© Pediatr Med Rodz 2022, 18 (1), p. 98–101
DOI: 10.15557/PiMR.2022.0014

Otrzymano: 10.06.2021
Zaakceptowano: 25.03.2022
Opublikowano: 31.05.2022

Izzah Akashah[1], Jeyasakthy Saniasiaya[2]

Naczyniak wewnątrzmięśniowy u dziecka

Intramuscular haemangioma in a child

[1] Department of Otorhinolaryngology, Hospital Selayang, Lebuhraya Selayang-Kepong, 68100 Selayang, Selangor Darul Ehsan, Malaysia
[2] Department of Otorhinolaryngology, Faculty of Medicine, Universiti Malaya, Jalan Universiti, 50603 Kuala Lumpur, Malaysia
Adres do korespondencji: Jeyasakthy Saniasiaya, Otorhinolaryngologist and Head and Neck Surgeon, Department of Otorhinolaryngology, Faculty of Medicine, Universiti Malaya, Jalan Universiti, 50603 Kuala Lumpur, Malaysia, tel.: +6012-6841223, fax: +6075564333, e-mail: shakthy_18@yahoo.com

ORCID iDs
1. Izzah Akashah https://orcid.org/0000-0002-3402-3150
2. Jeyasakthy Saniasiaya https://orcid.org/0000-0003-1974-4379

Streszczenie

Cel pracy: Przedstawienie problemu diagnostycznego związanego z rozpoznaniem naczyniaka wewnątrzmięśniowego mięśnia żwacza u dziecka. Naczyniak wewnątrzmięśniowy jest nietypowym rodzajem naczyniaka i często zostaje przeoczony lub błędnie rozpoznany przez pracowników służby zdrowia. **Opis przypadku:** W pracy omówiono złożony przypadek 9-letniej dziewczynki skierowanej na oddział otorynolaryngologii z powodu jednostronnego obrzęku okolicy przedusznej, początkowo leczonego jako zapalenie ślinianki przyusznej, a ostatecznie zdiagnozowanego – na podstawie badania obrazowego i wyniku biopsji – jako naczyniak wewnątrzmięśniowy mięśnia żwacza. W ciągu niemal roku poprzedzającego ustalenie rozpoznania pacjentka przeszła kilka kursów antybiotykoterapii i aspirację cienkoigłową, w której uzyskano niejednoznaczne wyniki. **Wnioski:** Autorzy pracy pragną zwrócić uwagę na wczesne przeprowadzenie diagnostyki obrazowej i biopsji pod kontrolą badania obrazowego, zwłaszcza u dzieci, w celu uniknięcia opóźnień w diagnostyce i szybkiego wdrożenia leczenia.

Słowa kluczowe: naczyniak, mięsień żwacz, dzieci, łagodny guz naczyniowy

Abstract

Aim of the study: To outline the challenge faced in diagnosing intramuscular haemangioma of the masseter in a child. Intramuscular haemangioma is an unusual type of haemangioma and is oftentimes overlooked or misdiagnosed by health care professionals. **Case report:** Herein, we discuss a complex case of a 9-year-old girl referred to otorhinolaryngology department for unilateral preauricular swelling, initially treated as parotitis and finally diagnosed as intramuscular masseter haemangioma following imaging and biopsy. However, the patient had to complete several courses of antibiotics and fine needle aspiration, which was inconclusive, for almost a year prior to attaining diagnosis. **Conclusion:** We would like to highlight early imaging and image-guided biopsy especially in children to avoid delay in diagnosis and prompt management.

Keywords: haemangioma, masseter muscle, children, benign vascular tumour

INTRODUCTION

Haemangioma is a not an unusual tumour among newborns, accounting for 7% of all benign neoplasms, and usually arises from endothelial cells, which line blood vessels of the skin and mucosal membranes[1]. Intramuscular haemangioma (IMH) of the masseter occurs mostly among childhood age group. However, it is usually only diagnosed following unexpected enlargement of the mass, pain, or cosmetic asymmetry, which often occurs later in life. Given the rarity of this entity, obtaining accurate diagnosis at presentation is usually not feasible especially by physical examinations only. It is noteworthy that above 90% of IMHs are misdiagnosed[2].

