Unusual presentation of head and neck swellings-a case series

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Abstract

Head and neck is the site of numerous swellings of different etiology and presentations. This article will describe various head and neck swellings with unusual presentation. 4 cases were evaluated i.e. lipoma of the tongue, lipoma as midline neck swelling, infected ranula, epidermal inclusion cyst. All these swellings were presented unusually because of their site, size and inflamed skin over the swelling.

To conclude, few head and neck swellings will show unusual presentation. So, each head and neck swelling case needs systematic evaluation for accurate diagnosis and treatment.

Keywords: Midline Neck Swelling, Lipoma, Epidermal Inclusion Cyst, Ranula, Ludwig's Angina.

Introduction

Head and neck region is a very complex area in the human body with various important structures. Head and neck is also site of numerous swellings of different etiology and presentations. Swellings are classified based on etiology-congenital or acquired, site-midline or lateral and consistency- cystic, firm or hard. We are reporting four cases of Head and neck swellings with unusual presentations in terms of site, size and inflamed skin over the swelling which was challenging during the diagnosis and management.

Case Series

Case 1: A sixteen year old girl came to our OPD with complaints of swelling below the chin since childhood. Swelling was gradually progressive in size and occasionally associated with pain. On examination a 4x5 cm smooth swelling was seen the sub mental region extending to the left submandibular region, firm in consistency, non-tender. Oral cavity examination was normal **(Fig. 1)**. Ultrasound of neck showed a thin walled well encapsulated cystic lesion of size 5.1x2.2cm in the sub mental and left submandibular region with features like thick turbid collection of about 20cc with lesion extending posterior to the strap muscles and hila of left submandibular gland **(Fig. 2).** Fine needle aspiration cytology was suggestive of epidermal inclusion cyst. After investigations, case was taken up for surgery under general anaesthesia.

Horizontal incision placed over the prominent part of the swelling and was dissected and separated. Superiorly it was extending up to the floor of mouth and laterally up to the submandibular gland. Incision was closed in layers **(Fig. 3).** Swelling was greyish white and was about 4.5x3.5x2cm in size **(Fig. 4)**. Histopathology showed fibrocollagenous tissue lined by stratified squamous epithelium and the lumen filled with keratin material, features suggestive of epidermal inclusion cyst **(Fig. 5).**

