Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Radiology

Imaging Features in Parotid Infantile Hemangioma: A Case Report and Review of Literature

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DOI: 10.36347/sjmcr.2023.v11i06.022 | **Received:** 30.04.2023 | **Accepted:** 25.05.2023 | **Published:** 10.06.2023

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Abstract **Case Report**

Hemangioma of the parotid gland is a benign growth of endothelial cells. It is the most common tumor of the salivary glands in childhood. The initial appearance is usually before 4 weeks of age and achieves full growth before first half of infancy. Diagnostic studies, either imaging or histological ones, are not necessary in the presence of a single lesion with typical clinical history and objective examination. Magnetic resonance imaging (MRI) is the best imaging modality to diagnose and define the extent of parotid hemangioma. Various approaches for treatment are available and Propranolol is considered first line therapy for these tumors. In this paper, we report the case of a female infant who presented with right parotid region swelling, in whom the diagnosis of parotid hemangioma was made based on imaging features.

Keywords: Hemangioma, endothelial cells, tumor, treatment.

INTRODUCTION

Hemangioma of the parotid gland is a benign growth of endothelial cells. It represents only 1% to 5% of all salivary gland tumors, however, it is the most common tumor of the salivary glands in childhood [1]. The female sex is affected three times more frequently than the male sex. The mean age of onset is around 4 months, and most of them are diagnosed during the first 16 months of life [2].

The diagnosis can be made on clinical grounds in typical and obvious cases, and may be confirmed with imaging. Magnetic resonance imaging (MRI) is the best imaging modality to diagnose and define the extent of parotid hemangioma.

Usually, treatment is required in cases with complications, functional impairment or for cosmetic reasons [3].

We report the case of a female infant who presented with right parotid region swelling, in whom the diagnosis of parotid hemangioma was made based on imaging features.

CASE REPORT

A 15-month-old female presented with a progressively increasing swelling over the right side of her face, that was initially noticed at 4 months of age. There was no history of pain, fever, lethargy, refusal to feed, discharge or bleed from the swelling, any rash or oral lesion.

Clinical examination revealed a large swelling in the right parotid region, soft and nontender on palpation, with normal appearance of the overlying skin.

Ultrasound demonstrated a diffuse enlargement of right parotid gland with lobulated contours, and a well-defined isoechoic area that involved almost all superficial lobe of the right parotid gland. On Color Doppler ultrasound, there was increased internal vascularization with numerous large blood vessels within the mass (Figure 1).

Computed tomography (CT) has been requested. Non-contrast images showed an enlarged right parotid gland with an isodense lesion that involve the entire gland. Post-contrast images showed intense enhancement of the lesion (Figure 2).

MRI demonstrated the presence of a lesion in the right parotid space replacing almost the entire gland. The lesion was isointense to the muscle on T1 weighted images and hyperintense and heterogeneous on T2 weighted images. There were also several tortuous and serpiginous vessels within the mass that could be seen as flow voids. After gadolinium administration, an intense and homogeneous enhancement was seen as well as multiple prominent vascular structures (Figure 3).

These imaging features confirmed the diagnosis of hemangioma of the right parotid gland. Treatment with Propranolol was started. The child did not develop any adverse effects from the treatment and after one year there was a marked regression of cervical swelling.

