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Inflammatory Myofibroblastic Tumour of the Buccal Mucosa in a Child: Case Report and Review of Paediatric Oral Cases

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ABSTRACT

Inflammatory myofibroblastic tumour (IMT) is an uncommon mesenchymal lesion of intermediate biological potential, characterized by a proliferation of myofibroblastic spindle cells accompanied by a prominent inflammatory infiltrate of lymphocytes, plasma cells, and eosinophils [1, 2]. While reported in various anatomical sites, its occurrence in the oral cavity is exceedingly rare. We report a case of IMT in the right buccal mucosa of a 9-year-old female patient with cerebral palsy. The clinical, radiological, and histopathological findings are detailed, underscoring the diagnostic challenges this lesion presents. The case highlights the importance of histopathological confirmation for accurate diagnosis and discusses treatment considerations, emphasizing conservative yet complete surgical excision as the primary management strategy. A review of the relevant literature on paediatric oral IMTs is provided to contextualize this case.

Keywords: *Inflammatory Myofibroblastic Tumour (IMT); Oral Cavity Neoplasms; Buccal Mucosa; Head and Neck Neoplasms; Myofibroblastic Proliferation; Rare Oral Tumor*

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1. INTRODUCTION

Inflammatory myofibroblastic tumour (IMT) is an infrequent growth of uncertain origin, characterized by myofibroblastic proliferation with variable inflammatory infiltration. It has been described by various names, including inflammatory pseudotumor, plasma cell granuloma, fibrous xanthoma, pseudosarcoma, and myxoid hamartoma, reflecting the historical uncertainty surrounding its biological nature [1].

First described in the lungs by Bunn in 1939, Umiker et al. later coined the term "IMT" due to its clinical, radiological, and histopathological similarity to malignant neoplasms [1, 2]. Although proposed aetiologies include reactive inflammatory processes, infections, autoimmune disorders, and neoplastic mechanisms, its exact cause remains unclear. However, cytogenetic evidence, specifically rearrangements of the anaplastic lymphoma kinase (ALK) gene on chromosome 2p23, and well-documented reports of local recurrence and rare metastasis support its classification as a true neoplasm with intermediate biological potential [1, 2, 11].

IMTs most commonly occur in the lungs, liver, and gastrointestinal tract (GIT). In the head and neck region, which accounts for approximately 14% of all cases, lesions have been reported in the epiglottis, parapharyngeal space, maxillary sinus, submandibular region, and oral cavity [3, 4]. Oral IMTs are exceedingly rare and are often clinically and radiologically

mistaken for malignancy, making diagnosis reliant primarily on histopathological evaluation [4-6]. Clinically, IMTs usually present as painless, firm swellings but may vary with location [7]. Imaging modalities such as computed tomography (CT) and magnetic resonance imaging (MRI) often reveal features that mimic malignant lesions, such as ill-defined borders and tissue infiltration [7].

Treatment strategies include surgical excision, corticosteroids, radiotherapy, chemotherapy, and, more recently, CO_2 laser excision [7]. The mainstay of treatment for localized disease is complete surgical excision with clear margins.

2. CASE REPORT

A 9-year-old female patient with cerebral palsy presented to our department with a chief complaint of swelling on the right side of her face, which had gradually increased in size over the past 6 months. Extraoral examination revealed a diffuse, firm, non-tender swelling of approximately 3 x 2 cm in size, with normal overlying skin. A slight drooping of the right oral commissure was observed, with no signs of facial nerve weakness or paraesthesia.

Intraoral examination revealed a solitary, oval-shaped, well-defined, mobile swelling in the right buccal mucosa, measuring roughly 2.5 x 1.5 cm. The overlying mucosa was intact and appeared normal in colour. The lesion was firm on palpation and non-tender. Ultrasonography (USG) revealed a well-defined heteroechoic solid lesion (2.5 × 1.5 cm) located in the subcutaneous and intramuscular plane. Colour Doppler imaging showed internal vascularity, which was suggestive of a neoplastic aetiology. Fine needle aspiration cytology (FNAC) yielded blood-mixed material. The smears showed fragments of fibroconnective tissue and numerous red blood cells, but were devoid of any malignant epithelial component, ruling out carcinoma.

