



Mucocele of Submandibular Salivary Gland Mimicking Deep Plunging Ranula in Radiological Examination- A Stepwise Surgical Exploration

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Abstract

We report an interesting case of a 45-years-old female patient of Philippines nationality with a left sided neck swelling for 6 months. Our clinical examination showed a swelling in the left upper lateral neck with a normal sublingual area. An ultrasound and MRI of the neck revealed a cystic mass in the left submandibular region appearing inseparable from the left submandibular gland and extending into the floor of mouth on the left side, thus, a possible radiological diagnosis of deep plunging ranula. We performed a stepwise surgical excision of the mucocele and confirmed it to be a case of submandibular gland mucocele only and not a deep plunging ranula despite having a 'tail sign' on MRI scan, which is pathognomonic of a ranula. The dissected specimen and histopathological examination revealed it to be a benign submandibular salivary gland cyst. In addition, preservation of the submandibular gland may be done with only the affected part being carefully dissected out to prevent recurrences.

Keywords: Mucocele; Plunging ranula; Submandibular salivary gland

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Introduction

Primary mucocele of the submandibular salivary gland is a very rare clinical entity but it is nevertheless one of the differential diagnoses of lateral neck masses. Primary mucocele of the submandibular salivary gland may mimic a deep plunging ranula but a ranula is often seen manifesting in the sublingual or oral submucosal area. We aim to present a case of mucocele of the left submandibular gland with a cystic extension into the sublingual space reported both on ultrasound and MRI Neck study although, clinical findings showed an otherwise normal sublingual area. We have, therefore, documented a stepwise surgical excision of the mucocele disproving that a sublingual extension of the cyst and a pathognomonic tail sign on ultrasound and MRI scanning, respectively, need not always be a definitive indication in each and every case of a deep plunging ranula and may include variations as seen in our case.

Case Presentation

A 45-years-old, female Filipino patient with type 2 diabetes was referred to our clinic on September 2017 for her complaints of left sided neck swelling of over 6 months duration. Her neck swelling started as a small nodule in the left lateral upper neck and gradually increased in size. There was no pain or difficulty in swallowing. The neck mass measured approximately 7 cm × 4 cm in size, soft to cystic on palpation, nontender, mobile and occupying the left submandibular triangle. Sublingual space was normal clinically. An ultrasound of the neck performed on 6th September, 2017 showed a cystic mass in the left submandibular region appearing inseparable from the left submandibular gland extending into the floor of mouth on the left side (Figure 1a). The radiologist reported it as a possible plunging ranula. MRI Neck done on 13th September, 2017 revealed a large thin walled cyst measuring approximately 7 cm × 3.2 cm in the left submandibular space. The left submandibular gland was distorted by the cystic lesion and flattened posteriorly. Superiorly, the lesion was seen extending minimally into floor of mouth behind the free margin of mylohyoid muscle (Figure 1b and c). Anteriorly, the lesion was extending partially into the submental space and inferiorly, up to the level of hyoid bone, respectively. Thin internal septations were noted within the cystic lesion. MRI Neck findings were suggestive of a cystic Warthin's tumor.

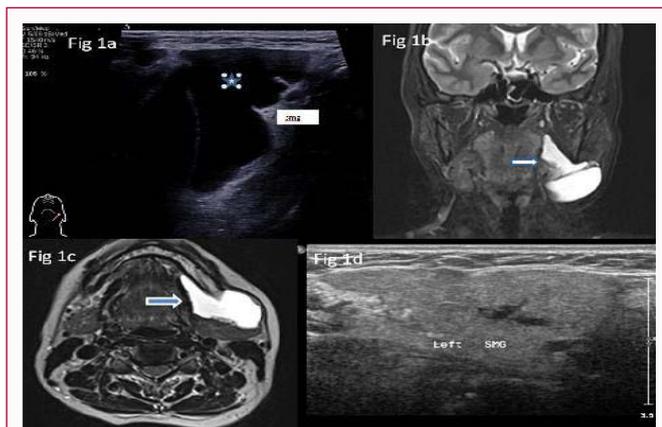


Figure 1 (a): Ultrasound neck shows an anechoic cystic mass (‘) in left submandibular region appearing inseparable from the Submandibular Gland (SMG) and having a deep extension.