CASE REPORT

A healthy 9-year-old girl presented with right-sided preauricular mass for the past eight months. According to her mother, the patient had multiple episodes whereby the swelling initially increased in size, accompanied by pain which subsided with oral antibiotics prescribed by various general practitioners. Additionally, the patient underwent repeated fine-needle aspiration, which was inconclusive. However, the child developed limited mouth opening concurrently with persistent preauricular swelling for the past 2 months. Apart from that, there was no fever, obstructive or constitutional symptoms. No nasal or aural symptoms were also documented.

Upon examination, the child appeared comfortable. There was right preauricular mass, measuring 2 × 1 cm with ill-defined borders, non-tender, firm, not mobile with no external skin changes (Fig. 1). No other neck nodes were palpable. Mouth opening was 1 finger breadth with no medialisation seen. Oral cavity and oropharynx subsites were unremarkable. Systemic examination along with vital signs were also normal.

We proceeded with neck ultrasonography which revealed an ill-defined mass posterior to the right parotid gland with minimal colour Doppler signal demonstrated. This was then followed by computed tomography of the neck, which revealed a mass arising from the right masseter muscle, with no calcification within. However, diagnosis was still inconclusive which led to discussion with an interventional radiologist. Hence, ultrasound-guided biopsy was carried out which revealed cavernous haemangioma. The tissue taken from the biopsy composed of small fragments of muscle bundle and loose myxoid connective tissue containing multiple abnormal blood vessels, which was highlighted by CD31 (Fig. 2). The blood vessels seen were lined by flat endothelial cells with no cell atypia seen (Fig. 3). Simultaneously, cervical magnetic resonance imaging showed right masseter intramuscular mass measuring 2.0 × 1.2 × 2.2 cm, which was iso-intense on T1 weighted and hyper-intense on T2 weighted images (Fig. 4), suggestive of right masseteric muscle IMH. The patient was then referred to arteriovenous malformation (AVM) clinic and underwent sclerosing agent injection. The preauricular swelling resolved after 6-months of follow-up.