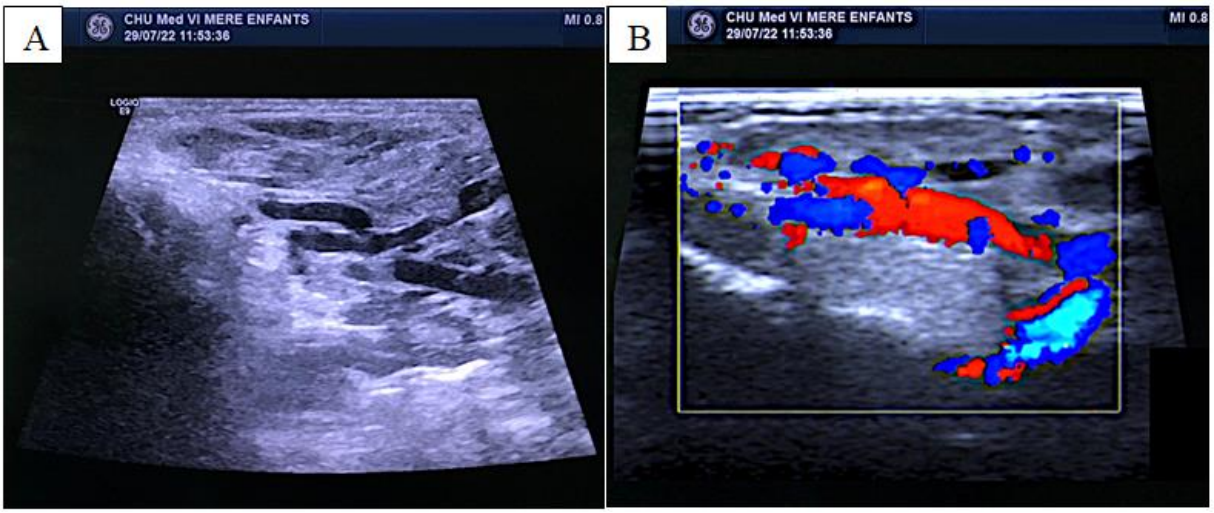

Figure 1: Ultrasound images (A) showing a diffuse enlargement of right parotid gland and a well-defined isoechoic area that involved almost all superficial lobe of the right parotid gland. Color Doppler ultrasound (B) showing increased internal vascularization with numerous large blood vessels within the mass

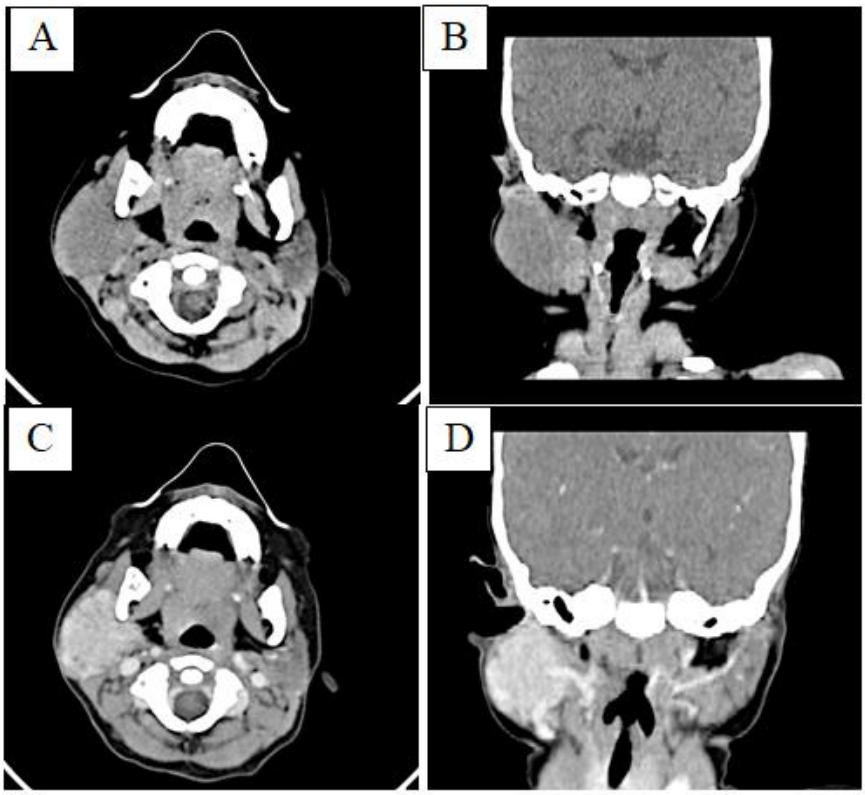

Figure 2: Non-contrast axial (A) and coronal (B) CT images showing an enlarged right parotid gland with an isodense lesion that involve the entire gland. Post-contrast axial (C) and coronal (D) CT images showing intense enhancement of the lesion, fed by a branch emanating from the external carotid artery with a venous drainage branch joining the homolateral external jugular vein

