Case Report

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# Rare Solitary Neurofibroma of the Palate in a Pediatric Patient: A Case Report

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**KEYWORDS**

Neurofibroma, palate, solitary, pediatric, Neurofibromatosis type1

**ABSTRACT**

Neurofibroma is a benign nerve tumour originating from the peripheral nerve sheath, a variable mixture of Schwann cells, perineural-like cells, and fibroblasts. it may arise either as a solitary tumour or be a component of neurofibroma and von Recklinghausen's disease. In rare cases, multiple neurofibromas may develop without any associated syndrome. The solitary type seldom affects the head and neck areas, particularly the oral cavity. Although some cases have been documented on the buccal mucosa, floor of the mouth, lips, and gingiva, neurofibromas on the hard palate are extremely rare. These tumours usually present as firm, well-defined, and elastic masses without a capsule. We present a case report with a very rare solitary neurofibroma of the palate in a four months old girl.

## INTRODUCTION

Neurofibromas are benign tumours originating from peripheral nerve sheaths are uncommon in the head and neck area and consist of Schwann cells along with varying amounts of mature collagen [1]. Neurofibromas found in the oral cavity typically manifest as a nontender, submucosal and discrete mass [2]. Reportedly, the occurrence rate of solitary neurofibromas in the oral cavity is 6.5%, specifically for lesions not associated with neurofibromatosis - 1 [3]. In rare cases, these tumours can occur within the bone, presenting with a diverse range of radiographic appearances, from well-defined to poorly defined margins, and may appear unilocular or multilocular [4]. Generally, neurofibromas grow slowly, are well circumscribed, and are non-encapsulated in nature. They can affect any area of the oral cavity, but the tongue and buccal mucosa are the most commonly involved sites, followed by the lips and gingiva [5]. Here, we report the first documented case of a solitary palatal neurofibroma in a four-month-old patient.

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## CASE REPORT

A 4-month-old Sudanese baby girl was brought to Omdurman Military Hospital with a primary complaint of a swelling in her palate, which had been present for two months and became more noticeable 15 days prior. Oral examination revealed a solitary, oval, dome-shaped swelling in the midline of the hard palate and extended to the alveolar bone with a labial protrusion. The mass had a smooth surface, variable consistency, and was fluctuant on palpation. The overlying mucosa appeared normal with a reddish hue (Figure 1).

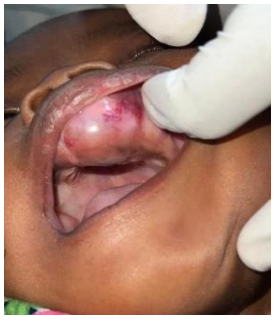

**Figure 1** Solitary dome shaped in the palate

A general physical examination showed no nodules or lymph nodes enlargement, and there was no family history of similar condition. A Computed Tomography (CT) scan with contrast revealed an expansile mass, measuring 3cm x 2cm, located at the midline of the maxilla and consisted of a soft tissue. The mass caused erosion and thinning of the maxillary bone, and the soft tissue showed faint enhancement, suggesting a benign bone tumour. no enlarged lymph nodes seen, with other facial bones appearing normal (Figure 2 and Figure 3). Surgical excision of the tumour was done under general anaesthesia (Figure 4), and two pieces of specimen were sent for histopathological analysis (Figure 5). Since the mass extended into the alveolar bone in the midline of the maxilla, during operation when we made exposure to the mass we found that the primary teeth specifically (A) in the right side and (A, B) in the left side were not involved within the tumour but were attached to it, so they were extracted with the tumour.

