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Case Report

Unilateral parotid gland double hemangiomas in an infant: A rare case report ☆

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ABSTRACT

Parotid hemangiomas, the most common salivary gland tumors in pediatric patients, are rare vascular tumors that typically present within the first year of life and are more common in females. We report the case of a 3-month-old female infant with a palpable, soft, noncompressible swelling at the right mandibular angle, which became more prominent in the evening. Ultrasonography revealed a large, multilobulated, hypoechoic lesion within the right parotid gland, along with a smaller, similarly vascularized lesion, and MRI confirmed the presence of 2 hemangiomas. This case is notable as no previous reports describe double hemangiomas in the same parotid gland. Imaging techniques such as ultrasound and MRI were essential in the differential diagnosis, distinguishing hemangiomas from conditions like acute parotitis and cystic lymphatic lesions. Early recognition and accurate diagnosis of parotid hemangiomas are key to determining the appropriate management, with treatment options ranging from observation to pharmacological therapy or surgery depending on the size and complications. In this case, the patient was referred for beta-blocker therapy.

Introduction

Salivary gland tumors are rare, comprising approximately 2% of all neoplasms. Most parotid gland tumors are benign. In children, parotid hemangiomas represent about half of all salivary gland tumors, with 75% appearing at birth, 90% in the first year of life, and a higher incidence in females compared to males, with a ratio of 3:1 [1,2].

Parotid hemangiomas are rare benign tumors composed of blood vessels lined with endothelial cells within a connective tissue stroma. They lack a capsule and can infiltrate the gland, replacing glandular elements and leaving only the ducts. These hemangiomas typically appear during the first months of life and reach their maximum growth within the first 10 years. They may undergo spontaneous regression during this period, a process more common in cavernous vascular malformations. Most parotid hemangiomas are

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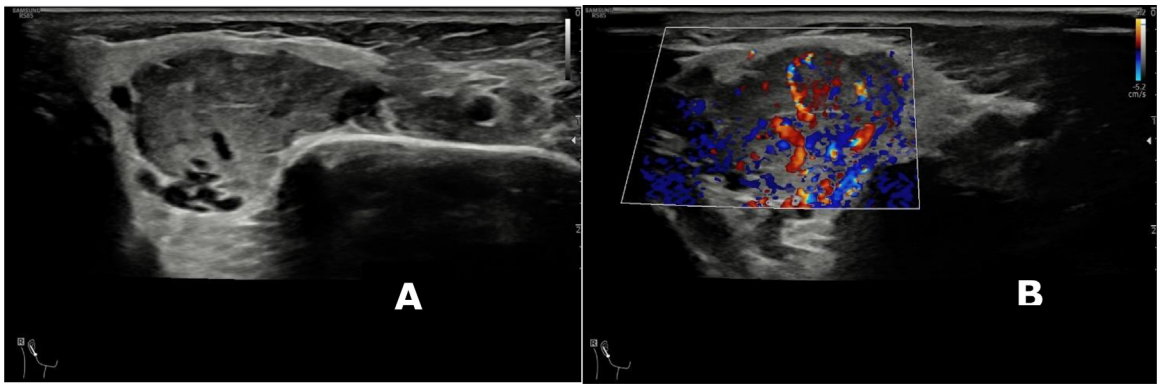

Fig. 1 – (A) Large formation occupying the right parotid gland. The lesion had polylobulated contours, a hypoechoic appearance compared to the small portion of the remaining parotid, relatively homogeneous content with the presence of fine hyperechoic septa and multiple and coarse vessels prominent in the context. (B) Multiple and coarse vessels prominent in the context and marked vascularization at the evaluation with color Doppler.

asymptomatic, but a small portion may cause complications such as rapid growth, visible facial swelling, obstruction of the ear canal or subglottic area, bleeding, ulceration, or infection [1,3].

Case report

We report the case of a 3-month-old infant with a palpable, soft, noncompressible swelling at the right mandibular angle. The swelling became more evident in the evening, prompting further evaluation. The patient underwent an ultrasound examination with a Samsung V8 ultrasound scanner using a high-frequency linear transducer LA2-14A, with a frequency range of 2.0 MHz to 14.0 MHz. Subsequently, she underwent an MRI of the neck at another hospital.

