Surgical Management Of An Atypical Case Of Multiple Mandibular Exostoses: A Case Report.
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Citation

Abstract
Multiple exostosis have long being described in the literature yet there is a continuing paucity in its documentation regarding the incidence and prevalence rates. An unusual case of multiple mandibular buccal exostosis reported to our periodontal office with a chief complaint of unaesthetic appearance. The novelty lay in the situation of the multiple lesions on the buccal mandibular alveolus, both the sites of least predilection (5.1:1); in a female patient whilst the text indicates a 1.66:1 incidence. The average dimension of the lesion was found to be 8X6X4mm. Under the clinical diagnosis of multiple exostoses surgical excision was performed under local anesthesia. Histopathological evaluation revealed a mass of dense cortical bone with interspersed areas of trabecular bone suggestive of exostosis. Deep bite resulting in continual occlusal trauma with consequential reactive proliferation of the bone can be hypothesized to be the concerned etiology in the present case. Uneventful healing with no recurrence was reported.

INTRODUCTION
Literature is replete with documentation of various constitutional bone disorders yet there is a continuing paucity regarding the prevalence rate of multiple bony exostosis. Also known as hyperostosis or hamartoma, bony exostosis have been described as non-pathologic, non-neoplastic exophytic nodular growth of dense cortical bone commonly located on maxillary and less frequently on mandibular buccal alveolus. Usually the bicuspid and molar areas are the afflicted sites yet occasionally they may occur in other parts of the jaw, either solitary or multiple, in a symmetrical pattern. A multifactorial etiology has been implicated though the exact nature of these causes has not been elucidated. Nevertheless some authors believe it to be caused by a complex interplay of genetic and environmental factors.

These multi-nodular masses commence in early adulthood and may very slowly enlarge over the years. The overlying mucosa is usually blanched and vulnerable to trauma. Bouquet JE clinically described them as nodular, pedunculated or flat painless self limiting outgrowths which seldom attained colossal sizes hence pronouncing them innocuous. However in the rare instances of untoward sporadic exacerbations they appear not only unaesthetic but also contribute to periodontal disease by promoting food lodgement. Additionally the massive proportions make them vulnerable to trauma, aphthae and herpetic ulcerations thus warranting a surgical intervention. No malignant potential has been reported till date.

A prevalence of only 0.09% was reported by Bouquot and Gundlach with 73% of the lesions encountered on the maxillary alveolus. Jainkittivong and Langlais examined 960 subjects and established a 5.1:1 prevalence of maxilla: mandible with men exhibiting exostosis much more than women (1.66:1). Men also exhibited a higher occurrence in nearly all locations. While these findings are in accordance with other authors however they should be treated with caution since racial and ethnic differences cannot be ruled out. Furthermore, an increased incidence with age has been identified by Neville et al as a consequence to the cumulative functional influences on the bone as indicated by significant attrition of teeth in older individuals.

Interestingly a strong pattern of concurrence of exostosis, Torus Palatinus and Torus Mandibularis has been recognized in literature which persuasively suggests general multiple exostosis syndrome. In both genders, exostosis occurs most frequently in the 35-65 year age group.

CASE REPORT
A 30 year old female reported to our private periodontal
office with chief complaint of unaesthetic facial appearance due to bulky lower jaw. The clinical examination revealed multiple small nodular excrescences of bone on the buccal surface of the mandibular alveolus located below the mucobuccal fold (Fig. 1). Dimensionally each bony protuberance was 8x6x4mm. The overlying mucosa appeared blanched and revealed focal areas of inflammation with associated ulceration. Moderate plaque accumulation on the facial surfaces of teeth was observed corroborating with the patient’s complaint of difficulty in instituting mechanical plaque control measures. Keeping the patient’s aliment in perspective, surgical intervention under local anesthesia was planned for total surgical excision. The patient’s family and medical history were found to be non-contributory. Routine blood and urine investigations revealed normal findings. Pre- surgically the patient was explained the treatment procedure with potential risks and benefits and an informed consent was obtained. Once adequate anesthesia was achieved following infra-alveolar block, a full thickness mucoperiosteal flap was raised to expose the exostosis (Fig. 2). To facilitate the major debulking of the osseous lesion a combination of Oshenbein chisel, Bard Parker blade number 15 (Fig. 3) and mallet were employed. Final gradualizing of the alveolar housing such that it mimics the original contours was performed with micro-motor mounted diamond burs under copious normal saline irrigation (Fig.4,5). Following alveoloplasty, the mucoperiosteal flap was trimmed to fit the reinstated newly established contours and interrupted sutures were placed with 3-0 black mersilk (Fig.6). The patient was discharged with post-operative instructions and recalled after 1 week for suture removal.

By the end of one week, the surgical area has healed uneventfully without any untoward discomfort reported by the patient.