Surgical excision of the lesion was performed under general anesthesia, and the excised mass was submitted for histopathological examination. Microscopic analysis of Haematoxylin and Eosin (H&E) stained sections revealed a proliferation of bland to mildly atypical spindle cells (myofibroblasts) arranged in irregular fascicles. The stroma was densely infiltrated by a mixed inflammatory cell population comprising lymphocytes, plasma cells, neutrophils, and histiocytes. Focal myxoid areas were also observed. There was no evidence of significant nuclear atypia or mitotic activity. Based on the correlation of clinical, radiological, and these characteristic histopathological findings, a definitive diagnosis of inflammatory myofibroblastic tumour (IMT) was established.

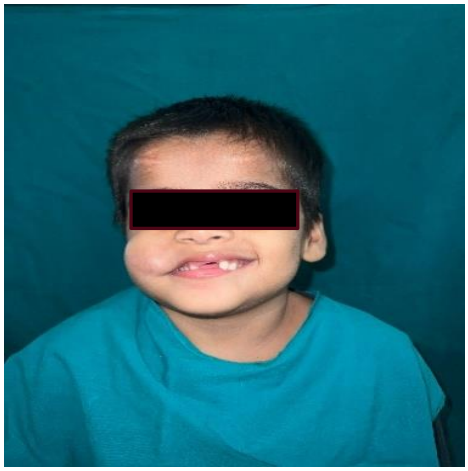

Figure 1. Extraoral photograph showing diffuse swelling on the right side of the face in a 9-year-old female patient diagnosed with an inflammatory myofibroblastic tumor.

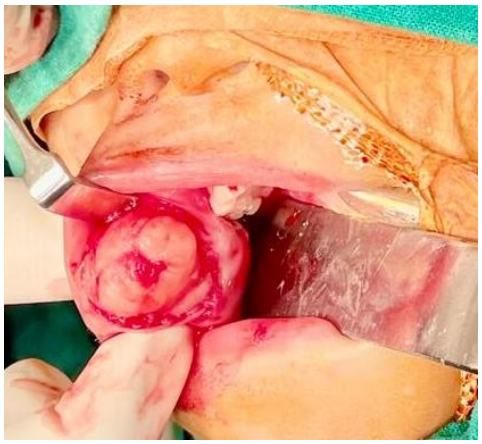

Figure 2. Intraoperative view showing surgical exposure of the well-encapsulated lesion in the right buccal mucosa prior to excision.

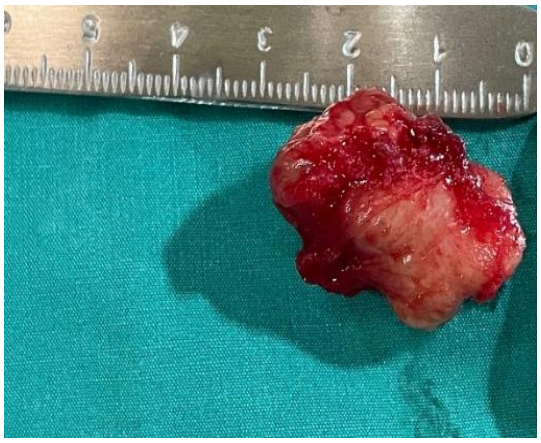

Figure 3. Gross specimen of the excised lesion from the right buccal mucosa, measuring approximately 2.5 × 1.5 cm.

Table 1: Reported paediatric cases of inflammatory myofibroblastic tumour in the oral and maxillofacial region

Author & Year	Country	Age/Gender	Site of Lesion	Size	Symptoms	Investigations	Treatment	Follow-up	Recurrence
Lourenço SV et al., 2007 [20]	Brazil	14/M	Tongue	2 cm	Painful mass	Incisional biopsy, IHC	Nil	5 years	No
Rao K, 2021 [21]	USA	41 weeks/F	Tongue	–	–	MRI, Frozen section, IHC, PET	Surgical debulking	1 year	No
Caporalini C et al., 2018 [22]	Italy	10 mo/M	Tongue	5 cm	Drooling, dyspnea, macroglossia	MRI, Biopsy, IHC	Prednisone, Crizotinib, Excision	4 months	No
Ribeiro AC et al., 2001 [23]	USA	6/F	Cheek	5 cm	Painless swelling, fever	Radiographs, CT, MRI	Corticosteroids	–	–
Rautava J et al., 2013 [24]	Finland	11/F	Maxilla	–	Painless swelling, fever	OPG, Histopathology	Enucleation, curettage	3 years	No
Tateishi Y et al., 2016 [25]	Japan	11/F	Mandible	–	Swelling	CT, Biopsy	Surgical excision	2 years	No
Patil PH et	India	8/F	Mandible	–	Painless	Radiographs,	Surgical	1 year	No