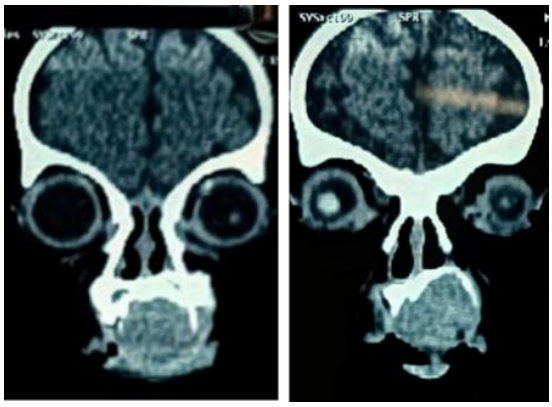

**Figure 2** CT scan with contrast (coronal view)

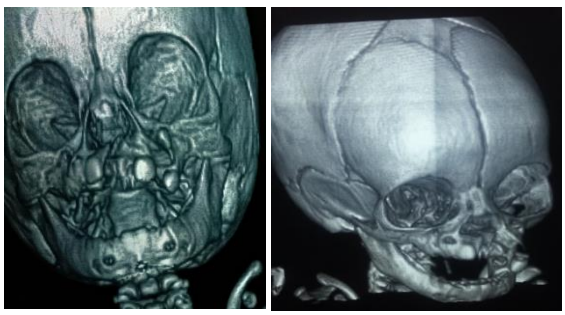

**Figure 3** CT scan with contrast (3D view)

Histopathological examination revealed small fragments containing benign spindle-shaped cells with wavy nuclei, without any evidence of malignancy. The final diagnosis was consistent with neurofibroma. After the mass was excised, a five-months follow-up showed no signs of recurrence.

**DISCUSSION**

Tumours of the peripheral nerve sheath are categorized as either benign or malignant [2]. Neurofibroma is a benign peripheral nerve sheath tumour that arises from Schwann cells and peripheral fibroblasts. Most solitary neurofibromas in the oral cavity appear in soft tissues, while intraosseous neurofibromas are rare [6]. Oral neurofibroma is a nonodontogenic tumour with differential diagnoses including schwannoma, lipoma, fibroma, neuroma, traumatic lesion or salivary gland tumours [7]. Common sites of extraosseous occurrence include the tongue (26%), buccal and labial mucosa (8%), and palate (8%). Floor of the mouth, gingiva, and alveolar ridge together constitute 2% of the cases. Intraosseously, the mandible, especially the posterior region, is more commonly affected than the maxillary area [8]. In the oral cavity, neurofibromas typically present as discrete, non-ulcerated nodules [9]. The mucosa can range in colour from normal to yellow or red [10]. These tumours are slow-growing, nodular, sessile, and mobile in character [11]. No particular racial or gender predisposition has been identified in oral neurofibroma [10], although Cherrick and Eversole, (1971) noted a higher occurrence among females [1, 12]. This tumour can occur in a wide range of ages, from 10 months to 70 years old, with an average age of 45 years old [13]. Solitary tumours are predominantly found in young adults, characterized by soft, slow growth, and painless lesions which range in size from small nodules to larger masses [9]. The direct cause of solitary neurofibromas is unknown [10].

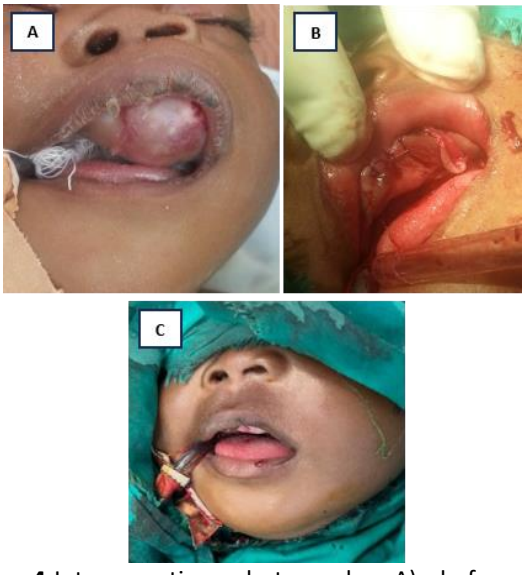


**Figure 4** Intraoperative photographs A) before excision the swelling and B) after excision the swelling (during suturing) and C) is after excision and suturing

