



A Tale of Two Neck Cysts

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Authors' contributions

This work was carried out in collaboration among all authors. The patient was admitted under Authors AKM. Patient was investigated by author AL under the guidance of author AKM. The idea was conceptualized by author AL. Data and report compilation was done by author AL. Final proof reading was done by author AKM. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

We present a case of 2 neck cysts in the same patient which were clinically diagnosed as plunging ranula and an uncomplicated thyroglossal cyst. This was supported by radiological imaging as well. But it is sought to share that sometimes clinical evaluation and imaging may not be as reliable, as intraoperatively we found it to be a right submandibular epidermoid cyst coexisting with an infected thyroglossal cyst. This was confirmed subsequently on histopathology. Thus we must always consider differentials before embarking on any surgery despite all preoperative investigations, lest we be found at a loss intraoperatively.

Keywords: *Plunging ranula; thyroglossal cyst; epidermoid cyst; neck swellings.*

1. INTRODUCTION

Epidermoid cysts are benign lesions encountered throughout the body, with 7% occurring in the head and neck area and 1.6% within the oral cavity [1]. They are rare lesions derived from germinal epithelium. While a dermoid cyst has an epidermal lining with skin adnexa, such as hair

follicles, sebaceous and sudoriparous glands, the epidermoid cyst contains no such adnexa [2]. It is very rare to have an epidermoid cyst to resemble and present as a plunging ranula, as was in our case.

Plunging ranulas are rare cystic masses in the neck that are mucous retention pseudocysts from

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an obstructed sublingual gland. They “plunge” by extending inferiorly beyond the edge of the mylohyoid muscle, or through dehiscence of the muscle itself, to enter the submandibular space. Surgical excision of the collection and the involved sublingual gland is performed for definitive treatment.

Interestingly, the plunging ranula was coexisting with a thyroglossal cyst. Thyroglossal duct cysts may develop at any place along the migratory path of the thyroglossal tract of the developing thyroid gland. The most common location is around the hyoid bone superior to the thyroid gland.

2. CASE REPORT

A 53-year-old gentleman presented to the Central Hospital, South Eastern Railways, Kolkata with complaints of 2 neck swellings. The first swelling was present in the right submandibular region for the past 6 months which was insidious in onset and gradually progressive. It was not associated with any change in size during chewing, nor associated with pain. The swelling did not cause any problems to the patient as such and was just concerned about the possibility of it being a malignant tumor. Intraoral examination revealed it to be a retention cyst which was bimanually palpable with a classical blue tinge and a diagnosis of a plunging ranula was made.

The second neck swelling was in the midline with typical features of a thyroglossal cyst, not moving on swallowing but moving with tongue protrusion. The patient had the swelling for as long as he could remember but did not seek medical attention as it was not bothering him.

Routine blood investigations were done. All were normal.

A preoperative ultrasound was done of the neck including the submandibular region. It revealed a well-circumscribed 4 X 3 cm homogeneous intraoral sublingual cystic mass which was separate from the tongue, extending towards the submandibular gland but not invading it and without intra-cystic flow, suggestive of a plunging ranula. It also revealed a 2.5 X 2.5 cm fluctuant cystic structure in the midline splaying the strap muscles with posterior acoustic shadowing containing hypoechoic fluid with thin walls without internal vascularity and no septa, separate from the thyroid gland, suggestive of a

thyroglossal cyst. Both the cysts had no interconnection. There was no lymphadenopathy. These findings suggested a right plunging ranula with an uncomplicated thyroglossal cyst.