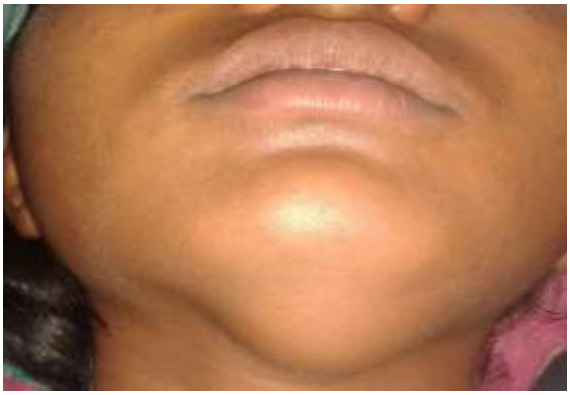

Fig. 1: Submental swelling & left submandibular swelling

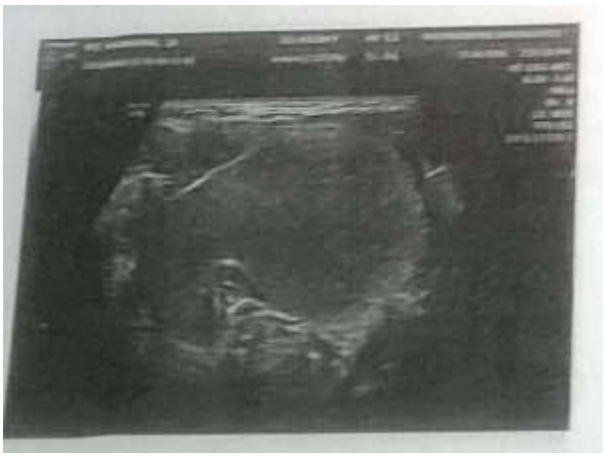

Fig. 2: Ultrasound picture of swelling

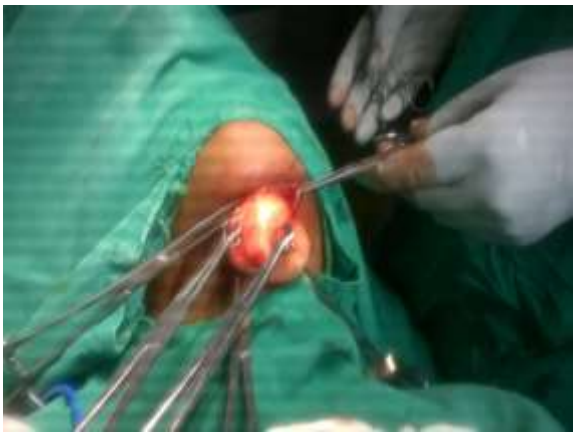

Fig. 3: Intraoperative picture during excision of swelling

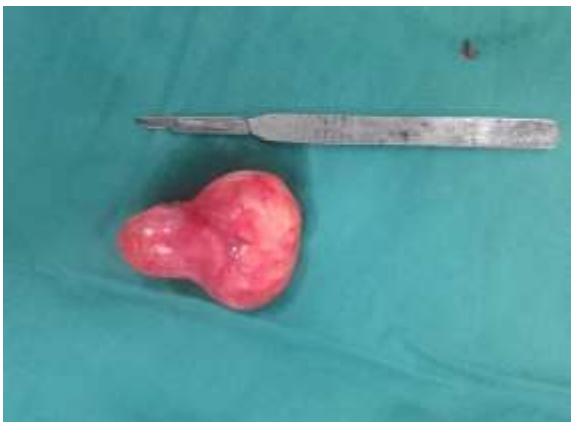

Fig. 4: Excised speciment

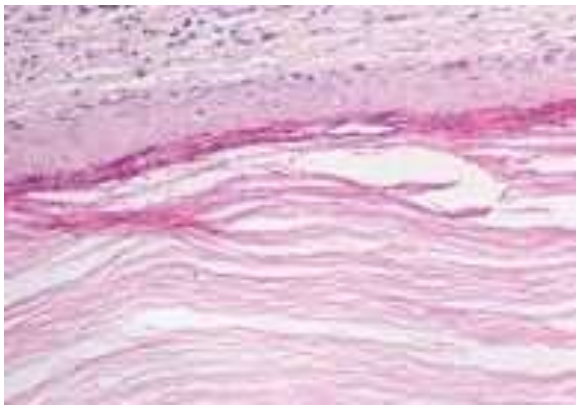

Fig. 5: Histopathology picture of epidermal inclusion cyst

Case 2: A sixty five year old female patient came with complaints of swelling in the tongue which was present since 15 years. It was gradually progressive in size with difficulty in swallowing, mastication and speech. On examination the swelling measured **8x7cm** and was extending from dorsal to ventral surface supero-inferiorly and right to left lateral border of anterior third of tongue. It was yellowish in colour, firm, non-tender with leash of blood vessels on its surface. **(Fig. 6 and 7).** After complete workup, case was taken for surgery under General anaesthesia. Swelling was totally excised and the mucosal layers were closed with absorbable sutures **(Fig. 8).** Gross examination showed a yellowish mass of 8x7 cm with cut surface showing fatty tissue **(Fig. 9).** Histopathology showed sheets of mature adipocytes which was arranged in lobules suggestive of lipoma.

