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Recurrent extensive plunging ranula: A rare case

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Abstract:

Plunging ranula (PR) is a cystic mass presenting on the lateral side of the neck with or without a history of the usual presentation of an intraoral lesion. "PR" is recognized as an uncommon lesion that has been found predominantly in the third decade of life mostly in males. However, the exact prevalence of "PR" is not yet known. We report a rare case of an extensive PR that reached the parapharyngeal space in a 17-year-old Saudi female. The diagnosis was made when she presented to the ENT clinic with swellings in the right submandibular and right floor of the mouth. The diagnosis of "PR" with the absence of an oral lesion is very challenging and requires a detailed history, clinical examination, and radiological imaging. Different modalities of treatment have been discussed. However, the excision of ranula and the sublingual gland is the most effective way of management.

Keywords:

Case report, extensive, plunging ranula, recurrent

Introduction

Plunging ranula (PR) is a spread of pseudocyst "Ranula"[1] to the submental or submandibular area.[2] Patients with "PR" present with lateral neck swelling in the submandibular triangle.[3] Most PRs are translucent with visible components, whereas all simple ranulas have them.[1]

The reported prevalence of "PR" showed a male predominance with a ratio of 1:0.74[1] as well as a predominance in the third decade of life.[4,5] Even though the prevalence of "PR" is believed to be lower than the simple type, the exact number has not yet been identified.[5]

Case Report

A 17-year-old Saudi female presented to an ENT outpatient clinic for an evaluation of a right submandibular swelling and a swelling of the right floor of the mouth.

History from the patient revealed that she had had recurrent swelling on the right side of the floor of the mouth in the last 6 months which had continuously changed in size. There was no history of dysphagia, odynophagia, fever, night sweating, weight loss, or respiratory distress symptoms.

On examination, a right cystic neck swelling, with a size of 5 cm × 4 cm × 7 cm, was found. The oral component showed thin translucence with slight turbid cystic fluid.

Computed tomography (CT) scan showed right extensive cystic lesion reaching parapharyngeal space, right floor of the mouth, right submandibular, and right sublingual spaces. The cystic lesion was anterior to the sternocleidomastoid muscle with no evidence of septation or enchantment [Figures 1 and 2].

The swelling was treated with repeated evacuations in outside hospitals, after which she underwent right cystic and submandibular gland excision. However,

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2 months after the surgery, the cystic lesion recurred in the right sublingual, submandibular space, and the right floor of the mouth with a great deal of serous yellowish discharge. CT scan showed right submandibular cystic lesion extending to the floor of the mouth with no evidence of septation or enhancement to suggest an infected cyst.

The patient then underwent an excision of this cystic lesion together with the right sublingual gland by the cervical and floor of the mouth approach. Since march 2017 there was a complete resolution thereafter with no recurrence [Figures 3 and 4]. Written consent was obtained from the patient for this case to be published.

Discussion

A ranula is a mucus retention cyst that is formed in the sublingual space when one of the small ducts of the sublingual gland is obstructed. With time, further secretions accumulate in the sublingual space to give rise to an extension along that space anteriorly and posteriorly. This occurs if the posterior extension extends beyond mylohyoid muscle.[6] A ranula extension to the submental or submandibular area is called PR.[2]

A swelling ranging from 4 cm to 10 cm in size on the lateral side of the neck, precisely in the submandibular triangle, is the classic presentation of PR. The swelling is painless and fluctuant and progressively enlarges in size but does not usually change with swallowing or eating.[3] However, in advanced cases of PR, patients may present with airway obstruction or dysphagia.[4] In addition, it can spread to parapharyngeal space as far as the base of the skull superiorly, to the supraclavicular area inferiorly, to the retropharyngeal space posteriorly, or it may spread anteriorly to cross the midline.[3]

PR is diagnosed by means of a comprehensive history, clinical presentation, and imaging studies. Ultrasonographic studies show a cystic lesion (ovoid or lobulated) in the submandibular triangle with a mylohyoid defect observed in up to 90% of the cases. CT shows a unilocular cystic lesion with thin or imperceptible walls and homogeneous intracystic low attenuation similar to other simple fluid-containing structures. Magnetic resonance imaging shows a thin-walled cystic structure with low-to-intermediate

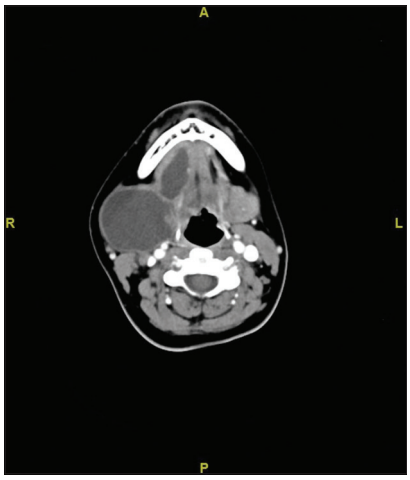

Figure 1: Computed tomography scan axial view showed right submandibular cystic lesion extended to the floor of the mouth with no evidence of septation or enchantment