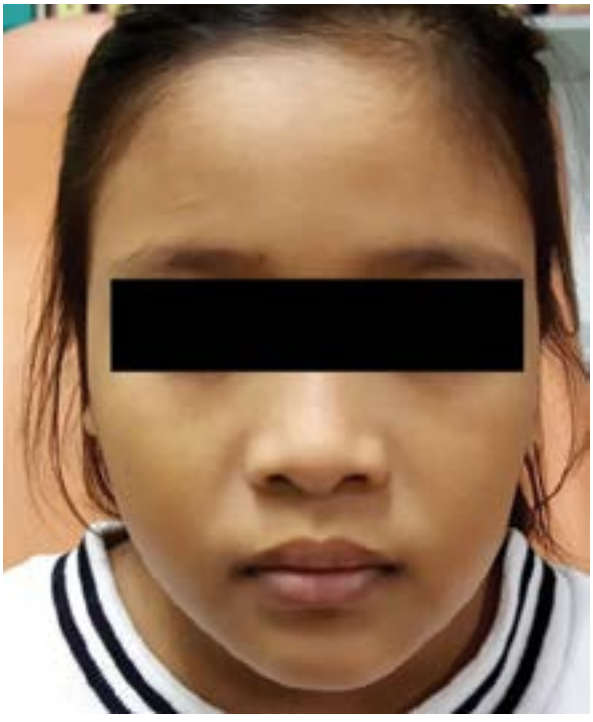

Fig. 1. Right preauricular mass measuring 2 × 1 cm with ill-defined borders

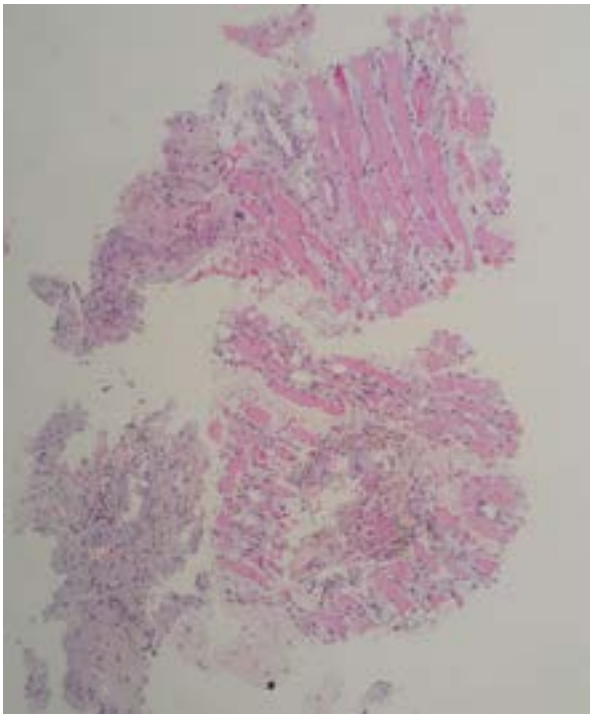

Fig. 2. H&E ×200. Fragments of skeletal muscle with abnormal blood vessels

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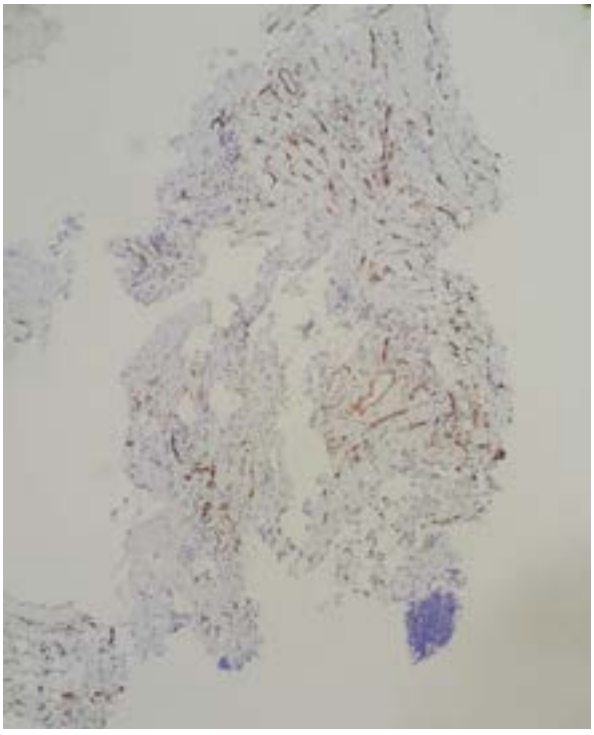

Fig. 3. CD31 ×200. Intramuscular and loose connestive tissue blood vessels with positive endothelial staining with CD31

DISCUSSION

IMH is an unusual type of vascular disease which involves skeletal muscle and accounts for only 0.8% of all haemangiomas. Nearly 10% to 15% of IMHs are found within the head and neck region, as in our patient. IMH of the head and neck has been reported to involve the masseter muscle (36%), the trapezius muscle (24%), periorbital muscles (12%), the sternocleidomastoid muscle (10%), the temporalis muscle, as well as the orbicularis oris muscle[2].

IMH is commonly found in children and young adults. Generally, IMH was shown to have equal sex distribution except for the involvement of the masseter muscle, which has shown male predominance[3] with a ratio of 3 to 1. However, our patient was a 9-year-old girl.

