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Case Report

Arteriovenous Malformation of the Floor of the Mouth Misdiagnosed as a Ranula and Complicated by Ludwig's Angina in Three Years Old Child-Case Report

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Abstract

Introduction: AVMs are abnormal congenital connections between arteries and veins bypassing the normal capillary bed, present at birth, although they may not become evident until adolescence or early adulthood, and they persist throughout life.

Case Presentation: A 3-year-old female patient presented with neck swelling, raised tongue with pus discharge from swollen floor of the mouth accompanied by foul smell. It was diagnosed as a ranula and advised by another doctor to be put on follow up until the child got older., incision and drainage of the neck swelling was done by three incisions (midline, right and left), Another incision at the floor of the mouth was done and communicated with extraoral incisions, drains put in place. The lesion at the floor of the mouth was excised. Biopsy sent to histopathology examination where the result revealed an arteriovenous malformation.

Conclusion: AVMs should always be considered in the diagnosis of sublingual swellings. Careful examination and diagnosis are critical to prevent fatal complications.

Keywords: *Arteriovenous Malformation; Ludwig's Angina; Infection; Floor of the Mouth Abbreviation AVM; Arteriovenous Malformation*

Introduction

Ludwig's angina as first described by Wilhelm Fredrick von Ludwig in 1836 is a rapidly progressive gangrenous cellulitis [1]. It is considered an infection mostly affecting adults with poor dentition. Current studies, however, show that 27% to 30% of Ludwig's angina cases occur in the pediatric population [2,3]. This infection remains a challenging threat to the pediatric patient population despite the availability of intravenous antibiotic therapy use in combination with airway securement, which has evidently reduced disease-specific mortality [4], As many as 1 in 3 to 4 cases of Ludwig's angina are reported to occur in children [2,3] and death from this infection in the pediatric population still approaches 10% to 17% [3,5]. Early recognition and treatment of this disease course is paramount to achieve a fortunate outcome.

Swelling in the floor of the mouth often represents a salivary glands disease. Rarely, such a swelling may be associated with an inflammation in a salivary gland. Such swellings are thought to arise from tissue normally located in the region, so the source may be, vascular, neurogenic, dentigerous, or skeletal. This case reported a three years old patient with a Ludwig's angina as a consequence of a swelling in the floor of the mouth which was clinically diagnosed as a ranula but the histopathological report revealed an arteriovenous malformation.

Case Report

A 3-year-old female patient was presented to emergency department complaining of neck swelling, raised tongue with pus discharge from swollen floor of the mouth accompanied by foul smell. The parents mentioned that there was a swelling at the floor of the mouth for one-year duration. It was diagnosed as a ranula and advised by another doctor to be put on follow up until the child got older. On examination she was febrile, dehydrated and fatigue. The floor of the mouth was swollen with greyish overlying mucosa. Pus was discharging from an opening at the right side of the floor of the mouth. The tongue was raised and obstructing the airway partially. On neck examination, there was a submental swelling extending

bilaterally to submandibular regions which was tender non-fluctuant on palpation (Figure 1).

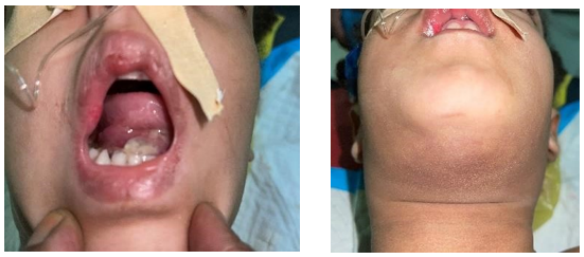

Figure 1: *Shows the sublingual swelling with discharging pus to the floor of the mouth, Submental and bilateral submandibular swelling.*

She was immediately admitted where antibiotic, analgesics and fluids started. Investigations revealed a high total white blood cell. CT scan revealed a diffuse edema within and between the affected submandibular, sublingual and submental spaces with cystic lesion within the floor of the mouth area. Patient was taken to operating room, incision and drainage of the neck swelling was done by three incisions (midline, right and left) (Figure 2).

