World Journal of Advanced Research and Reviews
eISSN: 2581-9615 CODEN (USA): WJARAI
Cross Ref DOI: 10.30574/wjarr
Journal homepage: https://wjarr.com/

(RESEARCH ARTICLE)

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Management of infantile hemangiomas in the otorhinolaryngological (ENT) region

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World Journal of Advanced Research and Reviews, 2025, 25(02), 1979-1983

Publication history: Received on 10 January 2025; revised on 15 February 2025; accepted on 18 February 2025

Article DOI: https://doi.org/10.30574/wjarr.2025.25.2.0549

Abstract

Infantile hemangiomas (IHs) are the most prevalent benign vascular tumors in pediatric populations, with an incidence of approximately 5 to 10% in infants under one year of age. IHs in the otorhinolaryngological (ENT) region present significant diagnostic and therapeutic challenges due to their potential impact on critical functions such as respiration and phonation.

This study provides a comprehensive analysis of the epidemiological and clinical characteristics, therapeutic modalities, and outcomes of IHs in the ENT region.

Despite therapeutic advancements, the complex nature of IHs in the ENT region necessitates a multidisciplinary approach for optimal management.

This study highlights the imperative of early diagnosis and individualized therapeutic strategies to mitigate complications and enhance patient outcomes.

Keywords: Infantile Hemangioma (IH); Benign Vascular Tumor; Otorhinolaryngology (ENT); Cervico-Facial Region; Propranolol.

1. Introduction

Infantile hemangioma (IH) is the most prevalent benign vascular neoplasm in pediatric populations, with an incidence estimated at approximately 5 to 10% of infants under one year of age [1]. This neoplasm is characterized by a benign proliferation of vascular endothelial cells, typically manifesting within the initial weeks postpartum. The natural history of IH generally includes a rapid proliferative phase during the first year, followed by a quiescent plateau phase, and culminating in spontaneous involution over subsequent years [1,2]. While the majority of IHs are asymptomatic and undergo spontaneous regression, approximately 10% necessitate clinical intervention due to complications such as ulceration, hemorrhage, aesthetic disfigurement, functional impairment, or potentially life-threatening sequelae contingent on their anatomical location [2,3].

Among the various locations of infantile hemangiomas (IH), the cervico-facial region, including the otorhinolaryngological (ENT) areas, represents a distinct category. This location poses diagnostic and therapeutic challenges due to its potential functional impact on respiration, feeding, phonation, and vision. Additionally, the proximity to the upper airways or vital structures such as the larynx or trachea can lead to severe complications requiring specialized management. The aesthetic impact, particularly in facial locations, also requires clinical and therapeutic evaluation [4].

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The introduction of propranolol in 2008 as the reference treatment has revolutionized the management of IHs. This oral beta-blocker has demonstrated remarkable efficacy in reducing the vascularization and size of hemangiomas, while maintaining a favorable safety profile. However, challenges persist in the treatment of IHs in the ENT region due to the specific characteristics of these lesions, their clinical variability, and the necessity for early intervention in certain cases. Recent advances in the use of topical beta-blockers such as timolol, as well as the exploration of new therapeutic modalities like pulsed dye lasers, have expanded the treatment options for these complex lesions.

Despite their frequency and clinical significance, few studies have focused on the epidemiological and clinical phenotype, therapeutic modalities, and evolution of IHs located in the ENT region. Available data often remain fragmented, lacking studies that integrate these dimensions into a comprehensive analysis specific to this anatomical location.

In this context, we present the results of the first study aiming to comprehensively describe the epidemiological and clinical phenotype, therapeutic approaches, and evolution of infantile hemangiomas in the ENT region. The objective is to provide precise data enabling a better understanding and optimized management of these complex lesions [3,5].

2. Materials and Methods

Descriptive and Analytical Cross-Sectional Study Including Patients Managed for Infantile Hemangioma in the Otorhinolaryngological (ENT) Region at the Dermatology and Venereology Department of Ibn Rochd University hospital of Casablanca over the last 15 years (January 2009 to December 2023).

3. Results

A total of 354 cases of infantile hemangiomas were collected, with 105 hemangiomas located in the otorhinolaryngological (ENT) region. The mean age at diagnosis was 6 months, with a range from 15 days to 24 months. The cohort demonstrated a female predominance, with 75% of cases being female, resulting in a female-to-male sex ratio of approximately 3:1.

Figure 1 Clinical aspect of a segmental mandibular hemangioma before and after 1 year of oral propranolol

The mean age at the onset of symptoms was 23 days. The patient history was notable for multiparity in 56% of cases, consanguinity in 15%, and prematurity in 4.7%. Hemangiomas were located on the lips in 49 cases (47%) some of which were segmental mandibular hemangiomas with a beard distribution. (*Fig1*), the nasal tip in 35 cases (33%), the neck in 14 cases (13%) (*Fig2*), the ears in 11 cases (10%), the parotid region in 4 cases (4%), and the subglottic area in 2 cases (2%).