Author & Year	Country	Age/Gender	Site of Lesion	Size	Symptoms	Investigations	Treatment	Follow-up	Recurrence
al., 2022 [26]					swelling	Biopsy	excision		
Satomi T et al., 2010 [27]	Japan	14/M	Mandible	–	Pain and swelling	CT, MRI, Biopsy	Segmental resection	2 years	No
Stringer DE et al., 2014 [28]	USA	16/M	Mandible	–	Swelling	Imaging, Histopathology	Excision	1 year	No
Lai V et al., 2014 [29]	Hong Kong	14/F	Nasal cavity	–	Nasal obstruction	MRI, Biopsy	Excision	1 year	No
Rachel L et al., 2015 [30]	USA	14/F	Nasal cavity	–	Nasal obstruction	Imaging, Biopsy	Excision	6 months	No
Fawzy WM et al., 2018 [31]	Egypt	5/M	Nasal cavity	–	Obstruction, swelling	CT, Biopsy	Excision	1 year	No
Murthy AS et al., 2017 [32]	India	5/F	Maxillary sinus	–	Swelling, fever	CT, MRI, Biopsy	Excision	6 months	No
Murai A et al., 2012 [33]	Japan	2/F	Maxillary sinus	–	Swelling	CT, MRI, Histopathology	Excision	1 year	No
Biswas R et al., 2019 [34]	India	4/F	Maxillary sinus	–	Swelling	CT, Biopsy	Excision	1 year	No

3. DISCUSSION

IMT is a rare lesion that predominantly affects children and young adults, commonly arising in the lungs, soft tissues, and abdominal organs, with the oral cavity being an unusual location [8]. The first series of oral IMTs was reported by Liston et al. in 1981 in three paediatric cases [9]. These lesions were historically misclassified under various terminologies until the advent of immunohistochemical (IHC) studies, which confirmed their myofibroblastic origin (positive for vimentin, smooth muscle actin (SMA), and often desmin) and helped distinguish them from true sarcomas [10]. The present case, located in the buccal mucosa of a child, aligns with the demographic and clinical profile of oral IMTs. Our review of 16 paediatric cases (including the present case), as detailed in Table 1, indicates a wide age range at presentation (from 10 months to 16 years) with no distinct gender predilection. The tongue, mandible, cheek, maxilla, nasal cavity, and maxillary sinus are the most common sites [20-34]. The clinical presentation is variable, ranging from an asymptomatic, painless swelling (as in our case) to obstructive symptoms such as nasal blockage and dyspnoea. Systemic symptoms like fever and weight loss are occasionally reported and are thought to be related to cytokine release by the inflammatory cells within the tumour [23, 24]. Imaging findings, as seen in our patient's USG, are often non-specific and can suggest a malignant neoplasm due to features like tissue infiltration and vascularity, thereby necessitating histopathological confirmation for a definitive diagnosis [7].

The histopathological diagnosis of IMT can be challenging due to its overlap with several other spindle cell lesions. The main histological differential diagnoses include nodular fasciitis, solitary fibrous tumour, fibrous histiocytoma, calcifying fibrous tumour, myofibroma, fibrosarcoma, follicular dendritic cell tumor, and leiomyosarcoma [11]. IHC plays a crucial role in this differentiation. IMTs are typically positive for vimentin and SMA, and approximately 50-60% of cases show positivity for ALK-1 protein, which correlates with ALK gene rearrangements [11, 12]. It is noteworthy that oral IMTs are more frequently ALK-negative compared to those in other anatomical sites, which may suggest a distinct biological behaviour or aetiology in the oral cavity [12].

The World Health Organization (WHO) classifies IMTs as lesions of intermediate biological potential due to their capacity for local recurrence and rare metastatic potential [11]. The overall recurrence rate is estimated to be around 25%,

particularly in cases with positive margins, ALK-negativity, or lesions located in anatomically complex sites where complete excision is difficult [16]. However, the risk of metastasis is very low (<5%) [18].