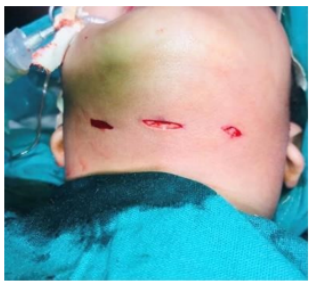

Figure 2: *Shows the incisions for the Ludwig's angina drainage.*

Another incision at the floor of the mouth was done and communicated with extraoral incisions. Vigorous irrigation by normal saline was done and drains put in place. The lesion at the floor of the mouth was excised (Figure 3) and biopsy sent to histopathology examination where the result revealed an arteriovenous malformation. Patient was admitted for three days and discharged on good condition.

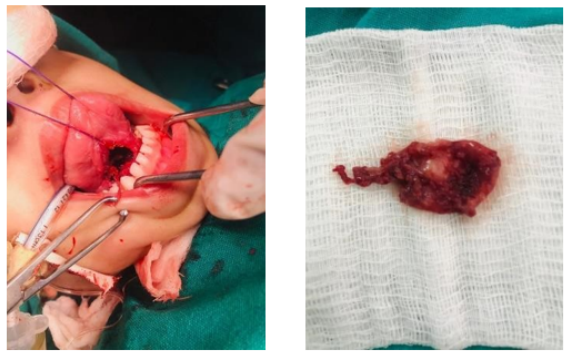

Figure 3: *Shows intraoperative image of the floor of the mouth and the excised AVM.*

Discussion

An arteriovenous malformation (AVM) is formed from abnormal communications between arteries and veins without the normal intervening capillary bed [6]. Although they are often found in the head and neck [7,8], they are rare in the oral cavity. In the current case, the occurrence of an arteriovenous malformation was not suggested by the preoperative assessment or the intraoperative behavior. An infected ranula was the first in mind since the typical history of recurrent swelling that increase and sometimes discharge some fluids as mentioned by the parents. Infected ranula is rarely happening although an infected plunging ranula was reported by Schiarelli C., *et al.* in an 8 years [9].

A sparse reported cases of AVM in the floor of the mouth was found in the literature [10,11]. The most common complication associated with arteriovenous malformation is bleeding. David O., *et al.* reported a sublingual arteriovenous malformation which was ruptured causing an emergent airway obstruction in 82 years old female. In the current case, the patient presented with a submandibular, submental and floor of the mouth swelling with fever and fatigue; atypical presentation of ludwig's angina in a pre-existing arteriovenoius malformation. Gharaibeh., *et al.* reported a similar case of Plunging arteriovenous malformation in the floor of the mouth with a description of a dermoid cyst [12] but it was not complicated by ludwig's angina.

AVMs can be grouped into; [1] congenital: occurring as a consequence of lack of differentiation of arteries, capillaries, and veins during vascular development, [2] acquired: linked to a previous history of injury or trauma [13]. Our case presented in a young age complaining of a sublingual swelling, incision and drainage was done as claimed by the parents, this may be caused a chronic recurrent infection that progressed into Ludwig's angina obstructing the airway. In the current case, we used an intraoral sublingual approach, there was negligible bleeding during excision and a lot of pus discharge. We hypothesize that chronic infection caused embolization of vessels and prevented fatal bleeding. Some authors suggested a median mandibulotomy approach for a bigger size AVMs for good convenience of the lesion [14].

In children, 1 in 3 to 4 cases of Ludwig's angina are reported to occur [3] with mortality rate of 10% to 17%]. A lot of possible causes of ludwig's angina had been mentioned in the literature, but concerning AVMs as a cause, literature is sparse. The only chance for cure of AVM is preoperative embolization and complete resection but with a possibility of recurrence and progression [12,15]. Vascular malformations may present in association with underlying disease or systemic anomalies including: Bonnet-Dechaume-Blanc syndrome or Wyburn-Mason syndrome, Parkes-Weber syndrome and Cobb syndrome [16], current patient was otherwise a normal child and no other systemic abnormalities were detected.

Conclusion

A wide variety of lesions can involve the floor of the mouth. AVM of the floor of the mouth is an unusual entity, AVM with static or low flow could always be theorized. Careful examination and diagnosis of the swellings at the floor of the mouth are critical to prevent fatal complications.

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