A 3-month-old girl, referred from the pediatrics department of our hospital, presented with a clinically palpable swelling at the right mandibular angle. The swelling was soft and noncompressible on palpation, with no skin alterations. The mother reported that the swelling was more evident in the evening and had increased in size over time. Laboratory blood tests were normal, with no signs of inflammation (normal ESR, CRP) and normal white blood cell count. The patient was afebrile, with normal temperature, and without any clinical symptoms.

Ultrasound examination revealed a large mass in the right parotid gland, which was swollen and enlarged (Fig. 1). The lesion had polylobulated contours, a hypoechoic appearance compared to the small portion of the remaining parotid gland, showing a relatively homogeneous content with fine hyperechoic septa and multiple coarse vessels, which were well visualized with marked vascularization on color Doppler evaluation (Fig. 1). Vascular peduncles were also visible at the base of the lesion. Another smaller formation with similar ultrasound characteristics was noted nearby (Fig. 2). Evaluation of the contralateral parotid gland revealed no structural alterations (Fig. 3). There were no bilateral cervical lymphadenopathies.

The findings detected by ultrasound, the clinical and laboratory investigations allowed us to diagnose double parotid hemangioma. An MRI performed later at another hospital, of which unfortunately no images were provided to us, confirmed the diagnosis of double parotid hemangioma.

Discussion

There are no reported cases in the literature of double hemangiomas in the same parotid gland. In a retrospective study by Harris and Phillips involving 15 pediatric patients with parotid hemangioma, most cases (73.3%) involved the left parotid gland, while 26.7% affected the right side. No cases of bilateral parotid hemangiomas were reported [4].

Ultrasound is the imaging modality of choice for studying salivary gland tumors because, like MRI, it is noninvasive and does not use ionizing radiation. Parotid hemangiomas typically present with polylobulated contours, well-defined edges, and variable echogenicity, ranging from homogeneous to heterogeneous. Color Doppler evaluation often reveals increased vascularity due to prominent vessels within the lesion. CT scans can be useful for determining relationships with surrounding structures. On CT, parotid hemangiomas have polylobulated, circumscribed contours, with the gland appearing enlarged and of homogeneous density. MRI is valuable for evaluating the characteristics and extent of parotid hemangiomas. On MRI, these hemangiomas appear as lesions with well-defined, lobulated contours, homogeneously hypointense on T1-weighted images, with an intermediate signal between muscle and fat, hyperintense on T2-weighted images, and show homogeneous enhancement on T1-weighted images after contrast administration. Hemangiomas may appear inhomogeneous if there are foci of adipose replacement within the lesion [5,6].

In considering differential diagnoses, acute parotitis should initially be excluded. Clinically, children with parotitis present with fever, swelling, and erythema of the preauricular