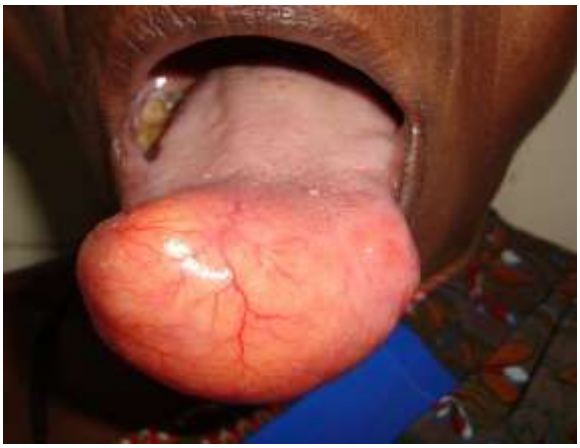

Fig. 6: Front view of swelling

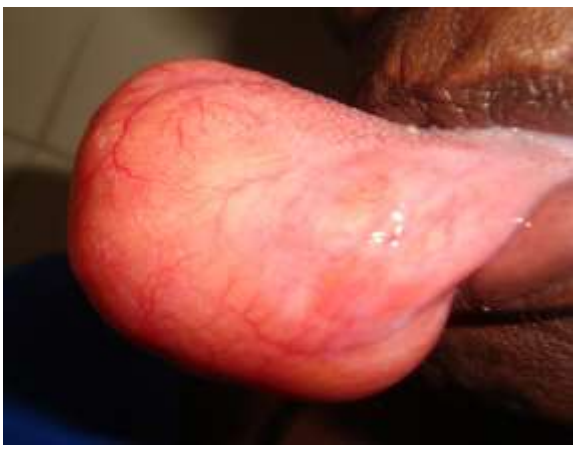

Fig. 7: Lateral view of swelling

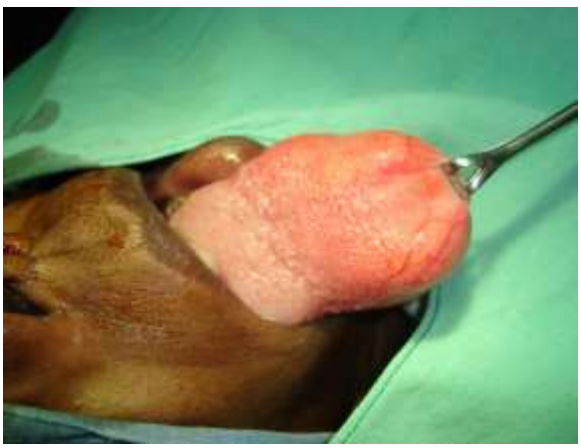

Fig. 8: Intraoperative picture during excision of swelling

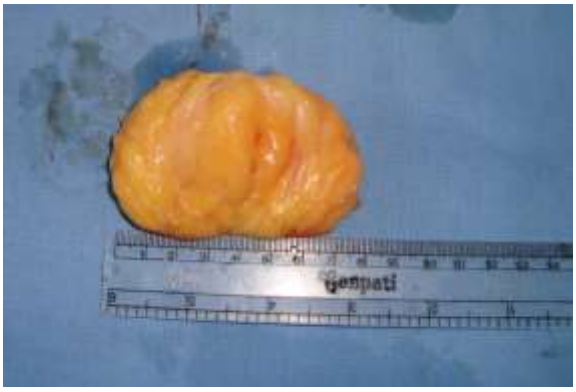

Fig. 9: Excised speciment

Case 3. A seventy year old male patient came with complaints of swelling in the neck since 6 months. Swelling was gradually progressive in size with no complaints of pain, loss of weight or appetite, difficulty in swallowing or speech. He is a known smoker. On examination a 3x3cm swelling was seen in the midline of neck about 2cm above the sternal notch. Swelling was firm in consistency, mobile, non-tender and not moving with deglutition **(Fig. 10).** Ultrasound examination of neck showed features suggestive of lipoma. Swelling was excised under local anaesthesia **(Fig. 11), (Fig. 12).** Histopathology examination showed features suggestive of lipoma **(Fig. 13).**

