Concurrence of torus palatinus with palatal and buccal exostoses

Case report and review of the literature

Demetrios Z. Antoniades, DDS, MD, MSc, PhD, Maria Belazi, DDS, PhD, and Panayiotis Papanayiotou, DDS, MD, PhD, Thessaloniki, Greece

Exostoses, also known as hyperostoses, are localized bony protuberances that arise from the cortical plate. These developmental anomalies, or hamartomas, frequently affect the skeletal jaw. Different types of exostoses have been described. Torus palatinus and torus mandibularis are two of the most common intraoral exostoses. Other types of exostoses, affecting the palatal aspect of the maxilla (palatal exostoses) or the buccal aspects of the jaws (buccal exostoses), are less commonly encountered. Concurrence of different forms of exostoses in the same individual is very rare. A 48-year-old woman manifesting excessive palatal exostoses, torus palatinus, and buccal exostoses is described. We present the clinical and histopathologic features and applied therapy and provide a comprehensive review of the current features of exostoses. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1998;85:552-7)

Under the general term exostoses are described non-pathologic, localized bony protuberances that arise from the cortical bone and sometimes from the spongy layer. Such developmental anomalies, or hamartomas, are not pathologically significant, and they most frequently develop in the human jaw bone. Two of the most common exostoses that occur in two specific intraoral locations—on the midline of the hard palate and on the lingual aspect of the mandible in the cuspid/premolar region—are termed torus palatinus (TP) and torus mandibularis (TM).

TP is an exophytic nodular mass of bone that arises along the midline suture of the hard palate. Morphologically, tori are classified as flat, spindle, nodular, and lobular. In the literature it is noted that an exostosis of the hard palate was first observed and reported by Santorini in 1724, whereas the earliest article describing the exostotic changes of the hard palate was written by Fox in 1814. Carabelli provided a detailed description of palatine torus in 1842 and even suggested a familial tendency for trait expression. Although the trait had been described under various names early in the 19th century, the term torus palatinus was coined by Kupffer and Bessel-Hagen in 1879 in a letter addressed to Virchow. It was further claimed that it was characteristic of East Prussian skulls. Late-19th- and early-20th-century researchers associated palatine torus with syphilis, tuberculosis, rickets, scurvy, cancer, insanity, and criminality and even with the regularity of sexual activity. In the 20th-century biologists made a concerted effort to include palatine torus in textbooks of oral anatomy.

TM, the other most common exostosis, is a bony exophytic growth located in the cuspid/premolar area of the lingual surface of the mandible and superior to the mylohyoid ridge, usually bilaterally. TM was first described by Danielli in 1884; however, the term torus mandibularis was first used in 1908 by Fürst, who used it to denote a bony protuberance that developed on the lingual surface of the mandible, most often in the premolar and canine areas.

Most dentists are familiar with these variations of exostoses. However, histologically identical lesions occurring on the facial aspect of the maxillary or mandibular alveolar bone, usually in the premolar/molar area and on the lingual aspect of the maxillary tuberosities, have also been described; these are referred to as buccal exostoses and palatal exostoses, respectively.

Woo first described TM as a hyperostotic formation that has developed on the alveolar portions of the maxilla. In 1968 Goldman and Cohen mentioned that a high incidence of large exostoses of bone present on the palatal alveolar process, extending from the mesial aspect of the maxillary first molar distally to the end of the tuberosity, is a possible factor in delayed healing of gingivectomy wounds in this area. The term palatal exostosis was later used for the description of this type of exostosis by other authors. Shafer et al. considered the term multiple exostoses to be more appropriate for the description of this form of exostosis; however, we must note that Shafer et al. referred to exostoses as those protuberances found mostly on the
buccal aspect in living subjects, whereas Wool11 had used skulls. As mentioned, Neville et al.1 described the aforementioned variations as buccal exostoses and palatal exostoses, whereas Sapp et al.2 and Regezi and Sciubba3 used the term exostosis.