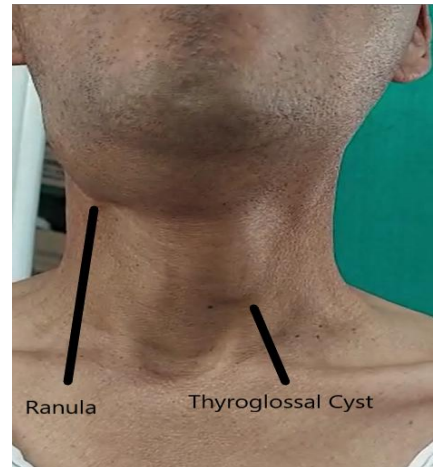


Fig. 1. Preoperative picture showing the neck swellings

A contrast-enhanced MRI scan was done for better delineation of the anatomy. The report gave a solitary cystic 4.4 X 3.5 cm right submandibular mass with low signal on T1 and high signal on T2, with non-enhancing thin walls and no internal septations or debris, with no invasion into surrounding structures, highly suggestive of a ranula. Another 2.7 X 2.5 cm homogenous midline mass was present, low signal in T1 and high signal in T2, with a 7.2 cm tract extending from the mass to the base of the tongue, again, suggestive of an uncomplicated thyroglossal duct cyst. No fat signal was seen in both swellings. The thyroid gland was normal with nothing significant to comment about. No connections were appreciated between the cysts. No significant lymphadenopathy was present.

No preoperative FNAC was done, as clinically and radiologically the diagnosis was straightforward.

The patient was put for surgery. A 4 cm transverse incision was made 4 cm below the mandible on the right side and the ranula was tackled via the cervical approach. During the dissection, unfortunately, the ranula ruptured. To our surprise, the contents were unexpected. There was a copious amount of thick white pus-like fluid, which was cleared and sent for culture. The rest of the cyst wall dissection was standard.

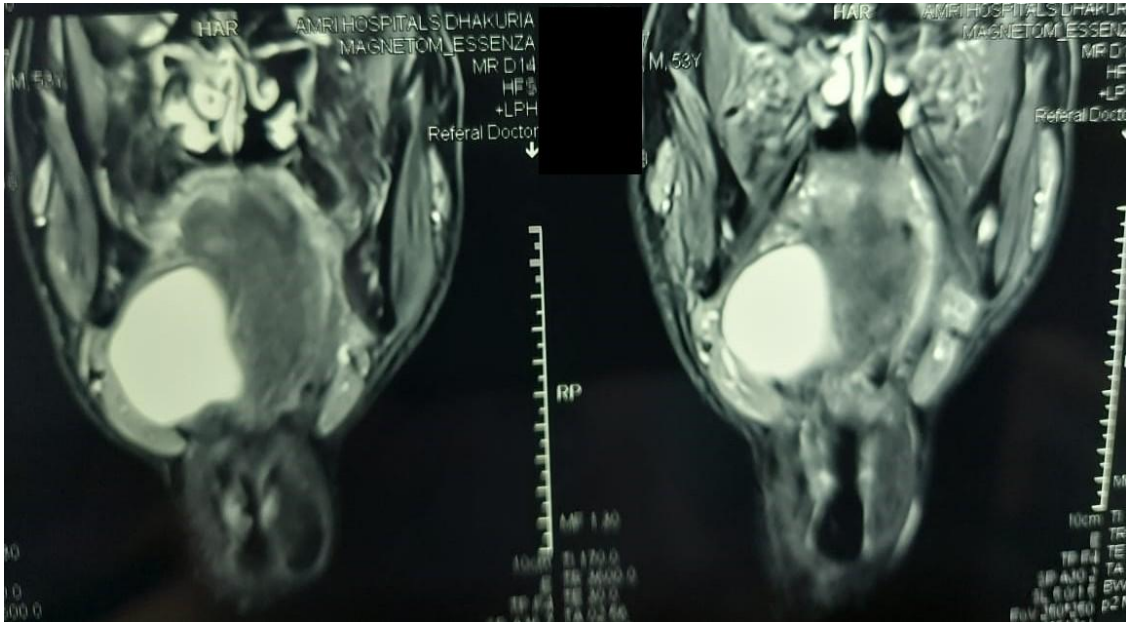


Fig. 2. MRI showing the submandibular swelling (Coronal view)