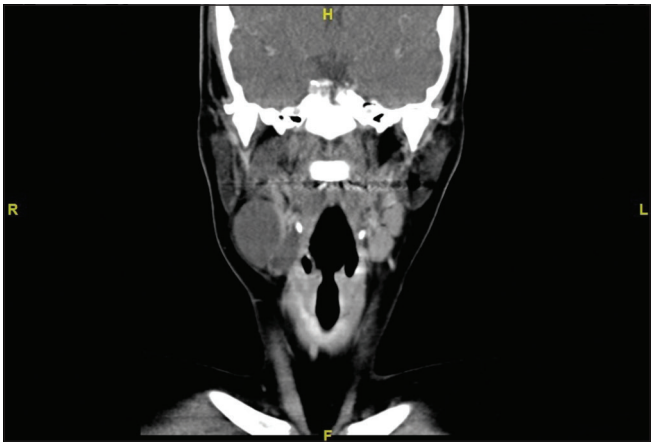

Figure 2: Computed tomography scan coronal view showed the extension of plunging ranula to the right parapharyngeal space

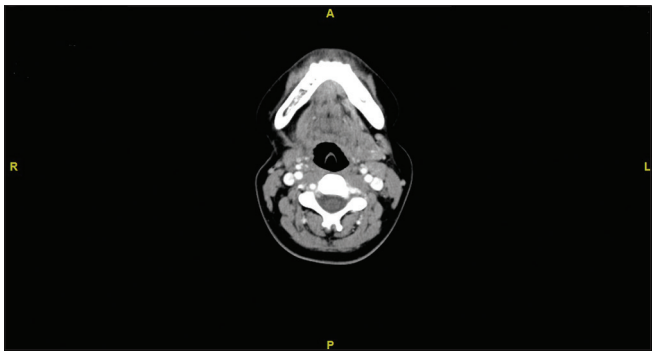

Figure 3: Computed tomography scan axial view showed a complete resolution after the last operation with no recurrence till date

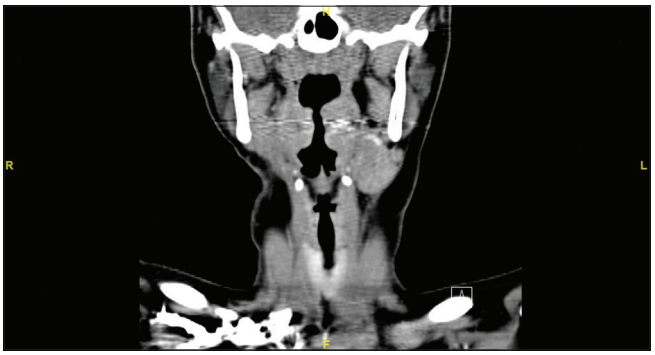

Figure 4: Computed tomography scan coronal view showed a complete resolution after the last operation with no recurrence till date

signal on T1-weighted sequences and high signal on T2-weighted images. In addition to the imaging studies, the use of fine-needle aspiration cytology would show yellow aspirate, positive amylase, and mucin. There are no epithelial/glandular elements, cholesterol crystals, and keratin in this condition.[3]

Clinical diagnosis of ranula may be difficult in the absence of oral swelling; thus, PR should be used in differential diagnoses in many cases. These cases include thyroglossal duct cyst, intramuscular hemangioma, lipomas, cystic/neoplastic thyroid disease, branchial cyst, submandibular sialadenitis, laryngocele, dermoid cyst, lymphatic or vascular malformations, and infectious cervical lymphadenopathy such as tuberculosis, Epstein–Barr virus, cervical thymic cysts, dermoid cysts, cystic hygroma, and benign teratoma.[3,7] Therefore, imaging studies or fine-needle aspiration may be required.[8]

Since the spontaneous resolution of PR is rare, intervention is necessary.[8] Several approaches are used in the treatment of PR with different rates of success, including sclerotherapy, marsupialization, ranula excision, sublingual gland excision, or sublingual gland with ranula excision.[3,9] In addition, cryotherapy after marsupialization has been proposed.[10] The most appropriate treatment procedure for children remains uncertain.[8]

Treatment with sclerotherapy can be considered as the primary treatment modality in the pediatric age group. The efficacy of sclerotherapy depends on many factors including a number of injections, interval between injections, and the initial size of the lesion. Moreover, this modality may be complicated by transient fever and painful cyst as a result of the inflammatory reaction.[3]

On the other hand, ranula excision alone has a high recurrence rate of up to 36.7%. Consequently, it is best to avoid this modality,[3] which may also have numerous complications with such high risks as postoperative infections with 20% risk and 40% risk of tongue hyperesthesia.[9]

Excision of ranula and the sublingual gland which is the source of both intraoral and PR has 0% recurrence rate. This means that this modality of treatment is the most effective technique.[3]

Conclusion

The diagnosis of PR is very challenging in the absence of an oral lesion. As a result, a comprehensive history, clinical presentation, and radiological findings are the main diagnostic tools. Several modalities of treatment can be used with different recurrence rates. However, sublingual gland and ranula excision are considered the main modality of treatment as it has the lowest recurrence rate and low postoperative complications.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understand that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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