**HISTOPATHOLOGIC FINDINGS**

Histopathologic examination revealed mature bone tissue organized as a mass of dense, lamellar cortical bone with interspersed areas of trabecular bone. Haversion canals with numerous viable osteocytes lodged in lacunar spaces were observed suggesting the diagnosis of multiple exostosis.

**DIFFERENTIAL DIAGNOSIS**

Exostosis may present as one of the signs of Gardner’s syndrome. However in this patient no typical findings such as multiple polyposis of large intestines, multiple epidermoid or sebaceous cysts of the skin were detected.

Additionally absence of hydroxyproline in urine suggested that this patient did not manifest Gardner’s syndrome.

**Chronic Multiple Sclerosing Osteomyelitis** is an unusual reaction of the bone to mild bacterial infection entering the bone through multiple carious lesions in persons with high degree of tissue resistance and tissue reactivity. Typically the patients exhibit multiple large carious lesion with mild pain associated with an infection pulp which was characteristically absent in our patient.

Multiple Odontogenic Keratocysts are benign cystic neoplasms which may clinically manifest themselves as medullary expansion of bone with associated pain, swelling, discharge, occasionally paresthesia of the lower lip, fracture of the jaw or may be completely asymptomatic. However, a strong correlation of multiple OKCs with Naevoid basal cell carcinoma syndrome has been established characterized by congenital skeletal defects, ectopic calcifications, planter and palmer pits, central nervous system and ocular lesions, frontal bossing which were typically absent in our patients.

**Figure 1**

Fig. 1 Multiple buccal exostosis along the right and left mandibular alveolar ridges: Pre-operative view
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**Figure 2**
Fig. 2 Full thickness mucoperiosteal flap elevated to expose the exostoses

**Figure 4**
Fig. 4 Final Recontouring being done to attain anatomic configuration

**Figure 3**
Fig. 3 Excision in progress

**Figure 5**
Fig. 5 Final bone contour achieved conforming to the anatomic form
DISCUSSION

With a 0.09% prevalence\(^9\), exostosis certainly merits as an uncommon benign lesion of the oral cavity. In this case report, we attempt to bring forth a highly atypical variant of the same which awards novelty to our presentation.

Contrary to the clinical reporting of various authors\(^{18,19}\), multiple bony exostosis were found on the buccal alveolus of the mandible; both sites of least predilection. Moreover, it was observed in a female patient while the literature emphatically documents a male predominance\(^{11,12}\).

The etiology of exostosis has been investigated by several authors, however no consensus has been reached. Giving credence to the quasi-continuous genetic or threshold theory, Gould AW(1964)\(^20\) identified exostosis as an autosomal dominant lesion caused due to a complicated and unclear interplay of environmental and genetic factors. Curiously, literature documents cases of spontaneous sequestration of bone occurring with an almost certainty in patients genetically predisposed to develop exostosis.

A strong relationship between the increasing age and occurrence of exostosis has been established by many authors\(^{10,17}\) which Nery et al\(^19\) attributed to the exposure of the oral environment to more rigid functional activities. This finding contrasts with that of Sonnier et al\(^12\) who noted a decreasing prevalence of exostosis after 50 years of age and correlated it with edentulism.

Jainkittivong et al\(^10\) described it as a reactive outgrowth in response to functional influences in genetically predisposed population exhibiting marked exostosis and significant attrition of teeth in older subjects. A similar pathophysiology may be implicated in the occurrence of reactive subpontic exostosis which may develop from the alveolar crestal bone beneath the pontic of a posterior bridge\(^17\). Stress causes the crestal alveolar bone to grow under the pontic along a vector opposing the forces of occlusion\(^21\).

Less commonly, solitary exostosis may occur in response to local irritation from the alveolar bone beneath free gingival grafts and skin grafts. Presumably the graft acts as a stimulant to the periosteum thus commencing new bone synthesis.

Sonnier et al\(^20\) while discussing palatal exostosis commented on its practical implications while planning periodontal surgeries in the posterior maxilla. Buccal exostoses on the other hand are significant for prosthodontists since they may interfere with preparation and insertion of the prosthetic appliance. Hall\(^22\) reported incidences of frequent aphthe and herpetic ulcerations with exostosis. Additionally buccal exostosis may be traumatized and interfere with oral hygiene procedures as was the case in this patient hence warranting their surgical removal.

As mentioned previously cases of co-existence of multiple exostosis and tori have also been reported in literature indicating general multiple exostosis syndrome.

CONCLUSION

Owing to their benign innocuous nature, exostosis in majority of cases does not necessitate any surgical intervention unless in the event of tissue trauma, periodontal or prosthodontic complications or pathological outbreaks like herpetic or aphthous ulcerations. Careful surgical planning while keeping the basic flap design and the gingival anatomy in mind would definitely culminate satisfactory outcome for both the clinician and the patient.

References

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