Figure 1 (b): Coronal STIR

Figure 1 (c): Axial T2 image shows hyperintense cystic mass in the left submandibular region extending into the floor of mouth (solid arrow) poster lateral to the free margin of mylohyoid muscle and producing a tail sign.

Figure 1 (d): Post-operative follow up ultrasound shows a normal left submandibular gland. No residual cystic mass.

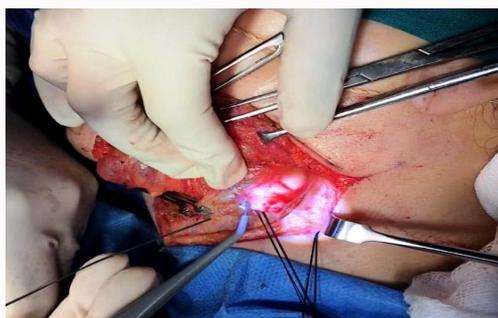


Figure 4: Showing separation of the cystic growth from left submandibular salivary gland.



Figure 5: Reflecting the cyst from the underlying left submandibular salivary gland.



Figure 2: Showing the patient is in position with intact nerve monitor with incision mark.



Figure 3: Showing the cystic swelling after subplatysmal dissection.



Figure 6: Partially preserved left submandibular salivary gland.

ranula. A repeat ultrasound neck done two years after the surgery on 25th March 2019 showed in Figure 1d of a follow up post-operative case of submandibular mucocele excision showed no residual mass or recurrence of the mass lesion.

Discussion

Mucocele of the minor salivary glands are common but mucoceles of the submandibular salivary gland is extremely rare and only 14 cases have been reported up to date in the English literature. About 60% to 70% of mucocele occur in the lower lip, 6% to 16% occur in the floor of the mouth [1]. One review of literature by Tzu Han Chi et al. [2] showed 13 reported cases till 1993 where he included one of their cases of mucocele of submandibular salivary gland in 2017 making the total number of up to 14. Thereafter, no other case reports could be found in English literature. Other factors like trauma and obstruction of salivary gland duct is thought to contribute to the formation of a mucocele.

Mucoceles can be categorized as retention mucoceles or



Figure 7: Resected specimen along with part of the left submandibular salivary gland.



Figure 8: Immediate post-operative neck wound.



Figure 9: Normal healed neck wound after 2 years of surgery.



Figure 10: Normal sublingual area in floor of the mouth.

extravasation mucoceles based on their histopathological appearances. A retention mucocele is a mucous containing cyst where the wall of the mucocele has an epithelial lining. Extravasation mucoceles like deep plunging ranula is also mucus containing cyst but the wall is lined by granulation tissue and without an epithelial lining [3]. Primary mucocele of submandibular salivary gland may mimic a deep plunging ranula. However, a deep plunging ranula develops when there is mucus extravasation extending through or around the mylohyoid muscle deep into the neck with a different histopathology of the cyst wall as in a ranula. In our present case, the mucocele measured 7 cm × 3 cm and cyst wall was found lined by flattened epithelium on histopathological examination.

Clinical symptoms of submandibular gland mucocele includes a painless slow growing mass in the submandibular region usually presenting as a soft well circumscribed mobile mass. Differential diagnosis of such submandibular cystic swellings is deep plunging ranula, cystic hygroma, branchial cysts, thyroglossal duct cysts, dermoid cysts [4,5].

