

Case Report

The Marvel of Surgery - Infantile Parotid Hemangioma

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ABSTRACT

Parotid hemangiomas represent 0.4-0.6% of the tumors of the parotid gland and are more commonly seen in the pediatric population compared to adults¹. The usual age of presentation is 3-4 months². Salivary gland tumors are rare in children, with an incidence of less than 5%. However, despite being rare tumors, hemangiomas are the most common benign tumor of the salivary glands in children². Treating parotid hemangiomas can be challenging due to their profuse growth, which in turn causes significant disfigurement and vascular shunting leading to congestive cardiac failure. There has been no sure treatment reported for large, deforming hemangiomas³. Surgery is challenging in Parotid Hemangiomas due to their resistance to medical therapy and proximity to the facial nerve². We report a 3-month-old, full-term female baby with a presentation of painless swelling in the parotid region for one month. She was planned for surgery and successful parotidectomy was done preserving the nearby structures.

KEYWORDS: Infantile Parotid hemangioma, Salivary gland tumor

INTRODUCTION

Hemangiomas are caused by the excessive proliferation of endothelial cells. Some may be present at birth or may develop a few months later. Hemangiomas can be seen superficially on the skin surface as a reddish or bluish macule or may involve underlying mucosa and organs. Several theories about the etiology of hemangiomas include hypoxia as an activator of GLUT1 (Glucose transporter 1) and VEGF (Vascular Endothelial Growth Factor), giving rise to vasculogenesis from progenitor cells⁴.

Hemangiomas of the parotid gland are quite common in the pediatric population, are benign and mostly asymptomatic. They have an association with premature birth, low birth weight, prenatal hypoxia, older maternal age, chorionic villus sampling, and twin pregnancy⁴. The growth pattern usually involves proliferative, plateau, and involution phases. These tumors can have serious complications like difficulty feeding, cosmetic disfigurement, airway compromise and congestive heart failure⁷. There are no fixed guidelines for treatment, however, the use of propranolol, corticosteroids, and interferon alpha has

shown some benefit⁷. Surgery is also an option, although this is not very common due to the risks of damage to nearby structures.

CASE REPORT

A 3-month-old female baby born full term, delivered via cesarean section, cried immediately after birth, and did not require NICU admission, presented with a swelling over the right side of the face that was first noticed at the age of two months and gradually increased in size. It was a painless swelling with no history of trauma, xerostomia, xerophthalmia, or facial weakness and was not associated with an increase in size during feeding. Clinical examination revealed a right-sided 2x3cm globular mobile swelling in the extent of the parotid gland over the right cheek with a smooth surface and margins (Figures 1a & 1b). The color of the swelling was similar to the surrounding skin, and there was no erythema or ulceration over it. The swelling was soft in consistency, non-tender, and non-pulsatile, with no sinus formation or bruit. There was no local rise in temperature, and

no other swellings were seen during the examination on both the ipsilateral and contralateral sides.

A contrast-enhanced Magnetic Resonance Imaging (CE-MRI) of the face showed a lobulated well-defined mass lesion measuring approximately 27mm (AP) X 21mm(TR) X 27mm(SI). The lesion was isointense on T1, hyperintense on T2 & STIR, and was seen involving the superficial lobe of the right parotid gland, showing homogenous enhancement with internal flow voids (vessels). These findings were suggestive of a benign lesion of the right parotid gland, possibly an infantile hemangioma. A diagnosis of infantile hemangioma of the right parotid gland was made based on the clinical examination and CE-MRI. All routine investigations were done, and the baby was planned for a right superficial parotidectomy under general anesthesia. A lazy S or modified incision was given (Figure 2), platysmal flaps were raised, and dissection proceeded.

The greater auricular nerve was identified, divided, and ligated (Figure 3).



Figure 1(a)



Figure 1(b)