Exostoses of another interesting type, termed solitary exostoses by Neville et al.,1 have recently been described. These develop from the alveolar bone beneath free gingival grafts24,25 and skin grafts.26,27 With respect to this type of exostosis, Siegel and Morton and Natkin28 in 1990. This variant is referred to as reactive subpontine exostosis (subpontic osseous proliferations; subpontic osseous hyperplasia) by Neville et al.1

With respect to the prevalence of tori, although a substantial number of investigations have been conducted in materials of various origins, global information on larger populations is incomplete.5 Many studies have shown considerable racial divergences or ethnic group differences in the occurrence of tori, even among groups with apparently identical or closely related ethnic and genetic backgrounds.5,29-45 The reported divergences and prevalences can be attributed to a number of factors.5 Haugen5 believed that the unqualified use of the term torus in doubtful cases is one of the main reasons for the highly divergent prevalences reported in ethnically and genetically closely related materials. In addition, ill-defined criteria for diagnosis or for the selection of material and its classification, doubts about its ethnic representativeness, and unsuitable methods applied in the handling of the material could be partly responsible.1,5,43

Another confounding factor is the matter of whether the studies were conducted in live patients or in skulls.1,5,43 It is interesting to emphasize that values for prevalence of the two torus variants obtained from skulls are higher than those obtained from living subjects. This is due to the fact that small tori are more likely to be found in skulls than living subjects, in whom they are obscured by mucosa and mucous glands.5,11,46 For this reason, Halfmann et al.47 have noted that skeletal and living samples must be treated separately and that direct comparisons of torus frequencies in extant and extinct groups should be made only with extreme caution. Thus our literature review of this field, which is far from easily surveyable, showed that reported prevalence values for TP ranged from 0.4% to 66.5% in a number of population studies, whereas those for TM ranged from 0.5% to 63.4%,1,31-46 With respect to palatal exostoses, studies on dry human skulls conducted by Larato21 in 1972 and Nery et al.22 in 1977 in different ethnic groups revealed prevalences of 30% and 40.53%, respectively. With respect to buccal exostoses, Bouquot and Gundlach42 reported in 1986 that the prevalence rate was 0.9 per 1000 persons and that approximately 73% of the lesions were encountered on the maxillary alveolar bone.

In addition, our review of the literature showed gender differences for the prevalence of tori. Some authors found that TP affected males more often than females.29,38; however, most investigators found that TP more frequently occurred in females.* The female:male ratio averaged 1.7:1, ranging between 1.3:1 and 2.3:1.1,45 In contrast, the prevalence of TM was found to be higher in males than in females5,29,44 except in one study.32 In both genders, the two types of tori occurred most frequently in the 35-to-65-years age group.5 This finding may indicate a general trend for tori to be more frequently observed during the middle phase of life than at younger or older ages.5,30,45,49

Although TP and TM have been the subject of numerous studies, these traits continue to be two of the most controversial and least understood skeletal characteristics.50 Recently Haugen5 pointed out divergent conclusions about whether the two traits are genetically correlated or are independent morphologic units. Although a few workers have acknowledged that more than one main factor may be significantly operative, most have tended to emphasize either genetic or nongenetic influences. Some authors have considered dietary habits and nutritional disturbances (avitaminoses) to be an underlying cause.29,30,43,49 Several studies have supported the hypothesis that torus formation is influenced by stress from masticatory hyperfunction.23,29,50-52 A correlation between the occurrence of tori and either abrasion of the teeth or additional stress on the jaws because of habitual clenching and grinding of the teeth for other

*References 5, 11, 15, 30, 34, 36, 42, 43, 45-48.
than chewing purposes (bruxism) has also been considered significant. Evolution, continued growth, heredity, and environmental factors have also been postulated as causes.

In general, the best explanation for the prevalence of both tori types seems to be an interplay of genetic (familial nature) factors and environmental factors, including functional (masticatory hyperfunction), nutritional, behavioral, and possibly, climatologic factors. Interestingly, Ossenberg has proposed a mechanism by which horizontal forces tip the lower teeth, first molar to canine, so that their root apices exert pressure on the periodontal ligament, causing formation of new bone on the outer cortical plate of the alveolar process on its lingual surface. Eggen and Natvig proposed that the presence of a torus was 30% genetic and 70% environmental in terms of occlusal stress. In studies conducted on white people of the same generation living in different environments, differences in the presence of TP have been found. This finding cannot be explained by race alone. We have mentioned that there is evidence showing tori to be more frequent in midlife; this observation indicates that a torus should be explained as a dynamic phenomenon rather than as a stationary or progressively growing lump of bone. Such an explanation is supported by the observed difference in the incidence of TP between edentulous persons and both dentate and partially edentulous persons (36.5% vs 48%, respectively), because patients who are edentulous would exhibit less masticatory hyperfunction.

