranula was recommended. Prior to the surgical procedure, however, the ranula spontaneously improved, his symptoms improved, and the decision was made to cancel the surgery. The patient remained asymptomatic without surgical treatment for the next six months but swelling along his left neck recurred. (Figure 1) CT imaging was obtained and revealed a large, 41.8-mm x 42.3-mm x 74-mm, cystic lesion involving the submandibular gland displacing it inferiorly with the lesion itself extending superiorly through the parapharyngeal space up to the skull base. (Figure 2) Left sided cervical lymphadenopathy at level I, II, and III were also noted. An MRI was deferred as the patient had metal dental braces. He was taken to operating room for excision of the cyst, left sublingual gland, and left submandibular gland. Evaluation of his floor of mouth intraoperatively revealed a site of previous mucosal disruption, presumably the site of his spontaneous drainage. Attempted cannulation of his left Wharton’s duct was unsuccessful, and no salivary flow was noted with pressure applied to the neck. A transcervical approach was performed and exploration of the neck revealed an inflamed cystic structure intimately involved with both sublingual and submandibular glands and extending superiorly toward temporomandibular joint. The left sublingual and submandibular gland were excised along with a transected portion of the pseudocyst. (Figure 3) The pseudocyst was transected superiorly along its tract toward skull base and temporomandibular joint, releasing 20cc of yellow mucoid fluid. The remainder of the cystic structure was irrigated thoroughly and a 7-Jackson Pratt drain was left in place for post-operative drainage. Post operatively, the patient’s symptoms improved and he was able to tolerate a PO diet on post-operative day 1. His drain was removed on post-operative day 2 and he was discharged to home in medically stable condition. The patient returned to outpatient clinic 5 weeks post procedure and continued to do well with no evidence of further mucocele formation.

Discussion:
A ranula is a mucocele of the floor of the mouth. The literature suggests that they may form through blockage of sublingual ducts by infection, trauma, or other cause which leads to obstruction, retrograde pressure of salivary contents and development of a pseudocyst. Simple ranulas remain within the floor of the mouth whereas violation of the mylohyoid muscle with extension into the neck are termed “plunging ranulas”. We believe that the development of this patient’s submandibular mucocele was related to his previously untreated ranula. Surrounding inflammation and infection during his previously untreated ranula may have caused injury and fibrosis in the floor of mouth and such trauma led to obstruction and development of a submandibular mucocele. Sublingual gland drainage occurs through several small individual ducts that number between 2-20 and are collectively called the ducts of Rivinus. However, there are anatomic variations and the sublingual gland may in addition share a common drainage paths with the submandibular gland, termed Bartholin’s duct, which then leads to Wharton’s duct. Because of this intimate association, it is advisable that the sublingual gland be removed in conjunction with the submandibular gland in the setting of a submandibular gland mucocele in order to prevent possible recurrence of a ranula within the floor of mouth. Because mucoceles do not have a true epithelial lining, complete excision of the cyst is unnecessary and removal of the source glands and drainage of the already collected fluid is sufficient treatment. In summary, submandibular gland mucoceles are a rare entity whose formation may involve duct obstruction secondary to injury or infection. Definitive treatment should include excision of the submandibular gland and drainage of the pseudocyst with strong consideration of removing the ipsilateral sublingual gland to prevent recurrences.

References: