

Infantile Parotid Hemangioma With Diagnostic Dilemma: A Case Report

Kamel T Abidi^{1*}, Naglaa M Kamal^{2*} , Khaled A Althobaiti¹, Sami D Althobaiti¹, Yasser A Halabi¹ and Saad A Alalayani¹

¹Department of Pediatrics, Alhada Armed Forces Hospital, Taif, Saudi Arabia. ²Department of Pediatrics & Pediatric Hepatology, Faculty of Medicine, Cairo University, Cairo, Egypt.

Clinical Medicine Insights: Case Reports
Volume 15: 1–3
© The Author(s) 2022
Article reuse guidelines:
sagepub.com/journals-permissions
DOI: 10.1177/11795476211073385



ABSTRACT

INTRODUCTION: Salivary gland tumors are uncommon in children; hemangiomas are one of them. We report a case of infantile hemangioma of the parotid gland which posed a diagnostic dilemma.

PATIENTS AND METHODS: Four-month-old infant presented with swelling at the parotid gland region, which was progressively increasing. There was a diagnostic dilemma with initial misdiagnosis as acute parotitis. Imaging studies with ultrasonography and CT with contrast were requested.

RESULTS: Imaging studies revealed hyperintense lobulated mass suggestive of parotid hemangioma. Vascular consultation recommended conservative management with follow-up after 3 months. On follow-up, there was an increase in mass size, and propranolol was started. Swelling showed a good response on the next follow-up visit and is still on medical management.

CONCLUSION: Parotid hemangiomas frequently pose a diagnostic dilemma, but the typical noninflammatory nature of the swelling and radiological evaluation confirms the diagnosis.

KEYWORDS: Infantile, parotid, hemangioma

RECEIVED: July 20, 2021. **ACCEPTED:** December 22, 2021.

TYPE: Case Report

FUNDING: The author(s) received no financial support for the research, authorship, and/or publication of this article.

DECLARATION OF CONFLICTING INTERESTS: The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

CORRESPONDING AUTHOR: Naglaa M Kamal, Department of Pediatrics and Pediatric Hepatology, Faculty of Medicine, Cairo University, Cairo, Egypt. Email: nagla.kamal@kasralainy.edu.eg

Introduction

Salivary gland tumors are uncommon in children with approximately less than 5% incidence. Among these tumors, are hemangiomas, although rare but still they are considered the most common benign tumor of the salivary gland, accounting for about 50% of all cases. Female affected 3 times more than male and age of 4 months is the usual age of presentation.¹⁻³

We herein report a 4-months-old female patient with infantile hemangioma of the parotid gland misdiagnosed as acute parotitis.

Case Description

MA is a 4-month-old girl, full-term, product of spontaneous vertex delivery who presented to the hospital with rapidly increasing left parotid swelling noticed initially at age of 3 months associated with bluish discoloration of the overlying skin, no other swelling, no fever, no history of trauma, and no contact with household animals. Her body system review was unremarkable.

Her parents sought medical advice multiple times at private and primary health care facilities. She received one course of antibiotics with no improvement.

On presentation to our hospital, she was initially assessed by an otolaryngologist, who requested ultrasonography (US) of the parotid gland. He diagnosed her as parotitis based on US's picture of heterogenous echo pattern mass with increased vascularity.

The patient was then referred for pediatric service for a second opinion. The patient was admitted under pediatric care for further evaluation. On examination, the patient looked well, vitally stable, not in distress with no dysmorphic features. Her growth parameters were appropriate for her age. There was left parotid swelling about 4 × 5 cm in diameter, soft, mobile, with bluish discoloration of the overlying skin. The swelling was not tender, non-pulsatile, had an ill-defined border, no sinus formation, and no bruit. There was no airway compromise. No other swellings, no organomegaly and no skin rash.

A computed tomography (CT) scan with intravenous contrast was requested and revealed an enhanced large soft tissue lesion at the parotid region suggestive of parotid hemangioma (Figure 1).

Accordingly, the vascular surgeon was consulted. He recommended conservative management with regular follow-up. Counseling of the parents about the condition and its usual natural course of spontaneous regression was carried and the patient was discharged home in good condition with regular follow up with multidisciplinary care team of a vascular surgeon, pediatrician, and otolaryngologist.

On 3 months follow up, the swelling size had increased to 4.6 × 5.5 cm, and decision to start medical management with propranolol 1 mg/kg/day in 2 divided doses. Education about drug mechanisms of action and side effects was provided.

The patient is currently in her third month of management with follow-up visits and phone calls for any urgent complaints during the treatment period. There was a good response to

*Authors share first author equally.



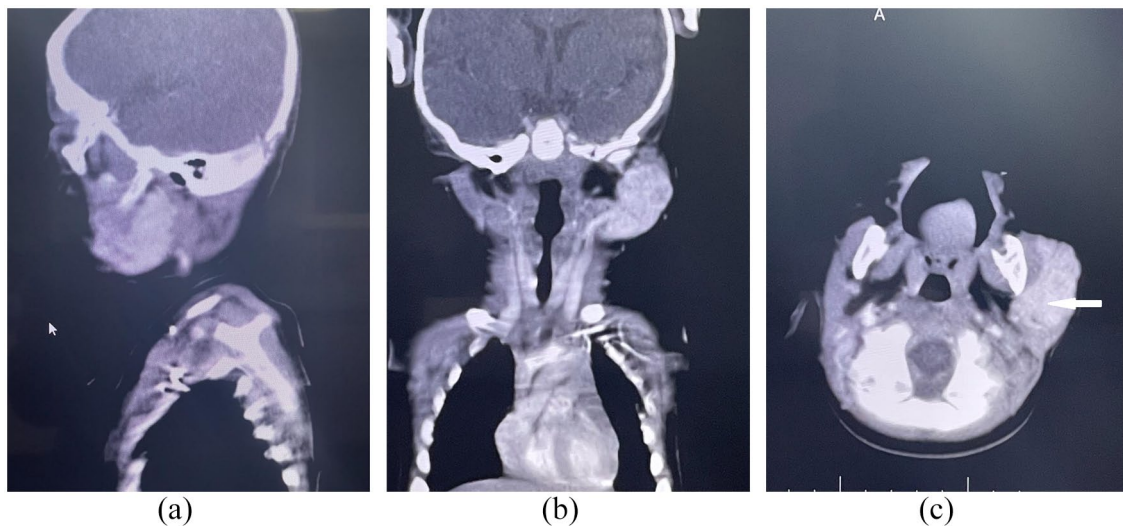


Figure 1. CT with IV contrast showing enhanced lobulated soft tissue mass at the parotid region, suggestive of parotid hemangioma.

Table 1. A literature review of reported cases of infantile parotid hemangiomas.

AUTHOR	AGE OF PRESENTATION/ GENDER	CLINICAL FINDINGS	RADIOLOGICAL FINDINGS
Greene et al ⁴	100 infants presented between year 1975 and 2002, female-to-male ratio was 4.5:1, 40% were on the right side, 36% were on the left, and 24% were bilateral. Diagnostic modalities used were not mentioned		
Mantadakis et al ⁵	37 days old, boy	Large right parotid swelling	US: engorged right parotid gland MRI: enlarged right parotid gland with multiple flow voids.
Kotrashetti et al ¹	2.5-month, boy	Left parotid swelling	US: enlarged parotid with increased vascularity MRI: well defined lobulated homogenous lesion
John et al ⁶	3-month, girl	Left parotid swelling	US: hypoechoic lobulated lesion CT: intense enhancement of parotid gland
Weber et al ⁷	Thirty-five infants presented at less than 4 months of age, Female to Male: 21:13	#19 patients had left sided hemangioma, 2 had bilateral lesions & 14 were right sided. #The majority (29 patients) presented due to localized swelling or palpable mass; the remainder had a cutaneous lesion, but no palpable mass at the time of presentation.	US: uniformly vascular lesion with pulsatile fast-flow seen on Doppler. MRI: hyperintense lesion on T2-weighted images, isointense on T1, with intense enhancement post-contrast.
Rio et al ²	1 month and 18-day old infant	Right sided parotid hemangioma of sudden onset, painless, no alteration of the overlying skin.	US: enlarged and heterogeneous parotid gland with increased vascularization. MRI: lobulated mass replacing almost the entire right parotid gland.
Siddiqui et al ⁸	2-month, boy	Progressively increasing swelling in left pre auricular and infra-auricular area for 5 days	US: revealed increased vascularity MRI: hyperintense lesion on T2 images with intense post contrast enhancement on T1
Our case	4-month, girl	Progressively increasing left submandibular swelling	US: heterogenous echopattern with increased vascularity CT: enhanced large soft tissue lesion with increased vascularity

propranolol therapy with a decrease in hemangioma size in all its dimensions and the patient is still on regular follow-up for further plan.

Discussion

Although infantile parotid hemangiomas are a rare condition, but they are still considered the commonest infantile

parotid tumor. Table 1 summarizes previously reported cases.^{1,2,4-8}

Parotid gland hemangiomas display a predilection for females. Bluish or purple discoloration of the overlying skin is a common clinical sign as noticed in our patient.¹⁻³

Although some affected children were noticed at birth, infantile hemangiomas typically appear during the subsequent

6 months.^{2,9} In our case, the growth was prominent beyond the first month of age.

