



## Taurodontism in association with the cusp of carabelli: report of two cases

**\*Sote EO, \*\*Ogunkola AO**

\*Department of Child Dental Health, College of Medicine, University of Lagos, \*\*Lagos University Teaching Hospital, Lagos, Nigeria

**\*Correspondence: Sote EO**

**Email: lizsote@yahoo.co.uk**

### Abstract

Taurodontism is a malformation of multirooted teeth characterized by abnormally large pulp chamber and abnormally short roots. It is diagnosed by the use of radiographs. The cusp of Carabelli is a morphological anomaly of the crown usually seen clinically on the mesiopalatal surface of the maxillary first permanent molars. The aetiology of both anomalies is not entirely clear, however, both genetic and exogenous factors have been proposed. There are recent reports that mutation of DLX3 (17q21) could be a cause of taurodontism. Taurodontism has been reported along with talon cusp, dens evaginatus and some syndromes.

In this paper we present an unusual occurrence of taurodontism in association with the cusp of Carabelli in a 15 year-old girl presenting with a toothache of 4 months duration and her 46 year-old mother. All the second permanent molars were taurodont. Other siblings investigated had cusps of Carabelli but no taurodont tooth. There were no associated syndromes in both mother and child.

Root canal treatment of the pulpally involved taurodont mandibular left permanent second molar of the 15 year-old was carried out. Appropriate restorations were placed on all other carious molars. Her mother had no carious tooth.

Taurodontism may present as pain when the affected tooth is carious or as a chance radiographic finding as it is the case in the mother. Preventive restorations of taurodont teeth is ideal treatment when detected early. The anthropological and clinical significance of the cusp of Carabelli is highlighted. The association of these two dental anomalies needs further investigation.

### Key word:

### Introduction

Taurodontism (MIM 272700) is a malformation of multirooted teeth, characterized by a large crown-to-root ratio. The roots are abnormally short, and the pulp chamber is abnormally large<sup>(1)</sup>. The condition was considered to be predominantly found in prehistoric hominids. However, the dental literature is now replete with numerous reports of taurodontism in modern man. The first report in modern man was in 1909 by Pickerill<sup>(2)</sup> where he used the term radicular dentinoma. The term 'taurodontism' was later coined by Keith<sup>(3)</sup> in 1913 from the Greek words tauros (meaning bull) and odonto (meaning tooth) to describe an unusual tooth form with "a tendency for the body of the tooth to enlarge at the expense of the roots".

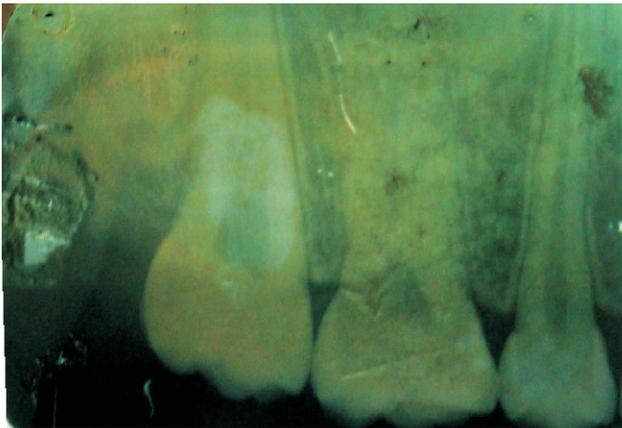
Taurodontism occurs in syndromes such as tricho-dento-osseous, Klinefelter, Down and in amelogenesis imperfecta hypomaturation-hypoplastic type<sup>(4-6)</sup>. It also occurs as an isolated trait with predilection for association in certain familial groups with common racial, geographical, and ethnic backgrounds and in patients with a female habitus and X-chromosomal aneuploidy and a male habitus and X-chromosomal states<sup>(7)</sup>. The aetiology of taurodontism is not entirely clear. Theories on the aetiology include, among others, a primitive pattern, a mutation, a continuous trait without discrete modes of expression, familial in nature and an autosomal dominant trait<sup>(7)</sup>. Development of taurodontism has also been explained in various ways such as a delay in calcification of

the pulp chamber floor, an odontoblastic deficiency, an alteration in Hertwig's epithelial root sheath with a failure of the epithelial diaphragm to invaginate at the normal horizontal level, and a delayed or incomplete union of the horizontal flaps of the epithelial diaphragm<sup>(7,8)</sup>. Recent investigations have revealed that mutation of DLX3 (17q21), a transcription factor involved in root sheath, delay the inward invagination of the sheath toward the pulp, resulting in taurodontism<sup>(1,9)</sup>.

