Inflammatory myofibroblastic tumor of gingiva in a patient with constitutional growth delay: A rare case report

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Abstract:
Several disorders related to growth and development affect children during their early stages of life. One rare disorder that affects about 15% of children is constitutional growth delay (CGD). Its results in the affected individuals appear unusually short and young for their age. The individuals are otherwise normal and appear as their normal counterparts as age increases. Inflammatory myofibroblastic tumor (IMT) is a rare soft-tissue tumor that commonly affects the lungs, liver, and gastrointestinal tract. The occurrence of this tumor outside the aforementioned areas is very rare. It resembles a neoplastic tumor clinically and histologically. It is also referred to as inflammatory pseudotumor. Here, we report the case of an individual with CGD who also had an IMT involving the mandibular left canine region.

Key words:
Inflammation, laser therapy, oral cavity, pseudotumor, spindle cells

INTRODUCTION
Constitutional growth delay (CGD) affects 15% of children at various stages of development. The rate of growth varies among individuals. Usually, in the affected children, normal growth and development is seen till 2–3 years of age. Changes in growth pattern typically occur around the time of puberty and manifest differently.[1] The affected children do not show any signs or symptoms that affect growth, but they reach puberty at a delayed time than normal children. It is also found that children with CGD show normal growth patterns after adult age. In short, children with CGD are described as those who appear smaller for their current age, but they grow at a normal rate. They are commonly referred to as late bloomers.[2]

Inflammatory myofibroblastic tumor (IMT) is a rare tumor consisting of numerous inflammatory cells and myofibroblastic spindle cells.[3] Brunn reported the first case of IMT in 1939.[4] Since then, numerous cases have been reported and several terminologies have been used to describe the tumor. Some other names include inflammatory pseudotumor, inflammatory fibrosarcoma, plasma cell granuloma, myxoid hamartoma, pseudo sarcoma, and inflammatory fibrohistiocytic proliferation.[5]

Commonly reported sites of appearance of IMT include lungs,[4] liver,[7] and orbit.[6] In head-and-neck region, the occurrence of IMT has been reported in the maxillary sinus, parapharyngeal spaces, and oral cavity. In the oral cavity, gingiva, tongue, buccal mucosa, mandible, and submandibular salivary gland have been commonly found to be the reported sites of IMT occurrence.[5] This case presentation shows a case of an individual with CGD who also happened to have an IMT involving his interdental gingiva.

CASE REPORT

Clinical examination
A 22-year-old male reported to the department of periodontics with a chief complaint of an overgrowth in his gums that was persistent for a week [Figure 1]. The overgrowth was painless, and mild bleeding on provocation was seen. Previous medical history revealed that the patient was a case of CGD. He was 22 years old but...
looked very young for his age. He had no facial hair growth, and his limbs and extremities did not appear to be that of a 22-year-old male. He was also short for his age [Figure 2]. He was 5 feet tall and weighed about 40 kg. He was not under any form of treatment for the same.

**Intraoral examination**
Gingival examination showed a gingival overgrowth in the lingual aspect of the mandibular left canine involving the interdental papilla. It was ovoid and measured about 10 mm × 8 mm in height and width. The soft-tissue overgrowth was firm in consistency, was mildly mobile, was painless, and did not contain any exudate. The probing depth was 2 mm all around and showed no signs of bone defects and did not warrant any radiographic investigation. A differential diagnosis was given as idiopathic gingival enlargement. To arrive at an exact diagnosis, it was decided to excise the overgrowth and send the specimen for a biopsy. Advanced diagnostic tests were not done on the sample due to poor economic status of the patient.

**Excision of overgrowth**
After adequate local anesthesia, the gingival overgrowth was excised using laser [Figure 3]. Indilase™ soft-tissue diode laser was used to excise the tissue overgrowth. Postsurgical analgesics were prescribed. The excised tissue was sent for biopsy [Figure 4].

**Histopathologic examination**
Histopathologic examination showed dense inflammatory cell infiltrate consisting of plasma cells, lymphocytes, and scanty neutrophils and eosinophils [Figure 5]. Also seen were proliferating spindloid fibroblasts and myofibroblasts with...
DISCUSSION

IMT is a very uncommon and rare lesion of the soft tissues. The WHO defined IMT as an intermediate soft-tissue tumor that comprises of myofibroblasts, inflammatory cells, spindle cells, plasma cells, and/or lymphocytes. The etiological factors behind the occurrence of IMT are not clearly understood. In the oral cavity, the occurrence of IMT is very rare. The presented case is an IMT involving the interdental papillary region of the mandibular left canine and premolar. Diagnosis of IMT can only be done through histopathological examination by performing a biopsy of the tissue mass. In the present scenario, the excision was performed using Indilase™ soft-tissue diode laser. The usage of laser provided the added advantages of minimizing hemorrhage during excision and increased patient comfort.

The etiology and pathogenesis of IMT is poorly understood. The etiology can vary depending on the area of occurrence of the tumor itself. Several reports suggest that the tumor may arise due to autoimmune, infectious, syndrome-related, or traumatic causes. The present individual did not have any such contributing factors other than moderate amount of calculus, which has never been reported as an etiologic factor for the lesion. IMF with involvement of a bony defect is extremely rare with only three cases reported so far.

The biopsy specimen was then studied using hematoxylin and eosin staining to identify the cellular and connective tissue elements. The histopathological picture showed the contents to be myofibroblastic spindle cells, plasma cells, lymphocytes, and eosinophils. This was in accordance with the previous studies reported by Coffin and Fletcher. The differential diagnosis of IMT based on histopathology includes several lesions such as benign fibrous histocytoma, fibrosarcoma, myofibroma, and solitary fibrous tumor. Benign fibrous histocytoma has a characteristic storiform appearance which was absent in the present specimen and was hence ruled out. The histopathologic view of fibrosarcoma shows extensive malignant features and collagenous areas. The present specimen did not show any such findings, and this helped in ruling out the fibrosarcoma. Absence of hemangiopericytoma-like areas and involvement of blood vessels helped in ruling out the myofibroma and solitary fibrous tumor.

CGD is a condition that delays the growth of the affected individuals. The individuals appear young for their age characterized by short stature and relatively slower developmental milestones compared to their normal counterparts. The two possible etiological factors associated with CGD are genetic and constitutional. The definitive etiology of CGD is not well established. The common finding is that all the body tissues are affected at a same rate, and there is an overall delay in the growth process of the affected individual. Diagnosis of a case of CGD is very complex involving an array of investigations including calculating the growth velocity of the child, monitoring growth hormone levels, and radiographic investigations. CGD resembles several other growth-related anomalies such as delayed puberty, Turner’s syndrome, and hypothyroidism. The affected children usually catch up to the others as they grow older. The individual in the present case report was diagnosed to have CGD at an adolescent age and was not under any specific treatment as he was otherwise healthy.

In the present case, the 22-year-old male was about 5 feet tall and weighed 40 kg, which is much less when compared to a normal 22-year-old male. His other physical attributes were in accordance with that of numerous studies done previously about the identification and management of children with CGD.

No specific intraoral findings have been reported so far in individuals with CGD. This case presentation is unique as it shows two rare clinical findings – a rare intraoral tumor and a rare growth defect, in a single individual.

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Conflicts of interest
There are no conflicts of interest.

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