In addition to the characteristic clinical presentation, radiological studies are very helpful to confirm the diagnosis and to avoid unnecessary biopsies.⁹ Ultrasound usually shows an iso-echoic or hypoechoic lobulated lesion with increased vascularity at the parotid region as noticed in our patient.^{2,6,7}

CT with contrast reveals hyperintense enhanced lobulated mass as observed in our patient.^{6,7} magnetic resonance imaging (MRI) is very helpful to determine the size and extension of the tumor and usually shows lobulated homogenous mass. MRI is preferred over CT to avoid radiation risk.^{2,5-7,10}

Among the rare complications of parotid, hemangiomas are heart failure and Kasabach-Merit syndrome. It is of note that our patient had a normal platelets count.^{4,7}

Various approaches for treatment are available including propranolol, steroids, interferon, sclerotherapy, and surgery but the definitive treatment is still controversial.^{3-5,10,11}

In the last decade, propranolol 1-1.5 mg/kg/day has emerged as a line of therapy for infantile hemangioma with good response in the form of evolution of the swelling, but recurrence may occur upon discontinuation of the medication patient.^{3-5,10,11}

In our patient, the vascular surgeon initially preferred to closely observe the child without medical or surgical intervention because of the benign nature of the tumor and expected regression within months in most cases. On follow-up, due to enlargement of the swelling, propranolol therapy was started with good response, no complications and the patient is currently in his third month of management with regular follow-up.

Conclusions

Infantile parotid hemangiomas usually pose diagnostic dilemmas but the typical history and clinical examination with radiological evaluation usually confirm the diagnosis. In the differential diagnosis of any nonpainful parotid swelling in infancy, it should be put in the differential diagnosis.

Acknowledgements

To the patient and his family. To Miss Sara AS Abosabie, Medical Student, 4th Semester, Charite' Universitätsmedizin Berlin, Berlin, Germany. We thank her for her help in collecting patient's data and for her help in performing the radiological part of the manuscript. She also helped in editing the manuscript and English proofreading.

Author Contributions

KTA: Diagnosed the patient, set the idea of the study, designed the study, and critically analyzed the data. KTA, NMK, KAA, SDA, YAH, SAA: collected data, reviewed literature, drafted the manuscript. All authors reviewed and approved the manuscript for final publication.

Availability of Data and Materials

All data and materials related to the study are included in the current manuscript.

Ethical Approval and Consent to Participate

The study was approved by the research and ethical committee of the participating hospitals. All parents of enrolled children signed written informed consents for the participation of their children in the current study.

Consent for Publication

All parents of enrolled children signed written informed consents for publication of the current study.

ORCID iD

Naglaa M Kamal  <https://orcid.org/0000-0002-8535-3838>

REFERENCES

- Kotrashetti V, Sonawane V, Bainade K, Agarwal R. Infantile hemangioendothelioma of the parotid gland. *Inter J Clin Pediatr*. 2015;4:184-185.
- Rio G, Costa NA, Fernandes JS, Torrao H, Silva PO. Parotid hemangioma in a child: an uncommon imaging diagnosis. *Acta Radiol*. 2019;31:19-22.
- Ji Y, Chen S, Xu C, Li L, Xiang B. The use of propranolol in the treatment of infantile haemangiomas: an update on potential mechanisms of action. *Br J Dermatol*. 2015;172:24-32.
- Greene AK, Rogers GF, Mulliken JB. Management of parotid hemangioma in 100 children. *Plast Reconstr Surg*. 2004;113:53-60.
- Mantadakis E, Tsouvala E, Deftereos S, Danielides V, Chatzimichael A. Involvement of a large parotid hemangioma with oral propranolol: an illustrative report and review of the literature. *Case Rep Pediatr*. 2012;2012:353812.
- John DR, Shingade R, Patel J. A case of parotid hemangioma of infancy: role of ultrasound and Doppler in the diagnosis. *Ind J Neo Medicine Res*. 2016;4:10-12.
- Weber FC, Greene AK, Adams DM, et al. Role of imaging in the diagnosis of parotid infantile hemangiomas. *Int J Pediatr Otorhinolaryngol*. 2017;102:61-66.
- Siddiqui A, Azad C, Singh DK. Infant with a parotid swelling: beware of the rare. *Sudan J Paediatr*. 2021;21:95-97.
- Harris J, Phillips JD. Evaluating the clinical outcomes of parotid hemangiomas in the pediatric patient population. *Ear Nose Throat J*. 2021;100:NP242-NP245.
- Sinno H, Thibaudeau S, Coughlin R, Chitte S, Williams B. Management of infantile parotid gland hemangiomas: a 40-year experience. *Plast Reconstr Surg*. 2010;125:265-273.
- Soltani AM, Reinisch JF. Algorithmic approach to the management of hemangiomas. *J Craniofac Surg*. 2011;22:585-588.