Taurodontism occurs in varying degrees. In an attempt to define the variations of expression, Shaw<sup>(10)</sup> in 1928 classified the condition into three subtypes, "hypotaurodontism", "mesotaurodontism", and "hypertaurodontism." Other more objective classification, most of them based on Shaw's subtypes are in the literature. Keene<sup>(11)</sup> developed the much criticized "taurodontism index", Blumberg et al<sup>(12)</sup> in 1971 developed a discriminant analysis, while Seow<sup>(13)</sup> introduced the calculation of specific range of values for the crown-body/root ratio for the cynodont, hypotaurodont, mesotaurodont, and hypertaurodont teeth.

The prevalence of taurodontism is reported to vary from 0.54% in the primary dentitions of Japanese children to 5.6% in the permanent dentition of Israeli adults. Its incidence is higher in mongoloid and capoid races<sup>(7)</sup>. In Nigeria, the prevalence is unknown possibly due to very rare reporting of the condition since diagnosis of the condition is mainly by the use of radiographs.

The cusp of Carabelli (MIM 114700) is a morphological



**Figures 1a & b** showing abnormally large pulp chamber and very short roots in taurodont maxillary right and left second molars in Case 1.

anomaly of the crown usually located on the mesiopalatal surface of the maxillary first permanent molars. It may present unilaterally or bilaterally in varying sizes. According to Mitchell<sup>(14)</sup> in 1892, the cusp was first described in 1842 by Georg Carabelli. It occurs rarely on the second or third molars, or on the maxillary first primary molars<sup>(15)</sup>. The aetiology of the cusp of Carabelli is unknown. However, most studies agree that it is a heritable feature, and both genetic and exogenous factors have been proposed<sup>(15-19)</sup>. The development of the trait is affected by multiple genes. The incidence and degree of expression differ and it is being used to detect, characterize and to compare different populations<sup>(16)</sup>. It is commonly seen in Caucasians, but rarely found in persons of Mongolian or Inuit heritage. The prevalence of the Carabelli cusp in Europeans is 75-85% while in the Pacific Islands 35 - 45% was reported<sup>(15-17)</sup>.

To date, the only known documented case of taurodontism in Nigeria was reported in a 12 year-old girl over 25 years ago<sup>(20)</sup>. Although the cusp of Carabelli is not an infrequent clinical finding among Nigerians, the authors are not aware of any documented case of the cusp of Carabelli from Nigerians in the recent dental literature. In our paper, we present two unusual cases of taurodontism in association with the cusp of Carabelli. Case report

#### Case 1

A 15 year-old girl attended the Paedodontic clinic of the Lagos University Teaching Hospital, with the complaint of tooth ache of 4 months duration on the mandibular left jaw. The pain was described as severe, not spontaneous, did not disturb her sleep but sometimes was referred to the head and face on the same side. Her medical history was nothing remarkable. Her dental history recorded an extraction of an erupting mandibular left third molar which was presumed to be impacted 3 weeks previously. She consumed a lot of refined carbohydrates including chocolates, biscuits, and cakes, and brushed once daily with fluoride - containing toothpaste

Clinical examination did not reveal any abnormality extraorally. However, intra-orally, her oral hygiene was poor. All the permanent second molars and the mandibular left first molar were carious. The mandibular right first molar had caries on its buccal surface. The mandibular left first and second molars were tender to percussion. An

accessory cusp was also observed on the mesiopalatal surface of each of the two maxillary first molars. Radiographic examination showed carious pulpal involvement of only the mandibular left second molar and all the second molars having very large pulp chambers and abnormally short roots.

A diagnosis of apical periodontitis of the mandibular left second molar, taurodontism of all second molars and cusp of Carabelli of the maxillary first molars was made. (Figures 1a & b; Figure 2).

Root canal treatment of the mandibular left second molar was carried out. Meanwhile, the crown was restored with amalgam but subsequently would be replaced with an appropriate crown coverage. The maxillary left second molar was also restored with occlusal amalgam while the carious buccal surface of the mandibular right first molar was restored with glass ionomer cement. Fissure sealant was placed on the maxillary right second molar with incipient caries occlusally. Oral prophylaxis was carried out and oral hygiene instruction and dietary advice was given. The cusps of Carabelli were well molded with no deep fissures, thus they did not require any treatment. The patient on review at 1 month and follow-up at 4 months was without any symptoms.

#### Case 2

The second case is the mother of the patient in Case 1 who was investigated for taurodontism out of interest because of the familial tendency of the condition. She was 46 years old; a mother of two boys and two girls, and a caterer by profession.

