Case Report

Congenital Infiltrating Lingual Chondromyomyxoid Lipoma in a Young Child: Case Report and Literature Review

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Abstract

Oral cavity lipoma is uncommon and the tongue is an unusual site in paediatric age. Most oral cavity lipomas arise from the buccal mucosal or floor of the mouth, whereas tongue involvement may be part of inherited disease syndromes such as neurofibromatosis, multiple familial lipomatosus, and Gardner syndrome or a sequela of trauma. We report case of congenital infiltrating tongue chondromyomyxoid lipoma in 21-month-old male child. This case illustrates a single lesion with multiple histological variants which may be misdiagnosed as a harmatomatous lesion.

Keywords: Tongue, Chondromyomyxoid Lipoma, Oral cavity, Child

INTRODUCTION

Lipoma of the tongue is an uncommon lesion and the first case was documented by Barling in 1858 though, ten years earlier in 1848, Roux described an oral cavity lesion as "yellow epuli".[1, 2] Since then, several cases of lipoma have been documented in the oral cavity. Lipoma is the most common benign mesenchymal tumour of fatty tissue, and occurs in body parts where adipose tissue is found.[2] However, lipoma is uncommon in the maxillofacial region and oral cavity lipoma accounts for 0.1% to 5% of all benign lesions affecting the oral cavity while tongue lipoma account for 0.3% of all tongue tumours.[2, 3, 4] It often arises from the buccal mucosa and rarely the floor of the mouth, gingiva and retromolar region whereas, extra oral lipoma most commonly involves the extremities. Occurrence of lipoma in the tongue is uncommon particularly as the tongue lacks fatty tissue, though, a handful of tongue lipoma cases have been reported in English literature in adults with an average age of 40years with no sex predilection unlike extra oral lipoma that has a female predilection.[3,5.] We report a case of congenital infiltrating chondromyomyxoid lipoma of the tongue in twenty-one months-old full term male child who had no gross congenital anomalies and or history of trauma.

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CASE SUMMARY

A twenty-one-month-old full term male child was brought to the maxillofacial clinic of our hospital by the mother who complained of a progressively increasing non-tender swelling on the anterior third of the tongue since birth. Swelling is not associated with difficulty in feeding but prevents full closure of the mouth. No other swelling or obvious anomaly on the child. He is the only child of the mother and his pregnancy was uneventful. He had all the childhood Immunizations and his developmental milestones is normal. Clinical examination showed a well child with an anterior tongue swelling which measured 6x6cm. The swelling was on the dorsal surface of the tongue, and was non-tender and non-mobile. The tongue moved freely and was not fixed to the floor of the mouth. There were no palpable lymph nodes or masses anywhere else on his body.

There were no obvious physical abnormalities. Investigations done included a full blood count of Hb 11.2g/dl, PCV 35%, WBC $8.1x10^9/mm^3$, and platelet count of 439. Electrolytes and urea were unremarkable, serology for HIV, HCV and HBV was negative and the clotting profile was within normal limits. He had surgical excision of the anterior tongue mass under general anaesthesia though, infiltrating strands of the mass into the intrinsic tongue muscle could not be dissected. He recovered fully post-operatively and was placed on antibiotics and analgesics.

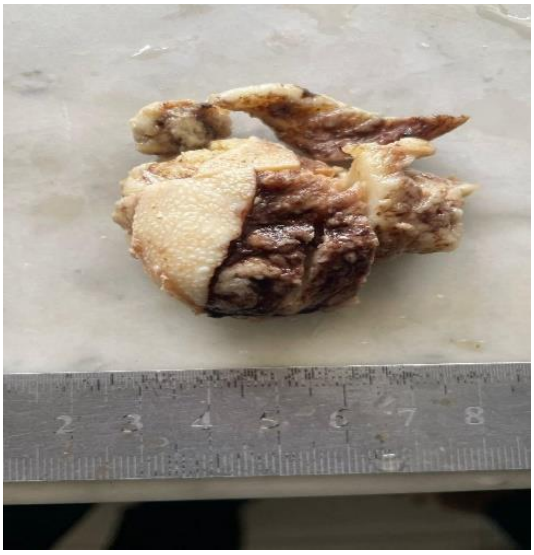

Figure 1: Gross photograph of the lingual mass

The three months follow-up at the clinic was uneventful. The surgical specimen sent to the pathology laboratory was fixed in 10% formalin and consisted of a soft to firm 6x6x4cm grey-yellow irregular tissue that weighed 7g (figure 1). It was processed in paraffin wax and then stained with haematoxylin and eosin. Histology revealed lobules of mature adipocytes infiltrating into the tongue

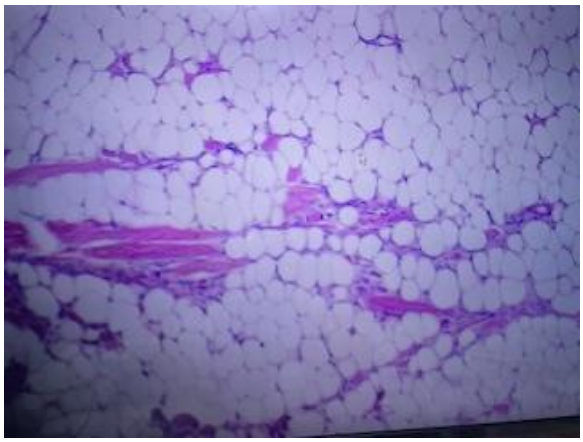

Figure 2: Photomicrograph showing lobules of mature adipocytes infiltrating into the tongue muscle bundles (H&E x200)