The primary and most effective treatment for localized IMT is complete surgical excision with clear margins [13, 14]. In the paediatric population, a conservative yet complete resection is favoured to minimize functional and cosmetic morbidity [17]. For unresectable, recurrent, or multifocal diseases, other modalities have been employed with varying success. Non-steroidal anti-inflammatory drugs (NSAIDs) and corticosteroids have led to tumour regression in some cases, supporting an inflammatory component to its aetiology [15]. In ALK-positive advanced IMTs, targeted therapy with ALK inhibitors like Crizotinib has emerged as a promising treatment option, as demonstrated in one of the reviewed cases [22]. Radiotherapy and chemotherapy are generally reserved for aggressive, unresectable cases where other options have failed [7].

The prognosis for oral IMT in children is generally excellent. As evidenced by the literature review, most patients treated with excision were disease-free at follow-up periods ranging from 6 months to 5 years. Rigorous long-term follow-up is recommended to detect recurrences early, with most recurrences occurring within the first two years [18, 19]. A suggested follow-up protocol includes clinical examinations every 1–2 months for the first year, every 3–4 months for the second year, and every 6 months thereafter for at least 5 years [19].

4. CONCLUSION

IMT represents a rare lesion with variable clinical presentation and biological behaviour. Although most oral IMTs in children follow a benign clinical course, their potential for local recurrence mandates accurate diagnosis and appropriate management. Histopathological examination, supplemented by IHC, is the cornerstone of diagnosis. Complete surgical excision with clear margins is the treatment of choice, offering an excellent prognosis. Long-term follow-up is essential to ensure early detection and management of any recurrence. This case adds to the limited literature on oral IMTs in the paediatric population and underscores the importance of considering this entity in the differential diagnosis of oral spindle cell lesions.

REFERENCES

[1] Poh CF, Priddy RW, Dahlman DM. Intramandibular inflammatory myofibroblastic tumour: A true neoplasm or reactive lesion? Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2005; 100:460-6.
[2] Volker HU, Scheich M, Holler S, Strobel P, Hagen R, Hermenlink HK, et al. Differential diagnosis of laryngeal spindle cell carcinoma and inflammatory myofibroblastic tumour: Report of two cases with similar morphology. Diagn Pathol. 2007; 2:1-7.
[3] Margaret S, Silloo BK, Gnepp DR. Nonsquamous pathology of the larynx, hypopharynx, and trachea. In: Gnepp DR, editor. Diagnostic surgical pathology of the head and neck. 4th ed. New York: W.B. Saunders Company; 2001. p. 287-8.
[4] Oh JM, Yim JH, Joon BW, Choi BJ, Lee DW, Kwon YD. Inflammatory pseudotumour in the mandible. J Craniofac Surg. 2008; 19:1552-3.
[5] Kujima M, Nakamura S, Itoh H, Suchi T, Masawa N. Inflammatory pseudotumour of the submandibular gland: Report of a case presenting with autoimmune disease-like manifestation. Arch Pathol Lab Med. 2001; 125:1095-7.
[6] Montgomery E, Speight PM, Fisher C. Myofibromas presenting in the oral cavity: A series of 9 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2000; 89:343-8.
[7] Al-Sindi KA, Al-Shehabi MH, Al-Khalifa SA. Inflammatory myofibroblastic tumour of paranasal sinuses. Saudi Med J. 2007; 28:623-7.
[8] Hammas N, Chbani L, Rami M, Boubbou M, Benmiloud S, Bouabdellah Y, et al. A rare tumor of the lung: inflammatory myofibroblastic tumor. Diagn Pathol. 2012; 7:83.
[9] Liston SL, Dehner LP, Jarvis CW, Pitzele C, Huseby TL. Inflammatory pseudotumors in the buccal tissues of children. Oral Surg Oral Med Oral Pathol. 1981; 51:287-91.
[10] Deshingkar SA, Tupkari JV, Barpande SR. Inflammatory myofibroblastic tumour of the maxilla. J Oral Maxillofac Pathol. 2007; 11:76-9.
[11] Binmadi NO, Packman H, Papadimitriou JC, Scheper M. Oral inflammatory myofibroblastic tumor: Case report and review of literature. Open Dent J. 2011; 5:66-70.
[12] Rautava J, Soukka T, Peltonen E, Nurmenniemi P, Kallajoki M, Syrjänen S, et al. unusual case of inflammatory myofibroblastic tumor in the maxilla. Case Rep Dent. 2013; 2013:876503.
[13] Gasparotti R, Zanetti D, Bolzoni A, Gamba P, Morassi ML, Ungari M. Inflammatory myofibroblastic tumor of the temporal bone. Am J Neuroradiol. 2003;24(10):2092-6.