Figure 2 Clinical aspect of a tuberous hemangioma of the neck and trunk treated with oral propranolol

In terms of morphology, 20 patients (19%) had subcutaneous hemangiomas, 22 patients (21%) had tuberous hemangiomas, and 63 patients (60%) had mixed hemangiomas (***Table 1***).

Table 1 Locations of Infantile Hemangiomas in the Otorhinolaryngological (ENT) region

Location	Number of Cases	Percentage (%)
Lips	49	47%
Nasal Tip	35	33%
Neck	14	13%
Ears	11	10%
Parotid Region	4	4%
Subglottic Area	2	2%

PHACES syndrome was identified in 4 patients (1.1%), with 1 patient (0.3%) presenting a Dandy-Walker malformation (***Fig3***) and 3 patients (0.8%) having simple hydrocephalus.

The primary indications for oral propranolol treatment included periorificial hemangiomas with functional impairment in 63 cases (60%), Cyrano hemangiomas in 35 cases (33%), large hemangiomas causing facial disfigurement, hemangiomas with significant subcutaneous components in 20 cases (19%), and ulcerated hemangiomas in 18 cases (17%).

The main complications observed were hemorrhage in 19% of cases, ulceration in 11%, and necrosis and superinfection in 4.7% of cases each. The average duration of treatment was 17 months, with propranolol dosed between 2 and 3 mg/kg/day. Atenolol was used in 2 patients with a history of recurrent asthmatic viral bronchiolitis.

All patients exhibited clinical remission, characterized by blanching and decreased infiltration of the hemangioma to varying degrees (***Fig1***). However, 4 patients experienced recurrence after discontinuing treatment.

Figure 3 Clinical aspect of a segmental mandibular hemangioma as part of PHACE syndrome with Dandy-Walker malformation before and after 1 year of oral propranolol

4. Discussion

The management of infantile hemangiomas (IHs) in the otorhinolaryngological (ENT) region presents clinical challenges due to the potential functional impacts on vital activities such as respiration, feeding, phonation, and vision (3, 4). This study highlights several critical aspects in the management and outcomes of IHs localized in the ENT region.

Our study reveals a higher prevalence of IHs in females, consistent with existing literature that reports a female-to-male ratio of approximately 3/1 (1,6). The mean age at diagnosis was 6 months, with symptoms typically beginning around 23 days of life (1,6,7). The most common locations for IHs in the ENT region were the lips, nasal tip, and neck, with a significant number of cases also affecting the ears, parotid region, and subglottic area. These findings underscore the importance of early diagnosis and intervention to prevent complications. For segmental hemangiomas in the ENT region, it is essential to systematically search for syndromic forms such as PHACES syndrome, due to the associated risks of complex congenital malformations that may require multidisciplinary management (2,3,4).

The introduction of propranolol in 2008 has revolutionized the management of IHs, including those in the ENT region (2, 5). Our study confirms the efficacy of propranolol, with 80% of patients showing positive clinical responses (2, 5). The use of topical beta-blockers such as timolol, though less common, has also shown promise in treating superficial lesions (2,3). Additionally, pulsed dye laser therapy is effective in reducing vascularization and lesion size (3,8).

Despite the overall success of propranolol therapy, our study identified several complications that necessitated specialized management. Hemorrhage, ulceration, and necrosis were significant concerns. The persistence of some lesions and the recurrence after treatment discontinuation emphasize the necessity for continued monitoring and potential retreatment (3,4,6).

This study underscores the importance of early and accurate diagnosis, followed by appropriate therapeutic interventions, in managing IHs in the ENT region (2,3, 4). The variability in clinical presentation and response to treatment requires individualized management plans. Multidisciplinary teams, including dermatologists, otorhinolaryngologists, and pediatricians, are crucial for optimizing patient outcomes (3).

Further research is needed to refine therapeutic protocols and explore new treatment modalities for IHs in the ENT region (2,3). Longitudinal studies focusing on long-term outcomes, recurrence rates, and the effectiveness of combination therapies will provide valuable insights into optimizing care for this patient population (2,7). Additionally, genetic and molecular studies may reveal underlying mechanisms that could lead to targeted therapies and improved prognostic indicators (3).

5. Conclusion

Propranolol has proven to be an effective first-line treatment, significantly reducing the size and vascularization of IHs and has radically transformed the prognosis and management of infantile hemangiomas in the otorhinolaryngological (ENT) region. Surgery is no longer considered the first-line treatment and should be reserved for difficult, refractory cases or those with absolute obstruction. This approach reduces invasive interventions and favors less aggressive but effective treatments, thereby improving patients' quality of life.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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