Figure 2

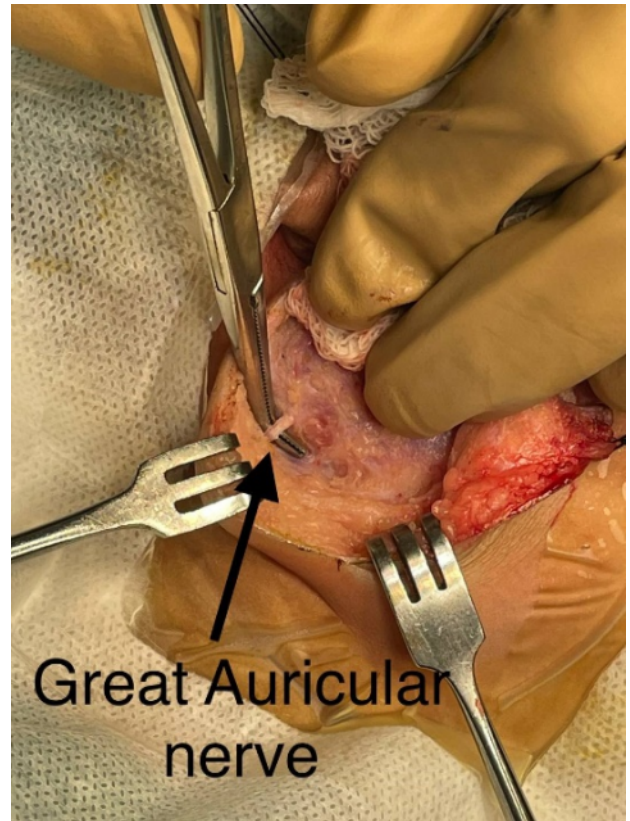


Figure 3

Then the hemangioma (Figure 4) was identified, and a superficial parotidectomy was done (Figure 5).

The facial nerve and all its branches were identified and saved (Figures 6a & b). The superficial lobe of the parotid gland was sent for biopsy, which reported intralobular growth with the

replacement of acini by capillary-sized vessels. These vessels were marked by increased cellularity, and compressed the vascular lumina, with a few showing inconspicuous lumens and entrapped adipocytes favoring a diagnosis of Infantile capillary hemangioma of the parotid gland.

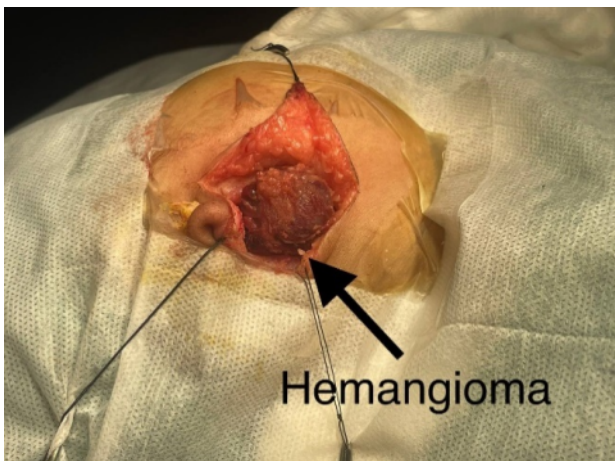


Figure 4

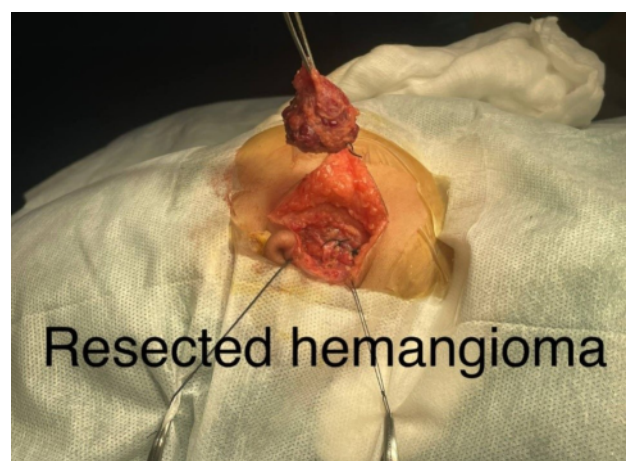


Figure 5

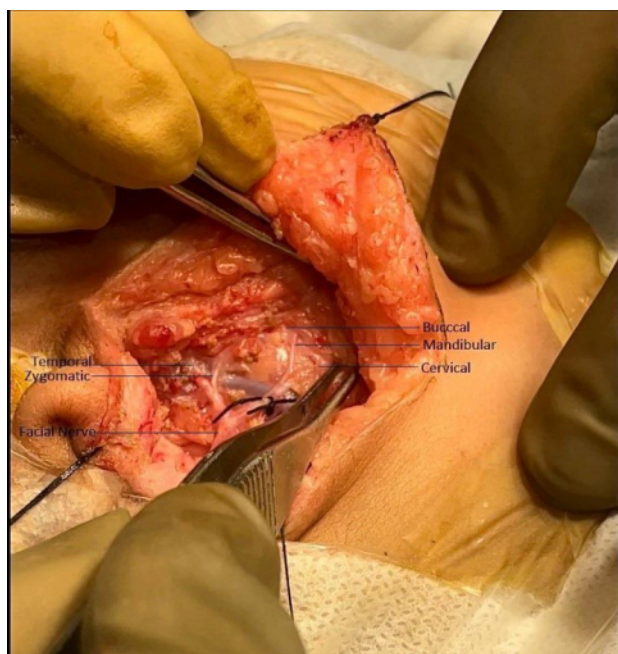


Figure 6(a)