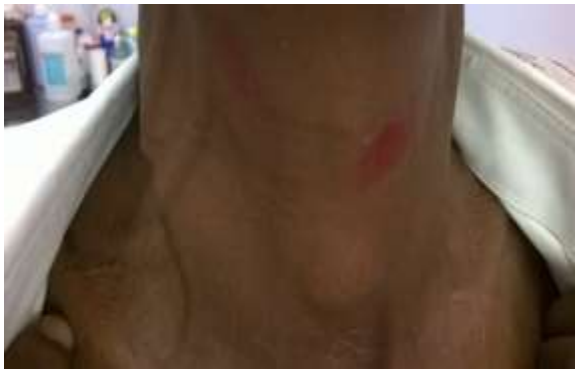

Fig. 10: Front view of swelling in the midline in lower neck

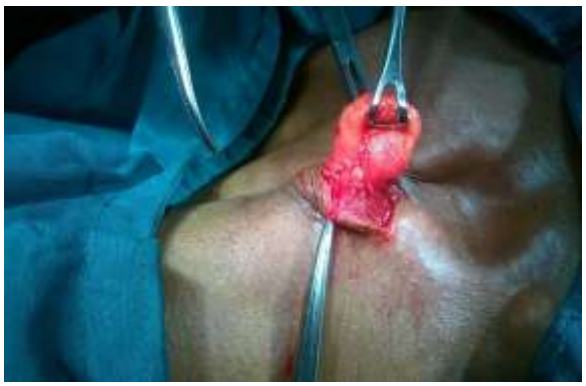

Fig. 11: Intraoperative picture during excision of swelling

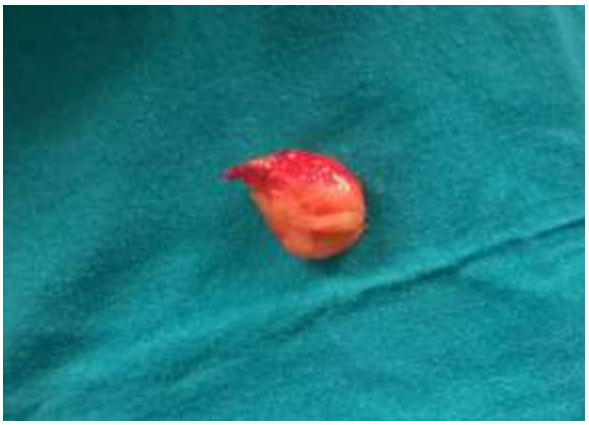

Fig. 12: Excised speciment

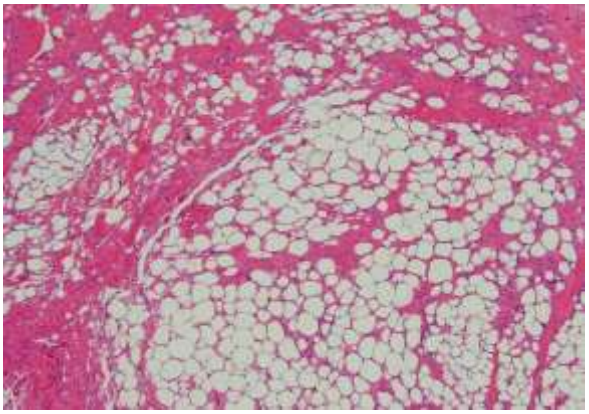

Fig. 13: Histopathology picture of lipoma

Case 4: A forty year old male patient presented with difficulty in swallowing, severe pain and swelling in the neck since four days. There was no significant past medical or surgical history. No history of toothache. On examination, patient had trismus, diffuse edema of floor of mouth with tongue pushed upwards. Diffuse reddish tender swelling noted on the neck mainly in the sub-mental and sub-mandibular region on either side. Bilateral submandibular glands were palpable **(Fig. 14), (Fig. 15).** A possibility of Ludwig's angina was thought and incision and drainage was attempted but there was no pus. Patient was put on intravenous antibiotics and anti-inflammatory drugs. Ultrasound of neck showed a thick walled homogenous swelling in the sub mental region suggestive of ranula **(Fig. 16)**. So, this case of infected ranula mimicked as Ludwig's angina.