The literature on the concurrence of different forms of tori is scattered. The studies were performed on populations of differing racial and ethnic backgrounds. All these studies exclusively refer to the relation of TM and TP and show a low prevalence of concurrence. We were able to find only two reported cases of the concurrence of tori with buccal or palatine exostoses: one, a case of maxillary exostoses, palatine exostoses and torus mandibularis, was reported by Blakemore et al. in 1975; the other was reported by Topazian and Mullen in 1975. Concerning the concurrence of exostoses and tori, Kolas et al. conducted a study in a large, ethnically complex group of white and black Americans. They found a concurrence of 3.03% and concluded that there is no causal relation between the appearance of TP and the appearance of TM.

Reichart et al. reported the concurrence of TP and TM at 0.7% in a population of German patients and at 4.7% in a population of Thai patients. Haugen reported low prevalences and little or no correlation between the two types of tori in patients from the area of Oslo, Norway. The concurrence of the two tori was 2.22% among 5000 patients, with no significant gender difference (2.34% for males and 2.07% for women). Haugen also found a low or nonexistent correlation between the appearance of the two tori; among 716 individuals examined, 111 showed concordance and 605 showed dissension for the trait "combined torus formation." However, by looking at individual probands, it has been shown that a person with TM has twice the normal risk of developing TP, and vice versa. Such considerations make it difficult to reject the notion that there must after all be some causal relationship between occurrences of the two tori. More recently, Eggen et al. conducted a meta-analysis on the concurrence of TM and TP on a data set of 2010 persons native to two different regions of Norway, the Lofoten Islands and the Gudbrandsdalen Valley. They concluded that there was a clear correlation between the presence of the two types of tori. This is the only study that has reported a significant correlation. Interestingly, the same study reported a statistically significant concurrence of TM and TP among women only. The authors hypothesized that such a gender-related dimorphism of the variable "combined occurrence of the two types of tori" might have something to do with the divergent conclusions between the two genders reported earlier.

As mentioned, the appearance of TM and TP appears to be a result of the interplay between environmental and genetic factors. With regard to genetic factors, many models have been proposed. On the basis of family pedigree analysis, an autosomal dominant mode of inheritance was suggested. This model has been rejected, however, because more recent studies have been unable to confirm complete penetration of the TM or TP trait from one generation to the next. Eggen and Natvig suggested that variations in the prevalence of TM and TP between the two genders may be explained by an X-linked dominant or X-linked recessive mode of inheritance. However, the hypothesis that
the trait is X-linked and dominant cannot be corroborated by the data, because it would require that fathers transmit the TM and TP trait to all their daughters. In addition, the possibility of an X-linked recessive trait has been rejected because such a model would require that the trait typically occur in men, a conclusion that is not in accord with the data.

More recently, Eggen et al. have suggested that TP and TM may be explained in terms of an underlying continuity in the population (termed liability) with a threshold value above which individuals will be affected. Liability in this case is defined as having a polygenic attribute expressed in combination with environmental factors. This model posits a “multifactorial” system that can explain the individual variation in the expression of TM and TP. Eggen and Seah have specifically defined TM as a multifactorial/threshold trait expressed in a quasi-continuous fashion. Quasi-continuous characteristics tend to cluster in families; however, unlike single-trait genes, genetic patterns are not clear-cut and do not fit any model of dominance or recessivity.

With respect to palatal and buccal exostoses, which were observed in the case about to be described, we have been unable to find an etiologic analysis for their appearance. Shafer et al. suggested that they are of unknown origin, and no figures are available as to their incidence or disposition. Because of the morphologic similarities between palatal/buccal exostoses and TM and because of the position of the former on the lingual/buccal aspects of the maxilla and of the latter on the lingual aspects of the mandible, it is reasonable to hypothesize that the quasi-continuous model of inheritance also applies to palatal/buccal exostoses. This view is reinforced by Ossenberg, who has argued that the same masticatory stress (ie, an environmental factor) may underlie the appearance of all oral hyperostoses.

Further evidence for the multifactorial model is offered by Nery et al., who have shown a differential appearance of palatal exostoses among four populations in Europe, Oceania, and Asia, Africa, and Latin America.