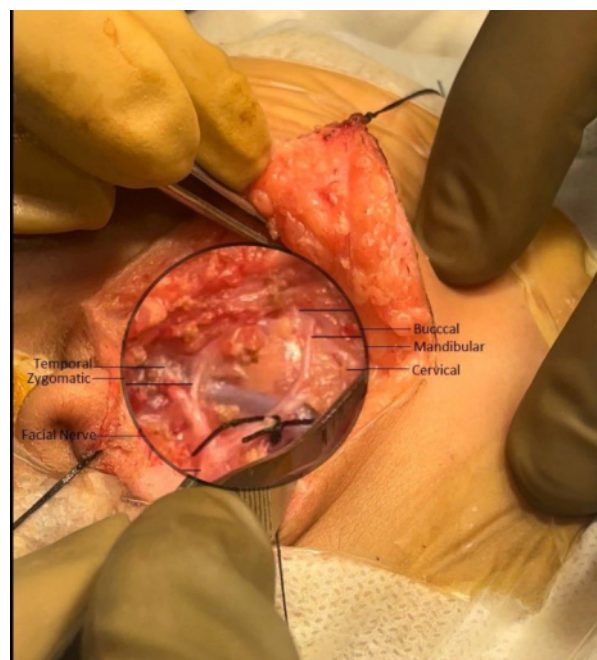


Figure 6(b)

DISCUSSION

Hemangioma of the salivary gland is seen in only 1-5% of all salivary gland tumors, but it is the most commonly seen tumor of the salivary gland in the pediatric population¹. The average age of presentation of these tumors is 3-6 months, and the incidence in males to females is 3:14. In our case, the growth was noticed at 2 months of age.

Hemangiomas have a clear-cut pattern with respect to their development. They grow rapidly during the early months of life and have two growth spurts; the first usually occurs at 1-2 months of life and the second at 4-6 months⁶. Then in the years after, dense fibrous fatty tissue takes the place of the vascular tissue; this is known as the phase of involution. Observations from a molecular point of view state that this occurs due to unevenness between positive and negative regulators of angiogenesis⁶.

Among the salivary glands, hemangiomas have the most affinity for the parotid gland, the reason for this being its embryological development. The parotid gland starts developing during the 6th week of embryogenesis, arising from the buds of the oral ectoderm. They then form solid cords after branching and growing upwards towards the ears. At 10 weeks, they canalize, become ducts, and start secretion at around 18 weeks. On the other hand, the submandibular and sublingual glands arise from the endoderm as opposed to the ectodermal origin of the parotid gland⁶.

The most common presentation of a parotid hemangioma is of a rapidly growing mass in the preauricular region. They are generally of two types, either part of a segmental V3 hemangioma or an isolated focal hemangioma⁴. In our case, it was an isolated focal hemangioma. Focal hemangiomas only involve the parotid gland⁶.

Clinical examination has a pivotal role in diagnosing infantile parotid hemangiomas. Accompanying the clinical examination, radiological studies, particularly MRI, help determine the size and the extent of the tumor² and are the investigation of choice. This diagnostic approach allows clinicians to solidify the diagnosis and avoid unnecessary biopsies.

The line of treatment of an infantile parotid hemangioma is influenced by several factors like size, stage, lesion location, ulcer/bleed over the swelling, and functional obstruction¹. However, there is no one treatment for the condition, and many modalities have been described in various literature. Surgery is one such route that is chosen keeping several factors in mind, like the risk of cardiac failure due to shunting, destruction of the normal anatomical structures, and disfigurement⁶. Additionally, children at the age of two years and above become aware of this disfigurement, hampering their psychological development⁶. Airway or visual axis obstruction or association with Kasabach-Merritt syndrome also calls for surgical intervention.

Surgery with preservation of the facial nerve and its branches and maintenance of facial symmetry can be achieved, as seen in this case. Reinisch et al. have documented 17 patients undergoing parotidectomy for parotid hemangiomas, and the post-operative results analysed were excellent. As reported, they also stated that in a parotid hemangioma, most of the hemangioma is situated superficially to the facial nerve³. Surgery reported in 16 patients by Weiss et al. for parotid hemangioma also reported full facial nerve function post-operatively⁶, also seen in this case. Therefore, surgical excision is a dependable and safe management modality for infantile parotid hemangiomas.

CONCLUSION

The idea of benign neglect, a practice of the past based on an "observe and follow-up" policy, was followed for many years. However, for a rapidly growing mass causing disfigurement and several other life-threatening complications, the absence of definitive medical treatment and stigma of facial deformity after involution advocates active intervention. Surgical excision via a modified Blair incision was performed in this case, where the most worrisome factor was the injury to the facial nerve. However, surgery by a trained surgeon has proven to be a reliable and a safe treatment modality with the preservation of facial symmetry. The scar from the incision also healed with excellent results due to the surgical expertise and the child's young age.

CONFLICT OF INTEREST: None

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