Her medical history revealed she had duodenal ulcer which was treated. She had a history of dental trauma some unspecified years back. Although this may seem difficult for a caterer, she claimed that she had stopped taking refined carbohydrates several years back. She brushed once daily with fluoride-containing toothpaste. Clinical examination revealed no abnormality extraorally. Intraorally, there was enamel fracture of the maxillary right central and lateral incisors but with no symptoms or discoloration. There was no evidence of loss of vitality of the traumatized incisors. There was no carious tooth either. Her oral hygiene was good probably owing to a recent scaling and polishing she had. She had mild crowding in the

Figures 1c & d



**Figures 1c & d** showing abnormally large pulp chamber and very short roots in taurodont mandibular right and left second molar in Case 1



**Figure 2a** showing the cusps of Carabelli bilaterally on maxillary right and left first molars in Case 1

**Figure 2b** showing root filled mandibular left second molar in Case 1



**Figure 3a** showing abnormally large pulp chamber with no cervical constriction in taurodont mandibular left first molar in Case 2.

**Figure 3b** showing cusp of Carabelli on maxillary left first molar in Case 2

mandibular anterior region and gingival recession on the mandibular right central incisor. On the mesiopalatal surface of the maxillary left first molar was a prominent accessory cusp (Figure 3).

Radiographic investigation showed her mandibular first molars with large pulp chambers and short roots. A case of

taurodontism of the mandibular first molars and cusp of Carabelli on the maxillary left first molar was diagnosed. None of the other children investigated had features of taurodont teeth while they all had cusps of Carabelli either unilaterally or bilaterally. Meanwhile, management of her other oral conditions were being handled by the appropriate specialties.

## Discussion

Taurodontism is a morphological dental anomaly of multirrooted teeth manifesting as an abnormally large pulp and short roots. The cusp of Carabelli is a morphological variant of the tooth crown. Two cases of taurodontism in association with the cusp of Carabelli were reported here in a mother and child. Although the father was not available for investigation, all other siblings investigated had cusps of Carabelli on one or more of their maxillary first molars but had no taurodont tooth. Taurodontism may occur in some syndromes and as an isolated trait. Both cases in this report did not occur in association with any syndrome. Its association with the cusp of Carabelli has rarely, if ever, been reported previously. The explanation of this occurrence is not easy to come by considering the fact that the other siblings had cusps of Carabelli but no taurodont tooth. The pathogenesis of taurodontism is not entirely clear. Abnormalities of root number and morphology arise from anomalous root sheath activity. DLX3 (17q21) is a transcription factor involved in root sheath activity. It has been reported that DLX3 mutations delay the invagination of the root sheath toward the pulp, resulting in taurodontism<sup>(19)</sup>. Recent reports among the indigenous southern African populations indicate that third molars are probably the most commonly affected by taurodontism while the first molars are the least<sup>(21)</sup>.

The cusps of Carabelli in this report were found on the characteristic site. The cusp in mother and all her children agrees with the suggestion that there must be a dominant gene responsible for the presence of the cusp<sup>(16, 19)</sup><sup>(15, 16)</sup>. To the best of the authors' knowledge the cases in our report are full-blood Nigerians. The prevalence of taurodontism and the Carrabelli trait in Nigeria is yet to be determined thus no valid comment or comparison with the other races can be made at present. The association of taurodontism, (an anomaly of the root) with the Carabelli trait, (an anomaly of the crown) needs further investigation. The only case of taurodontism previously reported in Nigeria was in association with bilateral congenitally missing maxillary lateral incisors and a maxillary canine with localized enamel hypoplasia<sup>(20)</sup>.

The most remarkable finding in our reports is probably the contrast between caries activity in the mother and the daughter, both with taurodont molars. While the daughter had caries on all the taurodont molars and cynodont mandibular left first molar, the mother had no carious tooth. Apart from age difference, the dietary and snacking habit was the main varying factor between them. This clinical observation again underscores the deleterious effect of excessive ingestion of refined carbohydrates on oral health, particularly on dental caries risk.

The clinical significance of taurodontism, an abnormality of root morphology becomes apparent when there is the need to carry out root canal treatment or vital pulp treatment because of the large pulp chamber and the short roots. There is a high risk of early carious pulpal involvement due to the abnormally large pulp chamber. Root canal treatment was carried out successfully on the mandibular left second molar in the first case of this report, albeit very challenging. Reports of successful root canal treatment of taurodont molars are documented in the literature<sup>(22)</sup>. Conservative treatment of other carious teeth was carried out as much as possible considering the characteristic large pulp at the expense of hard tissues.

Deep occlusal caries were restored with amalgam while glass ionomer cement restorations were used for minimal buccal caries to control the caries and to prevent pulpal involvement. Prevention of initiation of caries by placing pit and fissure sealants is recommended and should be carried out when taurodont teeth are detected early.