In their study Nery et al. showed that location is an influencing factor in the causation of palatal exostosis. In addition, they assumed that a gene plays an important role, although at that time they could not substantiate their conclusions. They stated that this was a worthwhile subject for further investigations. According to them, the different degrees of occurrence in various localities also suggest a hereditary type of exostosis, perhaps in the same category as that of hereditary multiple exostosis, described by Ehrenfried, Keith, Jaffe, Stark et al., and Gardner et al. Finally, Nery et al. suggested that this trait may be part of a general multiple exostosis syndrome in the rest of the skeletal system.

As noted, our review of the literature has revealed only two cases of concurrence between TP and palatal exostoses. Our interest in the following clinical case of multiple exostoses was thus prompted by the scarcity of evidence in the literature regarding concurrent appearance of oral exostoses.

**CASE REPORT**

A 48-year-old woman was referred to the clinic of the Department of Oral Medicine and Oral Pathology, Aristotle University School of Dental Medicine, for evaluation and treatment of multifocal maxillary exostoses. The patient was concerned about a discomfort associated with movement of her tongue and about frequent irritation of the palatal mucosa during mastication of hard food (eg, crackers, toast). Surprisingly, the patient had been unaware of the presence of the exostoses until 8 months before their removal; fearing that the lesions were cancerous, she had not visited her dentist until just a few days before the exostoses were removed in our clinic.
Fig. 3. Photomicrograph of histologic appearance of palatal exostosis shows dense bony tissue, presence of lacunae, and normal osteocytes. Scattered spaces of connective tissue containing dilated vessels are seen (hematoxylin-eosin, original magnification x120).

Physical examination of the oral cavity revealed extensive bony overgrowths located on both palatal alveolar processes extending from the mesial aspect of the maxillary first molar distally to the terminus of the tuberosity. In addition, in the midline of the posterior portion of the hard palate, a lobular exophytic bony protuberance (2 × 2 cm) was observed (Fig. 1). Finally, small bony nodules were noted on the right and left buccal aspects of the maxillary alveolar ridges, predominantly in the premolar area (Fig. 2). All the bony exostoses were covered by normal mucosa. Missing teeth (the maxillary right second premolar and left first and second premolars) had been replaced by stable prostheses. The oral health of the patient was generally satisfactory. Bruxism and other habits were not mentioned by the patient. To her knowledge, her parents had not experienced any growths in their mouths. Before surgical intervention a panoramic radiograph was obtained for the sake of estimating the thickness of bone between the exostoses and the maxillary antrum and floor of the nose. This radiograph indicated that complete removal of the exostoses and the torus palatinus could be performed without fear of antral or nasal perforation. In addition, an alginate impression of the maxilla was taken before surgery and a diagnostic cast was constructed. The surgical procedure was performed with the patient under local anesthesia induced by means of 2% xylocaine with 1:100,000 epinephrine. Removal of the exostoses was performed after standard procedures.

HISTOPATHOLOGIC FINDINGS

Multiple irregular hard-tissue fragments were submitted in 10% formalin for histopathologic examination. The examination of hematoxylin and eosin staining specimens revealed decalcified dense bony tissue, the presence of lacunae, and normal osteocytes. Scattered spaces of connective tissue containing dilated vessels could be observed (Fig. 3).

CONCLUSION

To the time of this writing, most studies of jaw bone exostoses have referred to two common types of tori, TM and TP. Most studies compared the occurrences of these tori in the two genders and in different ethnic groups and noted their variations with respect to age, gender, causation, and pathogenesis. Because the information from these studies was considered sufficient, the view that no more studies were needed had prevailed. However, it was suggested that more studies should be conducted to elucidate the controversies associated with the topic of exostoses—in particular, the notion that the proposed quasi-continuous genetic/threshold model could perhaps explain the biological processes underlying the formation of exostoses. On the other hand, Flynn et al. suggested that additional studies on tori were required for clarification of the reasons for their appearance and growth. Bouquot and Gundlach suggested that there has been no valid epidemiologic investigation of tori in the United States. Haugen reported that the number of studies focusing on the occurrence of tori in the population and the explanation of their biological nature, as opposed to the considerable number of ethnologic and statistical studies, is rather small.

In our country we have only found one study focusing on the clinical and epidemiologic aspects of TM. It was this scarcity of studies on tori that instigated our report of the unusual case we encountered, of which the striking feature was the concurrence of palatine tori with palatine/buccal exostoses.

REFERENCES


Reprint requests: Demetrios Z. Antoniades, DDS, MD, MSc, PhD School of Dental Medicine Department of Oral Medicine and Oral Pathology Aristotle University of Thessaloniki Thessaloniki 54006 Greece