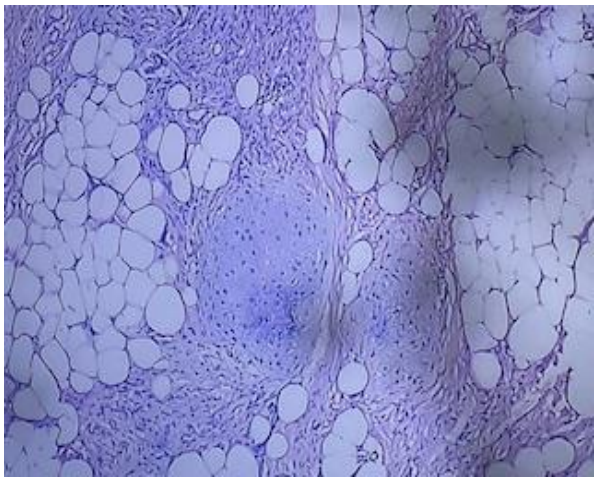

Fig 3: Photomicrograph showing chorndroid tissue admixed with mature adipose lobules (H&E x200)

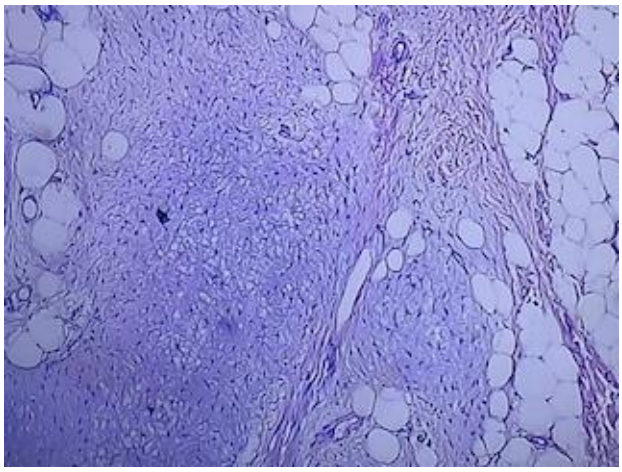

Figure 4: Photomicrograph showing myxoid admixed with mature adipose tissue (H&E x200)

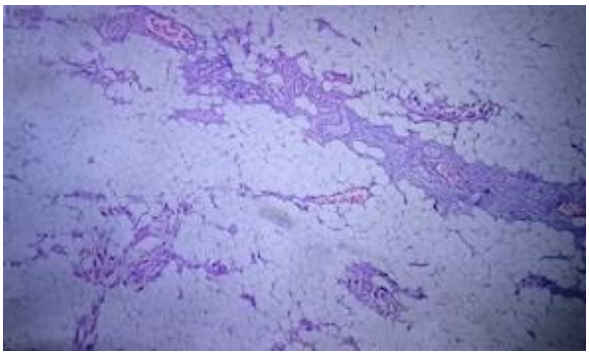

Figure 5: Photomicrograph showing dilated vessels within fibrous stroma (H&E x200)

muscle bundles (figure 2). Focal areas within the lesion showed chondroid, myxoid and angiomatous changes within the adipocytes lobules, and fibrous stroma (figures 3, 4 &5). This was diagnosed as congenital infiltrating chondromyomyxoid lipoma.

DISCUSSION

Lipoma is the most common benign mesenchymal tumour comprised of mature adipocytes indistinguishable from normal fatty tissue. It occurs as a painless asymptomatic slow-growing often yellowish soft tissue mass with a predilection for the extremities. The age of affected individuals ranges from 2 years to 87 years with a mean age of 40 years at presentation, and females are more affected compared to males.[3,6] Lipoma of the maxillofacial region is uncommon, even though head and neck lipoma accounts for approximately 15% to 20% of all lipoma cases.[3, 4, 7] Oral cavity lipoma represents 0.1% to 5% of all benign oral lesions and single lesions averages 2.5cm in diameter.[6, 8]

Lesions in the oral cavity often involve the buccal mucosa, floor of the mouth, gingiva, and retromolar region while the tongue is an unusual site.[3, 9,10] Lipoma in general may occur as single or multiple lesions and may be located superficially or within deep tissues with a female predilection. However, there is no gender preference in intramural lipoma. Multiple lesions are often associated with inherited diseases such as multiple familial lipomatosis, neurofibromatosis Gardner syndrome and chromosomal anomalies.[2, 4,11] Solitary cases have been associated with trauma though the pathogenetic mechanism is not clearly defined.[12] This child had the lingual lesion since birth with no history of birth trauma or obvious congenital anomalies or abnormalities on clinical examination.

