Multiple bilateral supernumerary teeth in a non-syndromic female patient - A case report with a comprehensive review

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INTRODUCTION

Supernumerary tooth (ST) or hyperdontia is defined as the presence of an extra tooth in comparison to the normal dentition. The prevalence varies among various populations; for primary and permanent dentitions is 0.2-0.8% and 0.5-5.3%, respectively. (Saluja et al., 2016) The etiology is not yet completely clear but some of the suggested theories of ST are inclusive of hyperactivity of the dental lamina, atavism, genetic and environmental factors. (Saluja et al., 2016; Guo et al., 2017) Males are affected twice and ST can be seen in either of the dental arch or in both, can be single or multiple, unilateral or bilateral, impacted or erupted, and can be found as an isolated dental finding or associated with syndromes. (Sulabha et al., 2013) Most of the times, they are found as an incidental finding during routine radiographic evaluation. (Saluja et al., 2016) Although they are asymptomatic in most of
the cases but sometimes they can present with various clinical problems like diastema, crowding, rotations, delayed eruption, resorption of the adjacent teeth and cystic lesions. (Saluja et al., 2016) This calls for a proper clinical and radiographic evaluation to make a suitable treatment plan for ST.

Non-syndromic multiple supernumeraries are rare, with the highest occurrence in the mandibular anterior region. (Sulabha et al., 2013) Furthermore, the occurrence of bilateral mandibular supernumerary premolars are uncommon. Therefore, the objective of this paper is to report a rare case of concomitant occurrence of multiple bilateral ST in a non-syndromic 22-year-old female patient with a detailed literature review.

Case report
A 22-year-old female patient reported for a routine dental check-up. The medical and dental histories were insignificant. She had no complaint of pain or any signs of infection and was in good general health.

Clinical examination
The extra-oral examination did not reveal any abnormalities. On intra-oral examination, a generalized plaque was observed along with the pit and fissures caries bilaterally in mandibular first and second molars. ST was noticed lingually between the 1st and 2nd premolars in the right mandibular region, whereas on the left side, it was present between the 2nd premolar and 1st molar. Their root development seemed to be active. Figure 1 Patient-reported a familial pattern where her elder brother had the same anomaly and had them extracted while undergoing orthodontic treatment. The patient was advised radiographic investigation to rule out the impacted ST.

Radiographic features
Radiographic examinations comprised of an orthopantomogram (OPG) that revealed the presence of additional bilateral impacted mandibular premolars. On the right side, ST was noticed just below the 2nd mandibular premolar, whereas on the left side, it was present between the 1st and 2nd premolar. Their root development seemed to be active. Figure 2

So in this patient, there were 4 ST in total; two were clinically evident, whereas the other two were impacted.

Treatment
For ST, the patient was educated about the radiographic findings and was adequately counselled regarding the potential complications and consequences associated with ST, both erupted and impacted. She was advised for extraction of both erupted supernumerary premolars and observation of the impacted ones. The patient refused any treatment related to ST since there was no immediate problem associated with ST.

Regarding plaque accumulation and caries, the patient was advised for oral prophylaxis and restoration of the carious teeth, for which the patient agreed.

Follow-up
Currently, the patient is under regular clinical and radiological examination for ST.

Discussion
Hyperdontia is an odonto-stomatologic disorder that is characterized by the presence of an extra tooth. (Santos et al., 2014)

Prevalence
In the general Caucasian population, the prevalence of ST for the permanent dentition ranges from 0.1-4%, whereas it seems to be more common in Mongoloid racial groups, with a frequency >3%. (Santos et al., 2014) Furthermore, the prevalence of supernumerary premolars is found to be 0.05-0.64% and it represents between 8.0-9.1% of all ST. (Hall and

Figure 1: Intraoral clinical aspect of the patient

Figure 2: Panoramic radiograph showing the presence of supernumerary teeth in mandibular posterior region
There are many categories to classify ST (Garvey et al., 1999) viz, (I) according to the morphology, they are of four types-conical, tuberculate, supplemental, and odontomes; (II) according to the topography, they are classified as mesiodens, paramolar, parapremolar and distomolar; and (III) according to age and chronologically, they are classified as predeciduous, pre-permanent and post-permanent.

According to Stafne, who had conducted the most comprehensive study on ST and one often cited, states that, unlike other ST, supernumerary premolars usually resemble normal premolars in shape and size. (Stafne, 1932) The ST, in this case, seemed to be with normal premolar morphology and belonged to the category of the post-permanent para-premolar variant.

Clinical findings
In the permanent dentition, males are affected twice as frequently as females. (Saluja et al., 2016) Supernumerary premolars are observed in about 10% of cases and nearly 75% of these cases occur in the mandible. Only 1% of non-syndromic cases present multiple ST, which mostly occur in the mandibular arch (61%). (Santos et al., 2014; Solares and Romero, 2004) Analysis for specific locations of ST revealed that there was a predominance of multiple ST in the premolar area (62%) with the highest frequency of occurrence in the mandibular premolar region (45%). (Solares and Romero, 2004)

For the specific site analysis, it was found that ST is usually present lingual to, or occasionally vertically below, the normal premolar teeth. (Panda and Ahmed, 2017) When the ST are asymptomatic, they are detected as a chance finding during radiographic examination. (Solares and Romero, 2004)

The present case is one of the rarest findings as multiple ST were found in a female patient that were not related to any syndrome or systemic disease.

Diagnosis
A proper clinical and radiographic examination is the most suitable way to determine ST. Usually, ST are asymptomatic and are often identified during the routine radiographic examination as an incidental finding. (Garvey et al., 1999) Moreover, the ratio of impacted supernumerary premolars to erupted ones is nearly 5:1, justifying radiographs as the only diagnostic aid.