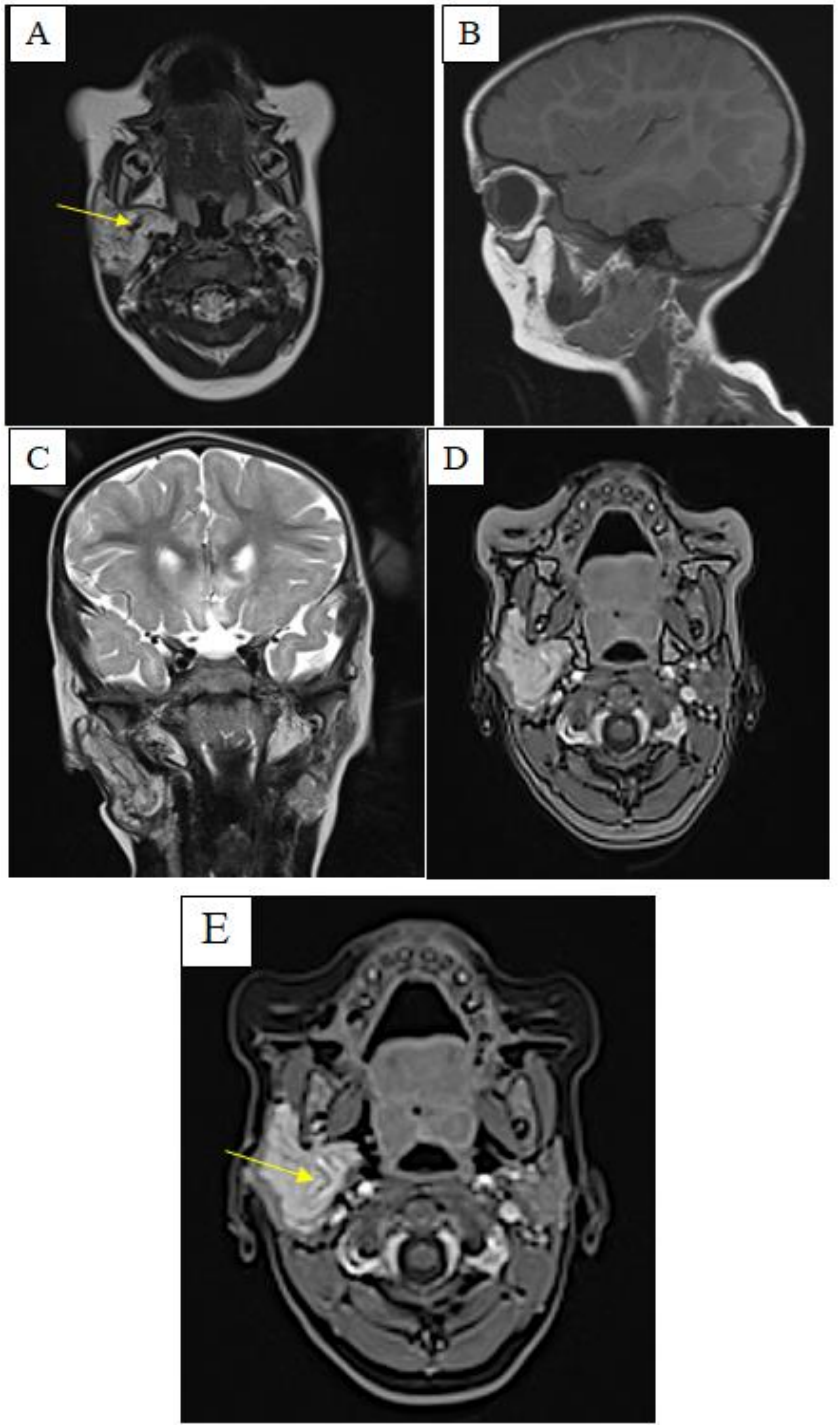

Figure 3: MR images demonstrating T1-isointense (A and B) and T2-hyperintense (C) lesion replacing almost the entire gland, with diffuse homogeneous enhancement on T1 (D) and T1 FAT-SAT images (E). Note the prominent vessels with flow voids in the lesion (yellow arrows)

DISCUSSION

Parotid hemangiomas are the most common benign parotid tumors in children, accounting for more than 50% of benign parotid tumors, and approximately 90% of salivary gland hemangiomas. It is characterised by abnormal proliferation of endothelial cells and aberrant blood vessel architecture. The initial appearance is usually before 4 weeks of age and achieves full growth before first half of infancy. The involution starts around one year of age and is complete in next 3 years. In comparison with infantile hemangiomas occurring in other sites, parotid hemangiomas tend to have a longer proliferative period and a higher incidence of complications because of their unique location and characteristics such as high metabolism, rich blood supply and secretion [3, 4].

Female gender, preterm birth, older maternal age, multiple gestations, low birth weight, maternal

vaginal bleeding during the first trimester, progesterone use, preeclampsia, placenta previa, use of in vitro fertilisation and positive family history have all been linked to the development of infantile hemangioma [3].