There are a few classifications for haemangioma in literature. In 1972, Allen and Enzinger histologically divided haemangioma into (1) capillary (<140 μm in diameter), (2) cavernous (>140 μm in diameter) or (3) mixed (includes both small and large vessels)[3]. Capillary haemangioma is predominantly cellular, thus contributing to its firm consistency as well as the presence of vague clinical signs to suggest its vascular nature, which may lead to delay in diagnosis. Cavernous haemangioma, on the other hand, generally presents with a longer history of symptoms, tends to be larger and painful. It is predominantly seen in the lower extremity with only 19% reported in the head and neck region. Mixed type is both clinically and histologically similar to the cavernous type. Our patient fits clearly clinically and histologically to the cavernous type. IMH of the masseter muscle frequently presents with facial mass associated with pain in 30–60% of cases[4], as in our patient.

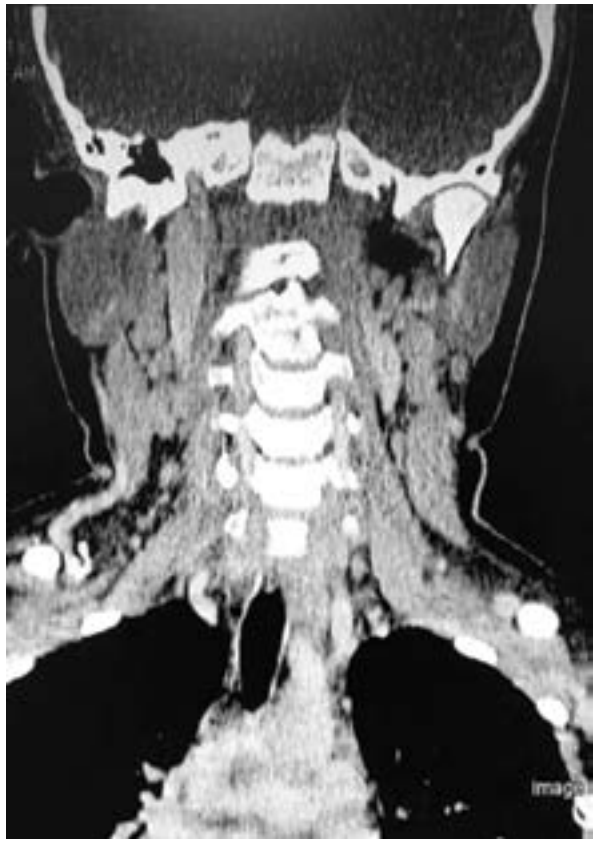

Fig. 4. Cervical MRI showed right masseter intramuscular mass measuring 2.0 × 1.2 × 2.2 cm which was iso-intense on T1 weighted image

The diagnosis of IMH can be established by the presence of phleboliths in radiographs, contrast-enhanced CT or CT in combination with sialography and angiography[4]. These tumours are however oftentimes confused with other diagnosis such as salivary gland tumours, cysts, lymphangiomas, rhabdomyosarcomas, masseteric hypertrophy as well as schwannomas[3]. FNAC is often inconclusive following blood tinged aspirate obtained[3], which unfortunately was also the case of our patient.

Magnetic resonance imaging (MRI) demonstrates superior ability to detect as well as provide better delineation of the extent of IMH compared to computed tomography following its multiplanar capabilities and the distinct contrast between normal muscle and IMH[3].

IMH ought to be managed according to the tumour location and extension, tumour growth rate, anatomical accessibility, patient age and cosmetic considerations[3]. Notable treatment for IMH ranges from steroids to injection of sclerosing agents, radiation therapy, and surgical excision[5], with surgical excision considered the current gold standard of treatment[3]. Surgical approaches include intraoral procedure, superficial parotidectomy as well as preauricular incision with lateral skin flap and parotid gland preservation[5].

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CONCLUSION

IMH ought to be considered as one of the differentials in a facial mass with soft tissue density encountered in young adults. MRI is the superior imaging as compared to computed tomography in diagnosing IMH, and though management should be individualised, complete surgical excision remains the optimal treatment of choice for IMH.

Conflict of interest

Authors do not report any financial or personal connections with other persons or institutions, that might claim authorship rights to this publication.

Piśmiennictwo

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