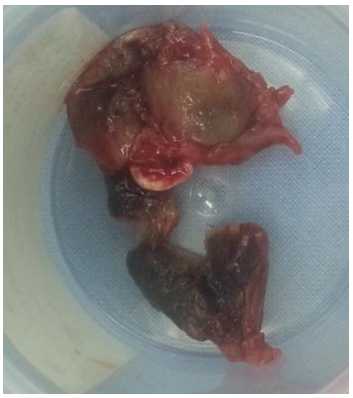

**Figure 5** Specimen of excisional biopsy

Radiographic characteristics become observed when neurofibromas develop centrally within the bone. These changes can appear as cortical erosion from adjacent soft tissue tumours or medullary resorption caused by intraosseous lesions. When located in the mandible, it may cause enlargement and branching of the mandibular canal, elevated bone density, concavity in the medial surface of the ramus, and expansion of the coronoid notch [14]. Histopathologically, neurofibromas are defined by the presence of spindle cells with wavy nuclei and are not encapsulated [9]. S-100 protein positivity is a significant factor distinguishing between schwannomas and neurofibromas, as neurofibromas exhibit positivity for S-100 protein, but less frequently than schwannomas [3]. This study compares the presented case with other lesions considered in the differential diagnosis of neurofibroma. pleomorphic adenomas, when they arise in the oral mucosa, the most frequently site is the mucosa over the anterior soft palate and posterior hard palate [17]. Fibromas, on the other hand, are most cases observed on the buccal mucosa near the occlusal plane but can also develop along the lateral border of the tongue and labial mucosa [17]. In the case of schwannoma, the tongue is the most frequent site in the oral cavity [18]. Schwannoma is also the least common oral tumour of nervous origin and is rarely seen in the palate. [19]. When it does occur in the palate, it is most often reported on the lateral aspect [20]. While schwannoma and neurofibroma can generally be differentiated by standard light microscopy, there can be considerable morphological overlap in some cases [18]. However, schwannoma and pleomorphic adenoma and fibroma most commonly appear between the ages of 30 and 50 years [17,22].

Mucoepidermoid carcinoma, when arises in minor salivary glands, it may be located in the following sites: the palate, retromolar area, tongue, buccal mucosa, lips, floor of the mouth [23]. A few are also found centrally within the mandible [17]. Adenoid cystic carcinoma (ACC), 31% of these tumours are found in the minor salivary glands, particularly in the palate [21], with the overlying mucosa being ulcerated in 50% to 60% of cases [17]. ACC and Mucoepidermoid carcinoma most frequently affect adults in their fifth and sixth decades of age [21,23].

Palisaded encapsulated neuroma has a predilection for the perioral facial skin and the palate. Lipomas in the oral cavity are most commonly found in the buccal vestibule, floor of the mouth, and tongue, with both palisaded encapsulated neuroma and lipoma most frequently affecting adults in the age between 40 and 60 years [17].

Hemangioma is also considered in the differential diagnosis of intraoral swelling [2]. Arteriovenous hemangiomas commonly develop in the mandible, maxilla, tongue, lower lip, or chin, usually emerging during the teenage or early adulthood. When these lesions affect the mandible or maxilla, their radiographic presentation can vary, appearing as a distinct radiolucent lesion or a well-defined multilocular structure often described as a "soap bubble." In some cases, they may exhibit a fine or mixed radiolucent-radiopaque pattern, resembling fibrous dysplasia. Although many vascular lesions are congenital, but remain subclinical at birth, and only become apparent years later [17].

Considering factors such as age (neurofibroma is more common in younger individuals compared to other lesions), location (when compared to schwannoma, lipoma, fibroma, and pleomorphic adenoma), radiographic appearance (when compared to arteriovenous hemangioma), and intact overlying mucosa (when compared to adenoid cystic carcinoma), these collective findings strongly support the diagnosis of neurofibroma and help rule out other differential diagnoses. However, the definitive confirmation of the diagnosis was achieved through histopathological examination.

The approach to treating solitary neurofibromas is a surgical excision, and recurrence is rare [15]. These tumours are not radiosensitive, and chemotherapy has shown limited benefit [2]. Despite the uncertain risk of recurrence or malignant transformation, the prognosis is excellent [16].

## CONCLUSION

We report a case that is very rare and unusual, although solitary neurofibroma in the palate rarely occurs generally, but this time it appeared in an infant with age of 4-months-old only. The mass was excised, and no recurrence occurred after 5 months.

It is important to note that neurofibromatosis carries a significant risk of malignant transformation, estimated at 5–10%. In young patients, solitary neurofibroma is considered a possible early manifestation of neurofibromatosis type 1 [8], emphasizing the necessity of regular follow-up evaluations.


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## DECLARATION OF INTEREST

Authors declare no conflict of interest.

## DECLARATION OF PATIENT CONSENT

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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