Clinically, parotid gland hemangiomas present as asymptomatic soft tissue swellings that cause the overlying skin to obtain a bluish hue. They have the potential to enlarge and cause serious facial deformities impacting the eyes or facial nerve dysfunction [5]. The presence of associated "strawberry skin" spots or the involvement of the skin overlying the lesion helps to confirm the diagnosis [1]. Infantile hemangioma may be part of the PHACE syndrome, characterized by changes in the posterior fossa, head and neck hemangiomas, arterial changes, heart defects or coarctation of the aorta, ocular or endocrine changes and malformations of the sternum.

In the presence of a single lesion with typical clinical history and objective examination, complementary diagnostic studies, either imaging or histological ones, are not necessary. However, in specific cases, imaging studies may be indicated to avoid unnecessary biopsies [6].

Ultrasound is the first-line imaging method given its safety, low cost and the possibility of being performed without sedation [7]. It also allows the exclusion of infectious etiology. The typical ultrasound findings of hemangioma consist of the presence of a homogeneous mass which may enlarge the entire parotid gland, with lobulated contours, fine internal septations and various intratumoral vascular structures which is confirmed by application of color Doppler. The identification of numerous vessels within the lesion is essential for the radiological diagnosis [8].

CT with contrast reveals hyperintense enhanced lobulated mass as observed in our patient [9]. While CT is useful in demonstrating the full extent of the tumor, its relationship to adjacent structures and any skeletal changes, the major drawback of CT is exposure to ionizing radiation [10].

MRI is therefore the secondary investigation of choice. It provides useful information on the size, deep extent of the tumor and its relationship to adjacent structures [11]. It demonstrates the presence of a mass in the parotid gland with lobulated contours, which is isointense to muscle in T1-WI and hyperintense in T2-WI, with prominent vascular flow voids within. After gadolinium administration there is homogenous enhancement and the identification of large tortuous vessels within the lesion [12, 13]. However, it is important to note that flow voids may not be easily identified if the study is performed in the neonatal period. The main drawback of MRI is that it is expensive and sedation is required in the majority of cases [10].

Red cell scintigraphy is considered highly accurate in the diagnosis of head and neck hemangiomas in children. It shows the presence of a uniform, well-circumscribed area of intense uptake [2].

The main differential diagnosis for infantile hemangiomas during infancy is acute parotitis. However, clinically these infants present with fever and swelling, with or without erythema in the pre-auricular area. On sonography, the parotid gland is typically enlarged and heterogeneous, with enlarged intraparotid lymph nodes. Hypoechoic foci may indicate areas of suppuration [10]. The solid component of the hemangiomas distinguishes these lesions from other vascular lesions, including cystic lymphatic malformations (cystic hygroma), as the later does not demonstrate extension beyond the parotid and does not contain prominent vessels. Other differential diagnoses consist of rhabdomyosarcoma, which is rare in childhood; congenital infantile fibroma, usually a heterogeneous lesion (unlike hemangioma); solitary infantile myofibromatosis, which is a hypovascular lesion; and sialoblastoma, which in addition to being very rare does not present flow voids on MRI [1].

The treatment of pediatric infantile hemangioma of the head and neck, and particularly of the parotid gland, is not codified. Given the high probability of spontaneous regression, parotid hemangiomas may not receive treatment [11]. It is recommended only with complications such as ulceration, infection, bleeding and severe growth leading to compression and/or deformity [13].

Various approaches for treatment are available including the administration of anti-angiogenic drugs such as propranolol and corticosteroids and, in some cases, laser therapy, surgery and embolization [6]. Earlier corticosteroids were used as first line therapy, but owing to the serious side effects and rebound phenomenon they are not used much anymore. Multiple studies have demonstrated an improved response rate with Propranolol when compared to corticosteroids. Propranolol is now considered first line therapy for these tumors, it acts via vasoconstriction, inhibition of angiogenesis and stimulation of apoptosis. Overall it is effective and well-tolerated and most serious side-effects can be avoided by appropriate screening and exclusion or minimized by appropriate monitoring [3, 10].

CONCLUSION

In patients with a typical clinical history, the diagnosis of hemangioma of the parotid gland is usually straightforward, obviating the need for further investigation. However, in atypical cases, when there are no overlying cutaneous hemangiomas, the diagnosis

can be challenging. Thus, it is important to know its imaging features in order to be able to identify this lesion on radiological studies and avoid unnecessary biopsies.

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