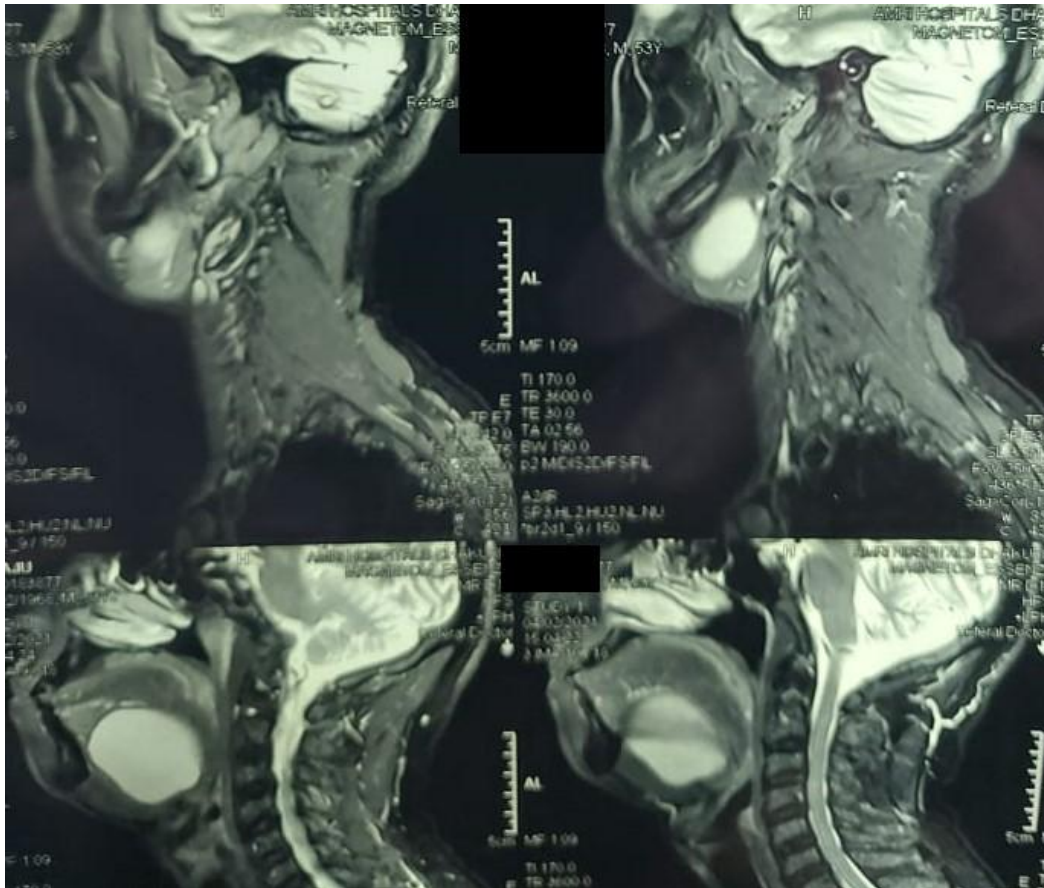


Fig. 3. MRI showing submandibular swelling (Sagittal view)

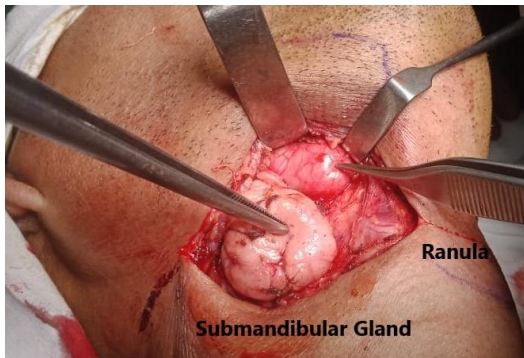


Fig. 4. Intraoperative picture of Ranula

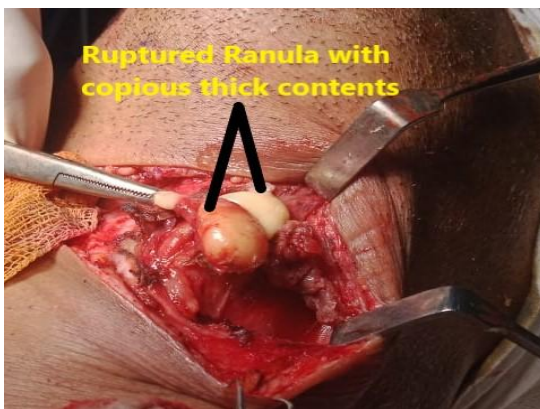


Fig. 5. Intraoperative picture of Ranula post rupture

The thyroglossal cyst was tackled via a standard Sistrunk operation, with a 3 cm midline transverse incision placed over the thyroglossal cyst. While dissection, the cyst also ruptured, and surprisingly, it contained the same thick white copious fluid, presumably pus. It was also drained off and the rest of the tract was dissected and ligated up to the base of the tongue. No connections between the ranula and the thyroglossal cyst were appreciated.