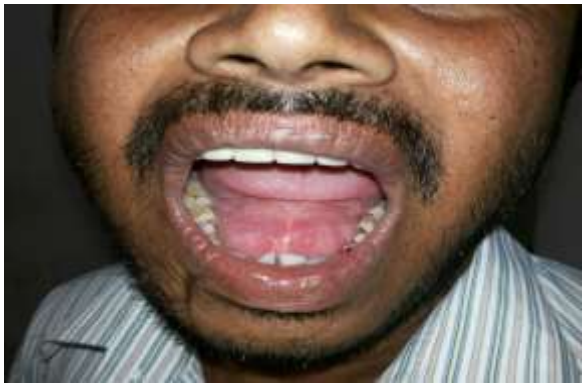

Fig. 14: Intraoral swelling in floor of mouth

Fig. 15: Reddish swelling in the submental region

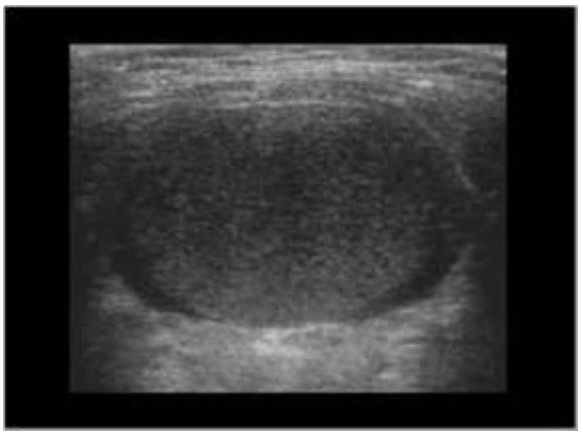

Fig. 16: Ultrasound picture of Ranula

Discussion

Head and neck swellings are commonly seen in outpatient department. Any head & neck swelling needs systematic evaluation. In this case series, we are discussing 4 cases of unusual presentation of head and neck swellings.

Epidermal inclusion cyst is a congenital cyst which occurs due to entrapment of ectoderm at the time of fusion of neural tubes however they can be acquired type also which occurs due to secondary inclusion of epidermal elements into dermis post trauma or iaotrogenically[4]. The incidence rate of epidermal cyst in head and neck region it is 1.6-7.0%. The epidermal cyst constitutes less than 0.01% of all the oral cysts in head and neck region[6].

Epidermal cyst is classified histologically by Meyer in 1955 in three types as epidermoid, dermoid or teratoid[4]. All three types of cyst contain a greasy, cheese like, white/grey/tan material[5]. The epidermal cyst is the slow growing in nature, varies in size from few mm to 10 cm having normal or yellow reddish colour, but are usually small in size. In most cases epidermal cyst is painless swelling with soft consistency. The clinical diagnosis should always be supported with the histological examination[6]. The un-intervened epidermal cyst can achieve increase in size causing discomfort during mastication, swallowing, and speaking[5].

The differential diagnosis of epidermal cyst includes ranula, mucocele, lymphangioma, lymphoepithelial cyst and thyroglossal duct cyst[5]. The choice of imaging in epidermal cyst of head and neck region is ultrasonography due to its reliability and is very economical[1]. On ultrasonography, the epidermoid cysts are seen as well defined, thick walled cysts with echogenic debris within them. Multiple well defined, dependent, echogenic nodules are noted in the cysts[2]. On computed tomography, the epidermoid cysts are found to have low attenuation. The epidermoid cyst of congenital and acquired type has no clinical or histological difference in spite of different pathogenesis mechanisms[1]. Histologically the epidermal cyst is characterized by stratified squamous epithelium with laminas of keratinization on the surface and lumen of the cyst cavity[6]. Treatment of choice is complete surgical resection or enucleation. The recurrence of epidermoid cyst is rare however the sporadic cases of malignant transformation have reported in epidermoid cyst[1].

Rudraksh et al has described a case of 24 year old female presented with a painless mid line swelling in the sub mental region of the neck of 5 to 6 months duration. On examination, a non-tender, firm, mobile and non transluminant swelling of 2 x 3 cms was noted in the mid line in the sub mental region. Ultrasound findings were characteristic of epidermal inclusion cyst[2]. **Case 1** of this series was unusual in terms of (1) size -4.5x5.5x2cm, (2) site-sub mental and submandibular region, (3) Duration-Since childhood and the Ultrasound picture was in favour of ranula as there were no echogenic nodules as seen in epidermal inclusion cyst. But in our case, histopathology report showed as epidermal inclusion cyst.