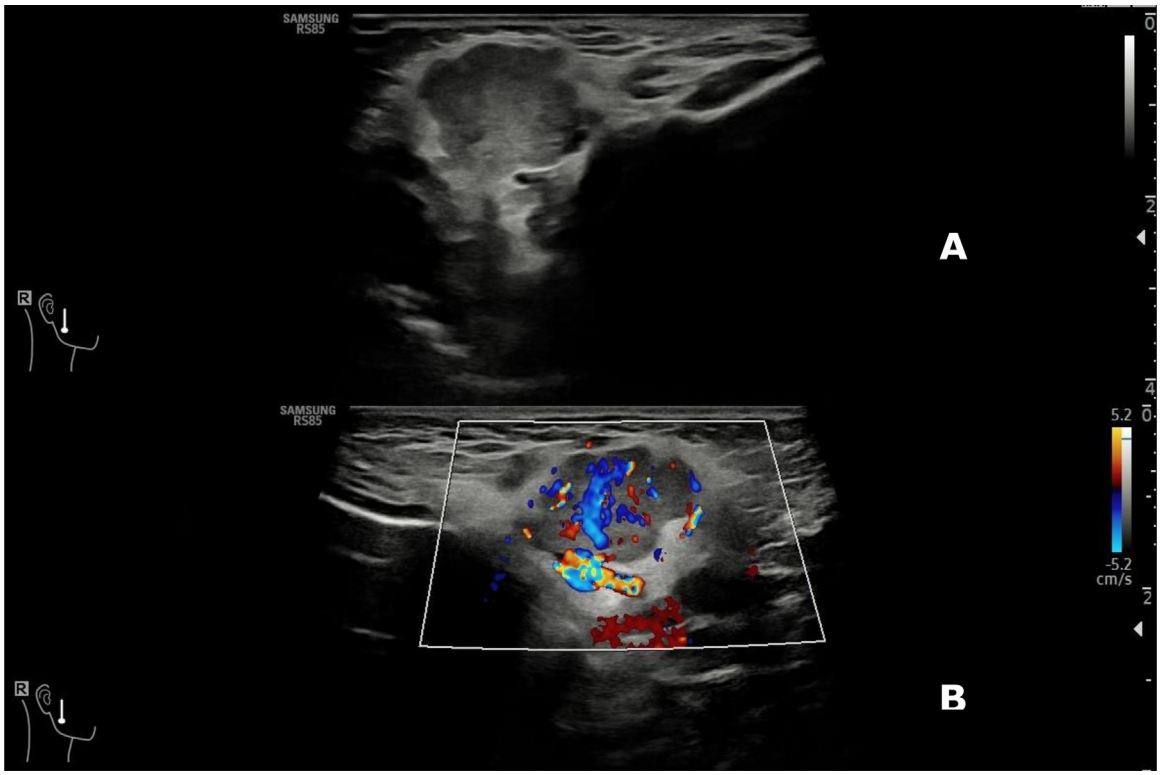

Fig. 2 – (A) Another smaller formation with the same ultrasound characteristics. (B) Multiple and coarse vessels prominent with marked vascularization at the evaluation with color Doppler in the context of the smaller formation.