Congenital lipoma as seen in this child is rare and the few cases documented in literature occurred shortly after birth with no other congenital anomaly.[11] Tongue lipoma may be associated with macroglossia, dental anarchy and malocclusion. This child had macroglossia and malocclusion significant enough to give the mother concern.

Common lesions that may involve the tongue include harmatoma, lymphangioma, haemangioma, neurofibroma and even a thyroglossal cyst at the base of the tongue. These lesions are particularly common amongst paediatric patients and manifest early as either congenital malformations or part of syndromes diseases. Harmatomatous lesions are histologically characterized by haphazardly arranged well differentiated or mature tissues native to the site of involvement while haemangioma and lymphangioma are composed of varying sized capillaries and lymphatic spaces. Cystic lesions that may occur in the tongue are characterized morphologically by the type of lining epithelium. The only abnormality seen in this boy's tongue was the lobules of mature adipocytes with chondroid, myxoid and focal angiomatous differentiation splitting the tongue musculature.

Lipomas have been classified into histological variants such as chondroid lipoma, spindle cell lipoma, osteolipoma, angiolipoma, fibrolipoma, myolipoma, myxoid lipoma, atypical lipoma, pleomorphic lipoma, and intramuscular (infiltrating) lipoma based on the constituent supportive tissues seen microscopically.[13, 14] This case had histological areas of chondroid, myxoid and angiomatous differentiation with mature adipocytes lobules infiltrating into the muscle tissue. This is the first report of a combination of many histological variants of lipoma in one lesion to the best of our knowledge.

Definitive diagnosis is by tissue excision for histological examination though the fine needle aspiration cytology technique also has significant diagnostic utility and is cheaper and less invasive. Supportive investigations such as radiological examination and magnetic resonance imaging are quite useful in identifying hypodense lesions of lipoma while computerized tomography and ultrasound scanning have limited utility.[4, 9]

The treatment of choice is surgical excision with free resection margins. However, the lesion was incompletely excised in this case due to infiltration of the muscles of the tongue and may recur. High recurrence is associated with infiltrating lipoma variants.[5 ,15 ,16] At present, there were no complaints during the 3months follow- up at the clinic however, long term follow- up is advocated. This case illustrates a commonly overlooked benign tumour that occurs in an uncommon site in an unusual paediatric age group.

REFERENCES

1. Junior OC, de Aguiar EC, Sartori JHF, Lima F de O. Spindle cell lipoma of the tongue: A case report of unusual occurrence. J Oral Maxillofac Pathol. 2013; 17:148.
2. Srinivasan K, Hariharan N, Parthiban P, Shyamala R. Lipoma of the tongue: A rare site for a common tumour. Indian J Otolaryngol Head Neck Surg. 2007;59::83-84.
3. Fregnani ER, Pires FR, Falzoni R, Lopes MA, Vargas PA.

Lipomas of the oral cavity: Clinical findings, histological classification and proliferative activity of 46 cases. Int J Oral Maxillofac Surg 2003;32: 49-53
4. Metgud RS, Kale AD. Lipoma of the tongue. J Oral Maxillofac Pathol 2004;8: 96-98
5. Sekar B, Augustine D, Murali S. Lipoma, a rare intraoral tumour- a case report with review of literature. Oral Maxillofac Pathol J 2011;2:174-177
6. Hatziotis JC. Lipoma of the oral cavity. Oral Surg Oral Med Oral Pathol 1971;31:511-524
7. Agarwal R, Kumar V, kaushal A, Singh RK. Intraoral lipoma: a rare clinical entity. BMJ Case Rep 2013: bcr2012007889
8. Roles DM. Lipoma of the tongue. Br J Oral Maxillofac Surg 1995;33:196-197
9. Magadum D, Sanadi A, Agrawal JM, Agrawal MS. Classic tongue lipoma: a common tumour at a rare site. BMJ Case Rep, 2013 (2013), Article bcr2012007987, 10.1136/bcr-2012/007987
10. Yang Y, Zuo H, Ding M, Ma F, Zhao L,Zhang J. A rare Lipoma of Tongue in a 4-Year old Child: A case Report with Literature Review. Ear, Nose &Throat Journal 2024
11. Navaneetham A, Rao A, Gandhi A, Jeevan CA. Lipoma involving the masticator space. Ann Maxillofac Surg 2011;1:93- 194.
12. Douglas Gneep R Diagnosticsurgical pathology of the head and neck WB Saunders
13. Anderson WJ, Doyle LD. Updates from the 2020 World Health Organisation Classification of Soft Tissue and Bone Tumours. Histopathology, 2021; 78 (5) : 644-657
14. Barisella M, Giannini L, Piazza C. From head and neck lipoma to liposarcoma: a wide spectrum of differential diagnoses and their therapeutic implications. Curr Opin Otolaryngol Head Neck Surg 2020; 28 :136-143
15. Pellissier A, Sawaf MH, Shabana AH. Infiltrating (intramuscular) benign lipoma of the head and neck. J Oral Maxillofac Surg 1991; 49:1231-1236
16. Betaineh AB, Mansour MJ, Abalkhail A. Oral infiltrating lipoma. Br J Oral Maxillofac Surg 1996;34:520-523