

ABSTRACT

Second branchial cleft anomalies (BCAs) are the most common branchial cleft anomaly. They frequently present as a cyst along the anterior border of the sternocleidomastoid muscle with or without a sinus tract. Fistulas are less common and traditionally present with an external opening in the anterior neck over the lower one third of the sternocleidomastoid muscle and travel along the carotid sheath between the external and internal carotid arteries, before terminating at an internal opening in the tonsillar fossa.

We present a case report of a 1 year old female who presented with recurrent drainage from a skin pit over the right angle of the mandible. Magnetic resonance imaging (MRI) with fistulogram revealed a sinus tract that began at the cutaneous pit, communicated with a 1.9 x 2.3 cm cyst in the submandibular space, and suggested extravasation into the oral cavity at the right floor of the mouth.

The patient underwent successful surgical excision of the cyst and sinus tract. No evidence of intraoral connection was found intra-operatively and the area of extravasation noted on the MRI fistulogram appeared to be through the cyst wall into the muscle of the tongue only. Pathology revealed a squamous lined cyst consistent with a branchial cleft remnant. This case report raises awareness to the variability in presentation of second branchial cleft anomalies and highlights the utility of pre-operative MRI with fistulogram.

CASE PRESENTATION

A 1 year-old female was referred to our institution with a three-month history of recurrent drainage and erythema from a skin pit over the right mandibular angle despite multiple courses of antibiotic therapy. Examination revealed a 2 cm mass in the right submandibular space and a sinus tract above the right mandibular angle with clear drainage. No intraoral connection was appreciated. Magnetic Resonance Imaging (MRI) fistulogram demonstrated a tract running from the skin surface toward the submandibular space before opening into a cystic structure measuring 2.3 x 1.9 cm. The tract appeared to continue medial to the cyst with contrast seen extravasating within the oral cavity at the right floor of mouth concerning for complete orocutaneous fistula (Figure 1). A diagnosis of a second BCA was favored on MRI.

The patient was taken to the operating room for excision of the cyst and fistula tract. Methylene blue injection through the skin pit failed to show extravasation into the oral cavity. Complete excision of the cyst and sinus tract was achieved via an inferior neck incision below the inferior aspect of the body of the mandible and a second small elliptical incision around the punctum of the sinus tract (Figure 2A). The tract was dissected inferior and medial to the mandible, leading to the cyst, located superior to the submandibular gland (Figure 2B). The tract continued beyond the cyst, extending medially deep to the floor of mouth and into the substance of the muscle of the right side of the tongue.

Her recovery was uncomplicated and she was seen in follow up 10 days post-operatively, healing well with no evidence of infection. Pathology revealed a squamous lined cyst consistent with a BCA.

CASE PRESENTATION



Figure 1: A) T2 axial and B) coronal MRI showing a 1.9 x 2.3 cm cyst in the submandibular space with extension toward the floor of mouth C) Fistulogram showing a sinus tract beginning at the cutaneous pit, communicating with a cyst in the submandibular space, and suggested extravasation into the oral cavity at the right floor of the mouth.



Figure 2: A) Preoperative surgical marking of the inferior neck incision and second small elliptical incision around the punctum of the sinus tract used to excise the cyst and tract. B) Dissection of the tract from the skin to the submandibular cyst. C) Cyst and tract excised in entirety.

DISCUSSION

Branchial cleft anomalies involving the floor of mouth, are rare, atypical entities with only a few cases reported in the literature.^{1,2} The literature does not clearly define best practice for imaging of these lesions.³ MRI or Computed Tomography (CT) with/without fistulogram are often obtained to help define the relationship between the tract and neighboring structures. At our institution, MRI is favored to avoid radiation exposure in the pediatric population. Due the atypical presentation of the skin pit in relation to the cervical cyst, preoperative MRI fistulogram was obtained in this case. It prompted our team to evaluate for complete fistula via methylene blue injection preoperatively. However the findings on MRI fistulogram did not, in fact, represent a true fistula. Rather the contrast material appears to have ruptured through a weakened portion of the cyst wall into the submucosal tissues of the floor of mouth. Nonetheless, the information provided by an MRI fistulogram can help facilitate complete excision, and thus help reduce risk of recurrence.⁴

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CONCLUSION

Second BCAs can have variable presentations. MRI fistulogram and perioperative methylene blue injection serve as complementary tools to delineate the anatomy of the BCA and its relationship to surrounding structures, thus facilitating complete surgical excision.

REFERENCES

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