The usefulness of the Carabelli cusp in comparing and characterizing populations is very well documented in anthropology<sup>(16)</sup>. However, this dental anomaly is also of clinical significance in that the presence of a cusp of Carabelli may present some challenges clinically during orthodontic treatment, moreso because it is most frequently located on maxillary first molars. The presence of deep fissures and grooves on such a tooth can also render it more susceptible to caries because of the tendency to trap plaque and food debris. Root canal treatment may be the last resort in order to avoid extraction of taurodont teeth. Root canal treatment when performed skillfully can be successful. Preventive restorations including fissure sealants are ideal when taurodont teeth are detected early.

## Conclusion

Taurodontism may present as an incidental radiographic finding or as pain when the affected tooth is carious. Thorough investigation and correct diagnosis are of paramount importance so as to prevent instituting erroneous treatment.

More attention should be drawn to the cusp of Carabelli for both clinical and other reasons.

## Acknowledgement

The authors gratefully acknowledge the cooperation of the family in this report.

## References

1. Nanci A. TenCate's Oral Histology, Development, Structure, and Function. Seventh Edition. Mosby Elsevier Inc. 2008; pp 187-188.
2. Pickerill HP. Radicular aberrations: bilateral radicular dentomata. Proc Roy Soc Med (Odont Sec) 1909; 2: 150-152.
3. Keith A. Problems relating to the teeth of the earlier forms of pre-historic man. Proc R Soc Med 1913; 6: 103-110.
4. Crawford PJM, Aldred MJ. Amelogenesis imperfecta with taurodontism and the tricho-dento-osseous syndrome: separate condition or a spectrum of disease? Clin Dent 1990; 38: 44-50
5. Crawford PJM, Evans RD, Aldred MJ. Amelogenesis imperfecta: autosomal dominant hypomaturational-hypoplasia type with taurodontism. Br Dent J 1988; 164: 71-73
6. Schulman GS, Redford-Badwal D, Poole A, Matthieu G, Bursleson J, Danser D. Taurodontism and learning disabilities in patients with Klinefelter syndrome. Pediatr Dent. 2005; 27(5) 389-394.
7. Jaspers MT, Witkop (Jr) CJ. Taurodontism, an isolated trait associated with syndromes and X-chromosomal aneuploidy. Am J Hum Genet 1980; 32: 396-413.
8. Gleiser I, Hunt EE. The permanent mandibular first molar, its calcification, eruption, and decay. Am J Phys Anthropol 1955; 13: 253-284



9. Dong J, Amor D, Alfred MJ, Gu T, Escamilla M, McDougall M. DLX3 mutation associated with autosomal dominant amelogenesis imperfecta with taurodontism. *Am J Med Genet* 2005; 133: 138-141.
10. Shaw JC. Taurodont teeth in South African races. *J Anat* 1928; 62: 476-498.
11. Keene HJ. A morphologic and biometric study of taurodontism in a contemporary population. *Am J Phys Anthropol* 1966; 25: 208-209.
12. Blumberg JE, Hylander WL, Goepp RA. Taurodontism: a biometric study. *Am J Phys Anthropol* 1971; 34(2): 243-256 erratum 36: 307-308, 1972.
13. Seow WK. Taurodontism of the mandibular first permanent molar distinguishes between the trichodonto-osseous (TDO) syndrome and amelogenesis imperfecta. *Clin Genet* 1993; 43: 240-246.
14. Mitchell WH (1892): Case report. *Dental Cosmos* 34: 1036.
15. Alvesalo N, Nuulila M, Portin P. The cusps of Carabelli, occurrence in first upper molars and evaluation of its heritability. *Acta Odontologica Scandinavica* 1975; 33: 191-197.
16. Mavrodisz K, Ržsa N, Budai M, Sož A, Pap I, Tarján I. Prevalence of accessory tooth cusps in a contemporary and ancestral Hungarian population. *The European Journal of Orthodontics* 2007; 29 (2): 166-169.
17. Kraus, BS. Carabelli's anomaly of the maxillary molar teeth: observations on Mexicans and Papago Indians and an interpretation of the inheritance. *Am J Human Genet* 1951; 3: 348-355.
18. Lee, GTR, Goose DH. The inheritance of dental traits in a Chinese population in the United Kingdom *J Med Genet* 1972; 9: 336-339.
19. Dietz VHA. Common dental morphotropic factor: the Carabelli's cusp. *Journal of the Am Dent Assoc* 1944; 31: 784-789.
20. Salako NO. Taurodontism. *Nig Dent J* 1985; 4: 34-37.
21. Constant D. A review of taurodontism with new data on indigenous southern African populations. *Arch Oral Biol* 2009; 46: 1021-1029.
22. Joseph M. Endodontic treatment in three taurodontic teeth associated with 48 XXXY Klinefelter syndrome: a review and case report. *Oral Surg, Oral Med, Oral Pathol Oral Radiol Endod* 2008; 105: 670-677.