The computed tomography CT or MRI examination or ultrasound neck are important diagnostic tools useful in the diagnosis of such cases for accurate location, extent and to distinguish between a benign or malignant lesion. On CT and MR images, most of plunging ranulas are seen as a well-defined, unilocular, homogeneous, non-enhancing cystic mass with fluid attenuation and signal intensity, located within the Submandibular Space (SMS), often with a contiguous involvement of the ipsilateral Sublingual Space (SLS). The posterior free edge of the mylohyoid muscle is where the communication between the sublingual and submandibular components occurs and typically appearing as a smooth tapered continuation anteriorly into the SLS, the so-called tail sign, which is a specific sign indicating that plunging ranulas originate from the sublingual gland [4-7]. The tail sign helps to differentiate plunging ranulas from other cystic lesions arising in or around the SMS, such as cystic hygroma, thyroglossal duct cyst, second branchial cleft cyst, abscess, and dermoid/epidermoid cyst [4,5]. In addition, fine needle aspiration cytology is useful for the diagnosis, sensitivity rate being about 90% to 100%. But false negative rate for cervical cysts is as high as 50% [8]. Diagnostic dilemma of large primary salivary gland mucocele with radiological extension of the cyst into the sublingual space on ultrasound and on MRI neck as seen in our case gives us the impression of a deep plunging ranula. Clinical evidence of sublingual swelling is prudent for a complete diagnosis. In the present case, we encountered a diagnostic dilemma of a large primary salivary gland mucocele with a radiological finding showing extension of the cyst into the sublingual space on ultrasound and on MRI neck that gave us an impression of a deep plunging ranula. Clinical evidence of a sublingual swelling is prudent for a complete diagnosis. The present case had a similar radiological finding but on table exploration did not reveal any involvement of the sublingual salivary gland.

With a preoperative diagnosis of either a submandibular salivary gland mucocele or a probable deep plunging ranula on the account of ultra sound and MRI findings, we posted our present case on September 20th, 2017. Before an on-table exploration, the patient was connected to a facial nerve monitor. A cervical incision with dissection commencing by a subplatysmal elevation of the cervical flap with the simultaneous preservation of cervical branch of the facial nerve was done. The cyst was very closely attached to the superficial lobe of the submandibular salivary gland and in fact penetrating deep into the submandibular gland parenchyma. The cyst was trapped in the sublingual space but sublingual salivary glands were not involved. The mucocele and part of the superficial submandibular salivary gland was excised completely and the sublingual extension of the mucocele was marsupialized. A stepwise surgical dissection of the mucocele documented here confirmed it to be a case of submandibular gland

mucocoele itself and not a deep plunging ranula despite having a tail sign on MRI scans, which is pathognomonic of a ranula. The dissected specimen and histopathological examination revealed it to be a benign submandibular salivary gland cyst. Our case presented with a unique diagnostic dilemma of a large primary salivary gland mucocoele showing a radiological extension of the cyst into the sublingual space on ultrasound and on MRI neck that gave us an impression of a deep plunging ranula. A clinical evidence of sublingual swelling should also be taken into consideration for a complete diagnosis. The present case had a similar radiological finding but on table exploration did not reveal any involvement of the sublingual salivary gland. A follow up ultrasound done 2 years after the primary surgery on March 24th, 2019 showed no recurrence of the mucocoele with a normal residual submandibular salivary gland.

Various management options of the mucocoele of the submandibular glands include marsupialization, cystectomy, and injection of sclerosing agent, incision and drainage, excision of a mucocoele along with sublingual gland with or without submandibular gland. In a review of literature of total 14 cases of mucocoele of the submandibular gland, 7 cases had undergone excision of mucocoele along with an excision of the submandibular gland and sublingual gland, 6 cases wherein excision of mucocoele with the submandibular gland was done and in one case excision of mucocoele alone was done which developed a recurrence. Surgical excision is the recommended line of management but whether surgical excision of a mucocoele with a combined excision of both the submandibular gland and sublingual gland to prevent recurrence is debatable as seen in our case.

Conclusion

Primary mucocoeles of the submandibular gland though rare must be differentiated from a deep plunging ranula. The importance of clinical examination in spite of radiological diagnosis is prudent in surgical decision making to prepare the surgeon where diagnostic dilemmas exist. Surgical resection of a primary mucocoele of

submandibular salivary gland need not include the resection of the sublingual salivary gland or the total excision of the submandibular salivary gland to prevent recurrences. A meticulous removal of the mucocoele with partial resection of the involved salivary gland, if necessary, will produce the desired result.

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