The surgery was otherwise uneventful and the patient had an unremarkable postoperative period.

The presence of the white fluid compelled us to consider epidermoid cysts as the differentials, but we waited for the histopathological reports expectantly.

The histopathological reports suggested that the submandibular swelling was indeed an epidermoid cyst. The midline swelling was confirmed as a thyroglossal cyst with secondary

infection. There were no foci of malignancy anywhere.

This made us very humble and awestruck as both the clinical and imaging findings had led us elsewhere.



Fig. 6. Intraoperative picture of ruptured thyroglossal cyst

3. DISCUSSION

Ranulas are retention collections of extravasated salivary secretions that form unilocular pseudocysts in the submandibular area. Ranulas superior to the mylohyoid muscle appear as a translucent bluish swelling under the tongue, resembling a frog's underside. Ranulas are termed "plunging" or "diving" when they extend beyond the sublingual space and "plunge" inferiorly into the neck. Ranulas occur when salivary duct obstruction results in increased pressure, duct rupture, and formation of a retention pseudocyst.[3].

A thyroglossal duct cyst is the most common midline neck lump, which is formed from the remnant of the thyroglossal tract.[4] Majority of patients present with asymptomatic midline mass usually located at or below the level of the hyoid bone. According to previous studies, most of the patients showed up with this problem with an age of below 30. But interestingly, most recent studies mention several older patients with thyroglossal cyst with their age in the 80s [5]. Often the patient has or recently had an upper respiratory tract infection, but whether the infection leads to cyst formation or simply increases the chance of detection of an already present cyst is uncertain[6].

An epidermoid cyst is a benign lesion that usually affects the scalp region, face, neck, back, and torso. It is commonly asymptomatic, however, it

may become symptomatic due to secondary infection.[7] It occurs in a wide age range, that is, from birth to 72 years, and frequently arises in individuals between 15 and 35 years. 7% of these cysts occur in the head and neck with the oral cavity representing only 1.6%.[8] Surgical enucleation is the recommended treatment.

The differential diagnosis of sublingual lesions in this patient includes ranula, lymphatic malformations, dermoid cyst, and epidermoid cyst. Dermoid cysts are more often seen in the submandibular space than on the floor of the mouth. They are lined by keratinizing squamous epithelium and appear as well-demarcated cysts. They may contain fatty and calcific components as well as fluid. Epidermoid cysts are lined by a simple squamous epithelium and are more frequently seen in the floor of the mouth than in the submandibular space. Rarely, as in our case, they may enlarge and extend to the submandibular space. With imaging, they appear as simple midline cystic lesions. A simple ranula manifests clinically as a swelling in the floor of the mouth and occasionally occurs in the submandibular space by either herniating through a mylohyoid defect or arising from an ectopic sublingual gland. The plunging ranula usually extends posteriorly from the sublingual space into the submandibular space [9].

Surgical excision with complete capsule removal is the treatment of choice for sublingual and submandibular/submental benign cystic lesions. The lesions above the mylohyoid muscle are approached intraorally and lesions below the mylohyoid muscles are removed by extraoral approach[10].

There have been very few cases where a diagnosis of plunging ranula was made preoperatively only to find it as an epidermoid cyst, such as the case of Verma et al [11] and Sadigov et al[12]. It is even rarer for an epidermoid cyst masquerading as a plunging ranula to coexist with an infected thyroglossal cyst. Retrospectively, we conclude that a preoperative FNAC is essential, even though the clinical examination and conventional imaging were clear and straightforward towards the diagnosis. Such a combination of findings makes this case report to be worth publishing.

4. CONCLUSION

This case makes us introspect that despite all the scientific and technological advancements made in the medical field, surprising and unexpected

events can happen at any stage in patient management. It would be rare for an epidermoid cyst to present as a plunging ranula and then co-exist with a thyroglossal duct cyst which got secondarily infected. This makes preoperative FNAC essential in these situations. Another intriguing fact was that the copious white fluid was found in both the swellings, without any inter cystic connections. While the patient returned home satisfied, we were in awe of the biology of the human body which never ceases to amaze us.

CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

Not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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