In recent years, a declining curve is noted in the diagnosis of ST that could be attributed to the fact that many healthcare professionals have reduced the number and frequency of radiographs taken owing to increased public and professional concern over increased public and professional concern over increased public and professional concern.

Classification
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Additionally, multiple ST is less commonly seen as compared to a single ST and is even rarer in the absence of a syndrome or systemic condition. The literature shows single ST account for 76-86% of all cases; double supernumeraries occur in 12-23% of all cases, whereas only in <1% of the supernumerary cases are three or more extra teeth found. (Santos et al., 2014)

Patients with cleft lip and/ or palate have a high prevalence rate for ST (~28%), especially in the anterior region. Gardner’s syndrome, in which multiple ST is a characteristic feature, a high occurrence rate of 21% has also been noted. A high incidence of ST is found in patients with cleidocranial dysplasia, with a combined 35% for both the maxillary and mandibular anterior regions and 27% for the combined premolar regions. A higher prevalence of supernumerary mandibular premolars was also seen to occur in these patients (15%). (Solares and Romero, 2004)

Origin and inheritance
The etiology of this anomaly is not fully understood even though many theories like phylogenetic reversion theory (atavism), hyperactivity of dental lamina and division of developing tooth bud to form two teeth are being proposed for its occurrence. (Marya et al., 2012) Although the theory of hyperactivity of the dental lamina is the most widely accepted theory that states that initiation of rests of dental lamina leads to the development of an extra tooth bud that manifests itself as an ST (Marya et al., 2012) but since many times supernumeraries are part of a syndrome, so there are lots of other theories that suggests the formation of ST being influenced by the genetic and environmental factors. (Açıkgöz et al., 2006)

As for the hereditary factor, it has been postulated that this anomaly does not follow a simple Mendelian pattern of inheritance since only a few supernumerary premolars among siblings are documented. A sex-linked mode of inheritance has been suggested as ST in the permanent dentition are twice as common in males as compared to their female’s counterparts. (Hall and Onn, 2006)
Complications

Many complications are associated with ST, like functional impairment, malalignment of teeth, unaesthetic appearance and less commonly, development of cysts and root resorption of adjacent teeth. (Saluja et al., 2016) Of all the aforementioned complications, the most troublesome is the association of ST with interference in normal occlusal development or with orthodontic mechanics such as crowding, impaction or delayed eruption of permanent teeth, rotations, malocclusion, abnormal eruption sequence, retained primary teeth, compromised space closure, and interference with root torque that is very rarely seen. (Solares and Romero, 2004; Bodin et al., 1978) Additionally, bilateral multiple supernumerary premolars are also found associated with odontogenic keratocyst or in close vicinity of permanent teeth. (Kasat et al., 2012; Bhardwaj et al., 2012) None of these problems were associated with the ST found in the present clinical scenario.

Treatment

The treatment of ST is still a debatable topic since many authors recommend the prophylactic removal of asymptomatic ST (Solares and Romero, 2004), whereas other suggests that asymptomatic ST should be untouched until the development of the adjacent anatomic structures.

Still, others propose that ST should only be removed if it is associated with any of the previously mentioned complications. However, regular clinical and radiographic follow-up is needed if the risks of surgery outweigh the benefits of removal. (Garvey et al., 1999)

Marré (1940) and Hanratty (1939) recommend surgical removal of supernumerary premolars in 2 stages:

1. Extraction of more developed supernumerary premolars is accomplished soon after diagnosis;

2. The remaining less-developed premolars are untouched and removed later after their root development is finished in order to avoid damage to adjacent structures and allow for bone regeneration.

Becker et al. (1982) suggested the late removal of supernumerary premolars in order to remove normal deciduous, permanent and supernumerary premolars in one session that considerably reduces the treatment time along with no damage to adjacent structures and psychological distress to the patient as well.

As the supernumeraries in the present case were not associated with any complications, it was decided to simply monitor the ST.

Recurrence and follow-up

The recurrence rate of supernumerary premolars after being surgically extracted has been found in 8% of the cases reviewed that is often hypothesized to be due to the reactivation of a portion of a follicle. (Solares and Romero, 2004) Another possible reason stated is that, in these patients, the dental lamina is incompletely resorbed which gets reactivated during the crown completion of normal permanent teeth, leading to the formation of multiple ST, especially in the premolar region.

This is the same mechanism found in patients with cleidocranial dysplasia. (Solares and Romero, 2004) All these scenarios and the related complications make the periodic follow-up of such patients an extremely important and mandatory step. (Sulabha et al., 2013) Owing to the above-mentioned facts, the patient is recalled every 6-months for the follow-up to avoid any complications.

CONCLUSION

This case is unusual as the bilateral occurrence of multiple supernumerary mandibular premolars is a rare event. A detailed history and clinical examination along with a thorough investigation for an early diagnosis and appropriate treatment of ST are mandatory. Symptomatic supernumeraries should be treated carefully and those without symptoms should be followed up for the long term. The unique features, in this case, include the symmetrical presence of multiple ST in the mandibular premolar regions with a resemblance to the normal premolars and without association with any syndrome.

Clinical significance

Routine, conventional radiographs as the diagnostic aid are justified, as shown in this case since supernumerary teeth are usually asymptomatic. Furthermore, the patient needs to be informed and counselled regarding the complications related to untreated supernumerary teeth.

Conflict of Interest

The authors declare that they have no conflict of
interest for this study.

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