The majority of tongue tumours are malignant in nature. Lingual lipoma, which accounts for 0.3% of tongue neoplasms, is a benign condition. Similarly, the occurrence in the oral cavity is rare and reported as 2% to 4% of all lipomas. It is typically described as well circumscribed, sub mucosal, with less than 1 cm

swelling, and located on the lateral edge of the anterior two-thirds of the tongue surface. Microscopically, it is composed of mature adipocytes; however, in 20% of cases, it demonstrates histological variants that include spindle cell lipoma, pleomorphic lipoma, angiolipoma, fibrolipoma, myxoid lipoma, and atypical lipoma. Their clinical course is usually asymptomatic until they grow to large sizes[7]. In the present case, the large size interfered with speech and mastication, similarly to a case reported by Gray and Baker[11]. On rare occasions, the infiltration is so extensive that it can cause muscle dysfunction or sensory changes due to pressure on nerve trunks. The average duration of the lipoma before excision is 3.2 years with a range of 6 weeks to 15 years. The usual range in size is 0.5 to 8 centimetres[7].

The differential diagnosis includes well-differentiated liposarcoma, ranula, dermoid cyst, thyroglossal duct cyst, ectopic thyroid tissue, pleomorphic adenoma, and mucoepidemoid carcinoma, angiolipoma, fibrolipoma and malignant lymphoma. The definitive diagnosis is by microscopic examination, which shows adult fat tissue cells embedded in a stroma of connective tissue and surrounded by a fibrous capsule. Lipoma has a characteristic radiographic appearance. On CT scan it shows a high density with well or poorly defined margins depending on the capsule[10].

Surgical excision is the treatment of choice for large lipoma over the tongue. Recurrence is reduced by wide surgical excision while preserving the surrounding structures. Well-encapsulated lipomas, as the present case, easily shell out with no possibility of recurrence or damage to the surrounding structures. It is still advisable to excise them with a little cuff of surrounding normal tissue[8].

In **case 2** of this series, the patient was a 65-year-old female with a slow growing mass present since the last 15years, which measured 8 cm in diameter. The mass was painless but she had difficulties in swallowing and tongue movement was impaired. however, taste and somatic sensation were intact. Chunkitchung reported a 62-year-old man with a 6cm mass in his tongue that was slow growing for 2 years. He had difficulties swallowing large food items. Moreover, his speech was not very clear due to the bulkiness of the mass[8]. Colella reported a 75-year-old man with a 10 cm mass in his tongue from 30 years ago. His speech was not very clear due to the bulkiness of the mass and he had difficulties in swallowing[9].

Lipoma is seen in all age group though mostly seen in fifth and sixth decade[13]. It constitutes five per cent of all benign tumours of body and can be found anywhere in the body. Lipoma in head and neck region is not commonly encountered (13%). Amongst the head and neck lipomas, commonest location is posterior neck. Anterior neck is a rare location for head and neck lipoma[12].

Lipomas are slow growing, painless, mobile, non-fluctuant, soft masses & are generally well encapsulated. Lipomas can be singular or multiple & are typically asymptomatic unless they compress neurovascular structures. Beside frequent aesthetic consequences, lipomas can also exert pressure on surrounding tissues and structures. Giant lipomas are defined by Sanchez et al as lesions with size of at least 10 cm in one dimension or weighing a minimum of 1,000 gm[16].

Although the diagnosis is mostly clinical, imaging tools are useful to confirm the adipose nature of the lesion and to define its anatomic border & exclude possible communication with the spinal canal. The characteristic sonographic appearance of head and neck lipomas is that of an elliptical mass parallel to the skin surface that is mostly hyper echoic relative to adjacent muscle and that contains linear echogenic lines at right angles to the ultrasound beam[16]. Computed tomography is modality of choice to confirm lipoma. Lipomas appear as homogenous low density areas with a CT value of -50 to -150 HU with no contrast enhancement. A thin soft tissue capsule may be seen surrounding a subcutaneous lipoma, distinguishing them from surrounding fat. Surgical excision of lipoma is the definitive treatment. Surgery is reserved for patients coming for cosmesis (most common indication) and pressure effects & to rule out malignancy. Differential diagnosis of lipoma in neck can be branchial cysts, epidermal cysts, thyroglossal cysts, haemangioma, lymph node and normal muscle[12].