[14] Coffin CM, Hornick JL, Fletcher CD. Inflammatory myofibroblastic tumor: comparison of clinicopathologic, histologic, and immunohistochemical features including ALK expression in atypical and aggressive cases. Am J Surg Pathol. 2007;31(4):509-20.
[15] Naveen J, Sonalika WG, Prabhu S, Gopalakrishnan K. Inflammatory pseudotumor of maxillary sinus: mimicking as an aggressive malignancy. J Oral Maxillofac Pathol. 2011;15(3):344-5.
[16] Browne M, Abramson LP, Chou PM, Acton R, Holinger LD, Reynolds M. Inflammatory myofibroblastic tumor (Inflammatory pseudotumor) of the neck infiltrating the Trachea. J Pediatr Surg. 2004;39(10): e1-4.
[17] Souid AK, Ziemba MC, Dubansky AS, et al. Inflammatory myofibroblastic tumor in children. Cancer. 1993;72(6):2042-8.
[18] Donner LR, Trompler RA, White RR. Progression of inflammatory myofibroblastic tumor (inflammatory pseudotumor) or soft tissue into sarcoma after several recurrences. Hum Pathol. 1996;27(10):1095-8.
[19] Ramachandra S, Hollowood K, Bisceglia M, Fletcher CDM. Inflammatory pseudotumor of the soft tissues: a clinicopathological and immunohistochemical analysis of 18 cases. Histopathology. 1995;27(4):313-23.
[20] Lourenço SV, et al. Inflammatory myofibroblastic tumor of the tongue in a child: case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2007;103(5):574-8.
[21] Rao K. Inflammatory myofibroblastic tumor of tongue in an infant: a rare case. J Oral Maxillofac Pathol. 2021;25(1):132-5.
[22] Caporalini C, et al. Inflammatory myofibroblastic tumor of the tongue in a 10-month-old child. Int J Pediatr Otorhinolaryngol. 2018; 105:151-4.
[23] Ribeiro AC, et al. Inflammatory myofibroblastic tumor of the cheek in a child. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001;92(3):307-10.
[24] Rautava J, et al. Unusual case of inflammatory myofibroblastic tumor in the maxilla. Case Rep Dent. 2013; 2013:876503.
[25] Tateishi Y, et al. Pediatric inflammatory myofibroblastic tumor of the mandible: case report. Oral Surg Oral Med Oral Pathol Oral Radiol. 2016;122(4): e109-13.
[26] Patil PH, et al. Mandibular inflammatory myofibroblastic tumor in an 8-year-old: case report. J Oral Maxillofac Surg. 2022;80(2):312-6.
[27] Satomi T, et al. Mandibular IMT with aggressive features in a 14-year-old. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2010;110(5): e45-9.
[28] Stringer DE, et al. Inflammatory myofibroblastic tumor of the mandible in an adolescent. J Craniofac Surg. 2014;25(6):2232-5.
[29] Lai V, et al. Pediatric inflammatory myofibroblastic tumor of the nasal cavity: case report. Head Neck Pathol. 2014;8(2):205-8.
[30] Rachel L, et al. Pediatric nasal cavity inflammatory myofibroblastic tumor: case report. Int J Pediatr Otorhinolaryngol. 2015;79(4):610-3.
[31] Fawzy WM, et al. Inflammatory myofibroblastic tumor of the nasal cavity in a child. Egypt J Ear Nose Throat Allied Sci. 2018;19(1):45-8.
[32] Murthy AS, et al. Inflammatory myofibroblastic tumor of the maxillary sinus in a child. J Pediatr Neurosci. 2017;12(1):90-2.
[33] Murai A, et al. Inflammatory myofibroblastic tumor of the maxillary sinus in a child. Auris Nasus Larynx. 2012;39(5):527-30.
[34] Biswas R, et al. Inflammatory myofibroblastic tumor of maxillary sinus in a pediatric patient: A case report. Oncol J India. 2019;3(1):25-7.