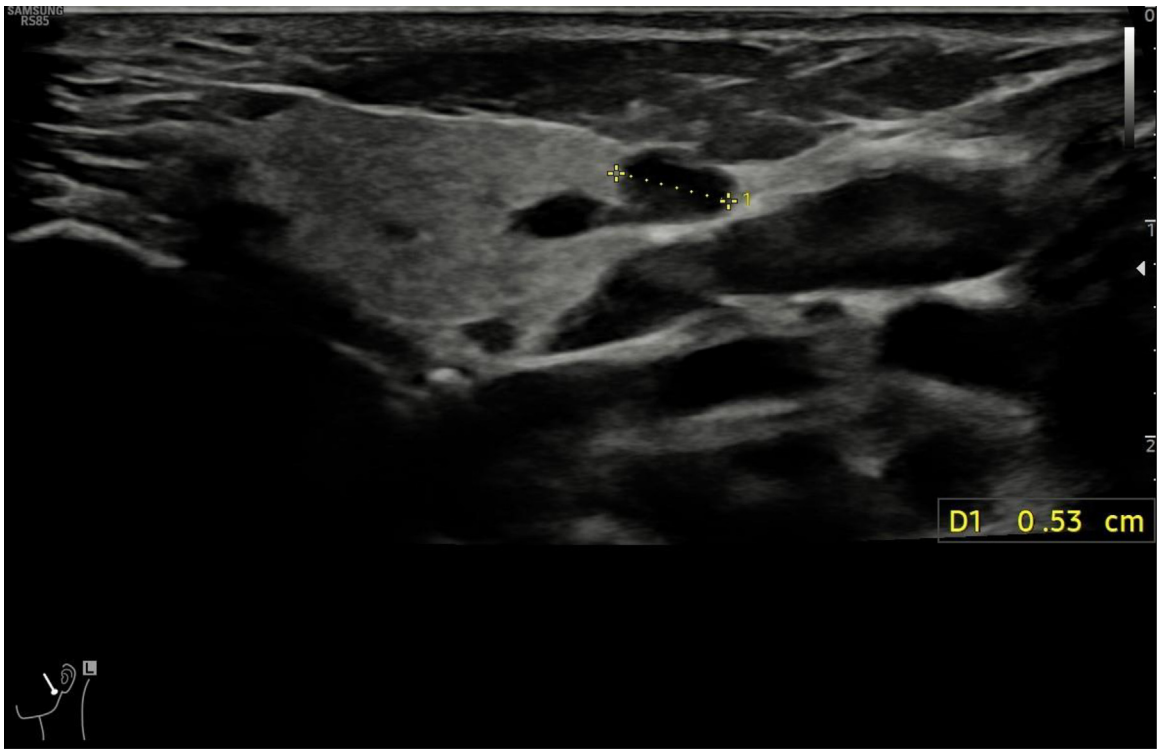

Fig. 3 – Regular contralateral parotid gland.

area. On ultrasound, the parotid gland appears enlarged and swollen, with heterogeneous content due to numerous hypoechoic foci [7].

Other lesions to consider in the differential diagnosis of parotid hemangioma include cystic lymphatic lesions (cystic hygroma), which do not contain prominent internal vessels; rhabdomyosarcoma, which infiltrates surrounding structures and is less vascularized; infantile fibrosarcoma of the parotid gland, which is highly vascularized but has a heterogeneous content; solitary infantile myofibromatosis, which may show rapid growth followed by involution and is poorly defined and nonvascularized; and sialoblastoma, a rare congenital tumor of the salivary glands that shows reduced vascularization and heterogeneous content on ultrasound or MRI compared to parotid hemangioma [8].

The association of parotid hemangiomas with PHACE syndrome should be considered, especially in cases involving large facial hemangiomas. PHACE syndrome encompasses a spectrum of disorders, including Posterior fossa malformations, Hemangiomas, Arterial anomalies, Cardiac defects, and Eye abnormalities. Magnetic Resonance Angiography (MRA) plays a critical role in evaluating the cerebrovascular abnormalities commonly associated with PHACE syndrome. These anomalies, such as arterial stenosis, persistent embryonic arteries, or intracranial aneurysms, can significantly influence the therapeutic approach and management of the patient, as they may predispose to ischemic strokes or other complications [11].

Considering venous malformations in the differential diagnosis of parotid hemangiomas, the presence of phleboliths can serve as a distinguishing feature. Venous malformations, composed of abnormally formed, dilated veins, often present with phleboliths—small calcified thrombi—visible on plain radiographs or CT scans. These calcifications are not typically found in hemangiomas, making them a useful diagnostic marker. This distinction is crucial when planning treatment, as venous malformations and hemangiomas often require different therapeutic strategies [12].

Treatment strategies vary and should be personalized. A conservative approach may be appropriate for small lesions with minimal growth, as they may undergo spontaneous involution, requiring no intervention. However, in cases of rapid growth, where aesthetic concerns or complications such as cardiac failure due to shunts arise, close monitoring and evaluation of the patient's clinical condition are essential. Pharmacological treatments have included corticosteroids (both systemic and local) and interferon α-2a, but the most effective drug has been propranolol, a nonselective beta-blocker. Its mechanism of action is not fully understood, but it is hypothesized to induce apoptosis of capillary endothelial cells by decreasing proangiogenic vascular endothelial growth factors and fibroblast growth factors. Propranolol is well-tolerated with minimal side effects. Another treatment option is sclerotherapy with direct injection of bleomycin, which induces apoptosis of angiogenic cells.

Surgical treatment, due to potential complications such as facial nerve damage, is reserved for patients with large lesions, complications like ulceration or bleeding, functional obstructions, or facial deformities [4,9,10].

Conclusions

Recognizing the clinical and imaging characteristics of parotid hemangiomas is crucial for differentiating them from other lesions and avoiding unnecessary biopsies in young patients. Once a diagnosis of parotid hemangioma is made, treatment should be personalized based on the lesion's location, size, and associated complications or facial deformities. In the rare case of double hemangiomas of the right parotid gland presented here, the patient was referred to a specialized center for pharmacological therapy with a beta-blocker.

Patient consent

The author confirms that he obtained written informed consent from the patient's parents for the publication of the case.

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