INTERNATIONAL JOURNAL OF PAEDIATRIC DENTISTRY

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Editor-in-Chief Göran Dahllöf

Abstracts of the 22nd Congress of the International Association of Paediatric Dentistry Munich, Germany, 17–20 June 2009



Volume 19 – Suppl. 1 June 2009

The official journal of The International Association of Paediatric Dentistry The British Society of Paediatric Dentistry



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Oral Sessions O10 - Dental Anomalies 2

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Oral Session O10/Dental Anomalies 2 – Syndromes and Genetics 1

O10-72

Secondary retained molar with clinical, radiological, histological, immunohistochemical and SEM studies V. ROY¹, I. JAMAZI² & S. GHOUL-MAZGAR³

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Introduction: A 12-year-old girl presented a secondary retained left mandibular first molar, pink in colour, with neither carious lesions nor restorations. The patient was in good health and had no history of congenital anomalies, trauma or inflammation of the relevant molar area.

Clinical management: The vitality test and the sound on percussion were normal. Orthopantomogram showed an absence of the periodontal ligament space in the area of bifurcation with apparent blending between the root and the alveolar bone. The pulp cavity showed radiolucent and calcified areas. Roots exhibited dilacerations and rounding of the apexes. After extraction, the coronal part of the tooth underwent histological and immunohistochemical analysis using PCNA (proliferation) and p53 (apoptosis) antibodies, root surfaces were observed using scanning electron microscopy (SEM). Ankylosis was observed in the bifurcation and the pulp cavity showed bone lining the dentin with a mostly lamellar aspect and empty osteoblasts. The rarely observed osteocytes were apoptotic. Dentin showed figures of aposition and resorption with several lacunae occupied by multinucleated giant cells. Fibrous pulp contained mononuclear proliferating cells. The vascular proliferation was proved immunohistochemically. Roots observed with SEM were free of lacunae.

Conclusion: The pink colour of the retained molar reflects the subjacent resorbing process and the proliferation of pulp vessels. Although odontoblasts showed an attempt at healing, osteocytes had undergone apoptosis. As the root surfaces were free of resorption lacunae and the ankylosis figures demonstrated into the pulp chamber, it seems that the pathology of this case is mainly due to pulp origin.

010-73

Unusual tooth malformation involving the permanent mandibular incisors: a case report

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Introduction: Tooth malformations involving the crown or root in permanent teeth, may be caused by various factors that interfere with normal tooth development, such as traumatic injury of the primary teeth, genetic mutations and local inflammatory, hamartomatous or neoplastic processes.

Clinical management: We report a case of an 8 year old Caucasian girl who applied to our department with her parents because they were concerned with the aesthetics of her front teeth. Her past

medical history and family history were non-contributory. She has had sporadic and limited dental care, but no records or past radiographs were available to us. The patient and her family did not recall any relevant trauma history before eruption of these teeth. Upon clinical examination, teeth #32, 41, 42 exhibited exophytic malformations of white and yellow colour. The malformations appeared as hard tissue protuberances of the crowns and the cervical areas of the roots, both labial and lingual. Dental CT revealed that these protuberances were continuous to the tooth structure and most likely contained no pulpal tissue. Hard tissue biopsy revealed a non-homogeneous structure resembling both enamel and dentin by SEM and histology. The protuberances (exophytic malformations) were finally trimmed off and the teeth were covered with a layer of opaque resin composite. No pulpal sensitivity or other complains have been experienced at 18-month follow-up.

Conclusion: The possible aetiology and diagnosis of this case is open to discussion based on the presented review of relevant literature.

O10-74

Dental ankylosis and aplasia of successor teeth K. SALEM¹, B. MIRZAEE² & T. MOHTAVIPOOR³

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Introduction: Dental ankylosis is a common disturbance of tooth eruption. The purpose of this study was to evaluate the prevalence of primary molar ankylosis and associated anomalies including missing of successors in a group of Iranian children.

Patients and methods: 1023 school children between 7 and 9 years in the city of Rasht were screened for dental ankylosis. Periapical radiographs were then taken from 105 subjects with ankylosis and their matched controls. Follow-up examination took place after 2 years to determine other anomalies in the permanent dentition. Chi-square test was used for data analysis.

Results: The prevalence of ankylosis was 15% (13.4% in males, 16.6% in females). More than 60% had more than one ankylosed tooth and the most common site was lower first primary molar. Missing of permanent premolars was observed only in second premolars and in 0.02% of total teeth but none of the controls (NS). Other observed anomalies were hypoplastic defects on central incisors, macrodontia of upper incisors, and ectopic eruption of upper canines (23 subjects *vs* 5 controls, (P < 0.05). The radiographic examination confirmed ankylosis in 10.03% of the cases. Spontaneous mobility or exfoliation occurred in 98% of first primary molars during the next 2 years.

Conclusion: Ankylosis may be a predictor of other dental anomalies such as ectopic eruption of upper canines. It is advised to make a thorough clinical and radiographic examination in cases of observed dental ankylosis.

O010-75

A child-centred approach to seeking children's experiences of cleft lip and palate

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Introduction: Children and young people can provide valuable insights into their experiences of conditions and medical treatment. The aim of this research was to explore children's accounts of being born and living with a cleft lip and/or palate (CLP) and related treatment.