Smrity et al has reported a case of giant lipoma in the anterior midline with mediastinal extension.[12] In **case 3** of this series, we had an elderly male, chronic smoker with anterior midline neck swelling. Our first differential diagnosis was lymph node but the histopathology report suggestive of lipoma.

Ranula is a sublingual gland mucocele; it's a mucous extravasation cyst, which occurs as a result of trauma or obstruction of the sublingual or minor salivary gland or the duct itself. The term ranula is derived from the Latin word "Rana" which means "belly of a frog", and classic ranula presents as a translucent swelling bluish appearance in the floor of the mouth, resembling the bulging underbelly of a frog[19].

Ranula can be classified in 2 groups, simple (intraoral) and plunging (deep/diving/cervical). Simple ranula is a true cyst, located above the mylohyoid muscle, with a lining formed by the sublingual gland capsule; plunging ranula is a pseudo cyst partially contained by the remaining epithelium and inflammatory cells and it occurs when it becomes large, ruptures out of the posterior sublingual salivary gland into submandibular space below the level of the mylohyoid muscle. Ranula is an uncommon pathology in the oral cavity with a prevalence of 0.2 cases per 1000 persons. Presentation is most frequently in the

second and third decades of life, with an age range of 3–61 years.

The most frequent clinical presentation is painless swelling of the sublingual space or submandibular space; when it enlarges, may produce deviation of the tongue, interfering with speech, mastication, respiration, and swallowing. When it is an isolated, small and oral lesion the diagnosis is generally easy on clinical examination. Clinical diagnosis could be difficult when ranula presents as large cervical mass, because it can mimic other soft tissue masses of the neck (dermoid, branchial cysts, lipomas, lymphangioma)[17].

These entities cannot be distinguished by clinical evaluation alone, so diagnosis requires imaging and fluid aspiration from the cervical swelling. Ultrasound is a non-invasive, cheap, reliable examination commonly used to investigate a neck mass in young people as it enables differentiation between cystic and solid masses and identifies anatomic location. It could be useful to identify a possible relationship with hyoid bone. Ranula usually appears like hypo-echoic well defined mass. If ultrasound findings are unusual or inconclusive additional diagnostic imaging is required.[18]

There are different methods of treatment of ranula. For simple ranula the easiest and least invasive treatment is marsupialization with drainage naturally into the floor of the mouth. This treatment is associated with high recurrence rates, approximately 60% to 90%[17]. Ludwig's angina is a rapidly progressing poly-microbial cellulitis of the sub-lingual, sub-mental and sub-mandibular spaces that can result in life-threatening airway compromise. Patients with Ludwig's angina typically have a history of recent dental extraction or of poor oral hygiene and dental pain. Symptoms include swelling, pain in the floor of the mouth and anterior neck, fever, dysphagia, odynophagia, drooling, trismus, toothache, and fetid breath. Hoarseness, stridor, respiratory distress, decreased air movement, cyanosis are all signs of impending airway catastrophe.

On oral examination, elevation of the tongue, woody, brawny induration of the floor of the mouth and anterior neck, and non-fluctuant supra-hyoid swelling typify the disease process. There is typically a bilateral submandibular edema, with marked tenderness on palpation and occasionally subcutaneous emphysema[20].

Ghani et al has described a case of ranula with secondary infection presenting with difficulty in swallowing.[18]

In **case 4** of this series, we had a male patient with an inflammatory swelling in the submental and submandibular region in addition to a diffuse swelling in the floor of mouth with difficulty in swallowing. All the findings were suggestive of Ludwig' angina. But ultrasonography showed a thin walled well encapsulated cystic lesion measuring about 4x5cm suggestive of ranula. Thus a case of infected ranula presented as Ludwig's angina.

Conclusion

Head and neck swellings are commonly encountered in ENT practice. Some of them may have unusual presentations. Clinicians should keep in mind all the possible differential diagnosis while managing these cases.

Conflict of interest: None

Source of support: Nil

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