Patients and methods: Children and young people, under the care of the CLP multidisciplinary team at Sheffield dental hospital, UK, were invited to take part in this qualitative study. A child-centred research approach was taken incorporating interviews and activities such as timelines. Interviews were usually carried out in children's homes, audio-taped and transcribed verbatim. Data were analysed using a biographical approach to life stories.

Results: Eight boys and nine girls, aged 8–16 years, with a range of cleft types, each took part in two interviews. The first focused on their general life stories, although these often incorporated a discussion about CLP. A follow-up interview explored specific aspects of the condition and its treatment. 'Cleft stories' emerged which detailed the ways in which the children made sense of the condition. Since CLP-related interventions frequently commence in infancy, there were many aspects that children could not remember themselves. Accordingly, cleft stories were composed of a fascinating interplay of both children's and adults' recollections.

Conclusion: These data indicate the persistent, but not always significant, part that CLP plays in children's biographies. It demonstrates the constructed nature of cleft stories, and that adults occupy a key role in this construction, shaping their child's understanding by their own experiences and perspectives.

O10-76

Ectrodactyly with ectodermal dysplasia: dental and radiographic implications

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Introduction: EEC syndrome is a rare, genetic disorder of ectoderm - mesoderm origin exhibiting variable phenotypic expression. The dental status and management of a boy aged 3 years with severe ectrodactyly of the hands and feet paired with ectodermal dysplasia has been assessed and documented.

Clinical Management: The diagnosis of the EEC syndrome (ectrodactyly ectodermal dysplasia and clefting syndrome) was confirmed by molecular investigation. The affected boy had a full complement of primary teeth with severe dental caries, several anterior periapical abscesses, tooth mobility and enamel hypoplasia. He did not have labial or palatinal clefting. The dental implications of the condition have been reviewed with emphasis on the role of radiography as an adjunct for the determination of indications for dental management. The patient received endodontic treatment of several primary teeth, followed by the placement of anterior ceramic crowns under general anaesthesia. Because of the oligodontia in the permanent dentition it was decided to fully rehabilitate the primary dentition, where in normal circumstances a different treatment option could have been indicated. A week after the treatment the boy presented for a follow-up evaluation, all anterior peri-apical abscesses had now disappeared and mobility was zero. Routine three monthly assessment visits are to be scheduled.

Conclusion: The dental management has been tailored for this child where enamel hypoplasia, severe dental caries and oligodontia are additional complications. The importance of the diagnostic nature of dental radiographs as an adjunct to the treatment of patients with oligodontia and ectodermal dysplasia is emphasized.

O10-77

IgA secretory and Lysozyme concentration in whole saliva of patients with Prader Willi Syndrome

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Introduction: Prader Willi syndrome (PWS) is a rare, genetic disorder (1:15,000). It is a deletion in the 15 q11–q13 region of the father's chromosome. As a consequence of hypothalamus alterations there are typical characteristics as: hypotonia, hypogonado-trophic hypogonadism, mental retardation, central obesity, typical facies, xerostomic, thick and viscous saliva and amelogenesis. Brown spots were observed in the vestibular enamel teeth which lead to caries. In previous studies we have described alterations on the concentration of inorganic ions in the whole saliva. The aim of this study was to describe the concentration levels of IgA secretory and lysozyme in whole saliva within a group of patients with PWS.

Patients and methods: Eight Patients with PWS between 6–18 years of age were analysed and 8 patients in good health as a control group. The samples were collected according to the international protocol that will be described during the presentation.

Results: High IgA values were observed in patients with PWS in comparison with the control group: 37.6 mg/dl and 18.5 mg/dl accordingly. The levels of lysozyme were diminished in comparison with the control group 0.53 and 0.18 accordingly.

Conclusion: The increased levels observed in the concentration of IgA and the decrease in the concentration of lysozyme could be favourable factors for the occurrence of caries-free zones in patients with PWS:

O10–78

A novel DLX3 mutation associated with tricho-dento-osseus syndrome (TDO)

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Introduction: DLX3 is a member of the DLX homeobox genes. As in the case of other homeobox genes, DLX3 homeodomain is able to bind to its cognate DNA sequences and regulate transcription of its target genes. Mutations in DLX3 are associated with TDO. We report a novel mutation in the DLX3 gene. Informed consents and ethical committee agreement were obtained.

Patients and methods: The proband was examined clinically at the age of 1.5 year. Panoramic tomogram (ptg) was taken at the age of 4. Clinical and radiographic records (ptg, cephalometric and wrist radiogram) were available for her mother and maternal grandfather. Primary teeth of the child, extracted because of infection, were examined histologically. Swab samples from buccal mucosa were taken for the mutational analysis.

Oral Presentations

Results: The main features were typical for TDO: sparse colourless hair, increased bone density and thickness in some parts of the cranium, and abnormal teeth. Enamel was thin. Molars were taurodontic and their pulps contained unorganized hard tissue. The roots were short and dysmorphic. Histological analysis revealed abnormal enamel and dentin. Spontaneous infections developed in primary and permanent teeth soon after eruption.

Sequencing of the DLX3 coding regions revealed a heterogeneous C > T transition in the homeobox in the proband, mother and grandfather but not in healthy family members or 100 controls. **Conclusion:** A novel mutation in affected family members exclusively strongly suggests its causative role in TDO. By disrupting the highly conserved homeobox sequence the mutation interferes with the DNA-binding ability of the DLX3 protein.

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