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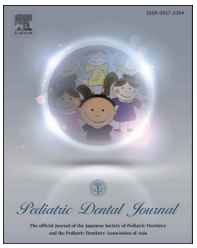

Case Report

"Life with an incomplete bite" – Preventive oral care and findings of a child with pre-existing 'Poland syndrome'

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ABSTRACT

Background: Poland syndrome is a rare syndrome named by Patrick Clarkson in 1962 and described by Sir Alfred Poland in 1841.
Case report: The dental anomalies with pre-existing poland syndromic child were noticed by the parents during child's regular eating-chewing more obviously, after the appearance of his primary teeth.
Follow-up: The patient is under oro-myofunctional therapy with other strict preventive regimen and is still being monitored every 6 months since last 2 years.
Conclusion: Specific attention must be paid to children with pre-existing conditions with preventive measures and implementation of muscular training very early in life.

1. Background

One of the early masticatory dysfunction signs is mandibular mobility model disorder. Symmetrical muscular work is one of the most important initiators conditioning the proper functioning of the masticatory system. Lack of muscle activity is just as harmful to the stomatognathic system components as their hyperactivity as in eg, bruxism. This may lead to shortening of muscle fibers, in extreme cases even to muscle atrophy [1].

Poland Syndrome is a rare congenital anomaly classically consisting of the combination of unilateral aplasia/hypoplasia of the sternocostal head of the major pectoral muscle and ipsilateral brachysyndactyly [2–4]. Other usual anomalies in Poland Syndrome are malformations of the anterior chest wall and breast [5]. Poland Syndrome is sometimes referred to as Poland sequence because sequence refers to a pattern of malformations derived from a single anomaly [6].

According to some investigators, the primary defect in Poland Syndrome may be impaired development of a certain artery or other mechanical factors that may result in diminished or interrupted blood flow during early embryonic growth. The term "Subclavian Artery Supply Disruption Sequence" has been suggested for a group of conditions that may occur due to disruption of blood flow through particular arteries (i.e.,

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subclavian artery, vertebral artery, and/or their branches) at or around the sixth week of embryonic development. Such conditions include Poland Syndrome, Moebius Syndrome, Klippel-Feil Syndrome, and Sprengel deformity [7]. In fact, associations with Moebius Syndrome, facio-auricolo-vertebral dysplasia and fronto-nasal dysplasia have been described [8].

In the specialized literature so far, there is no association listed about the co-existing conditions of the oro-facial muscular atrophy or oro-muscular dysfunction with Poland syndrome.

In the present case report the masticatory dysfunction syndrome co-existing with Poland syndrome were noticed. Oro-facial anomalies, the dysfunctional mastication as well as oro-motor disability and abnormal stereognosis in the child with a history of pre-diagnosed Poland syndrome during his infancy were evaluated. The findings maybe subtle, but hypoplasia of oro-facial skeleton along with hypotonicity and shortening of masticatory muscles should alert the pediatric dentists and impose instillation of myofunctional therapy.

2. Case report

2.1. *Medical history*

An eight-year-old boy pre-diagnosed by the team of medical professionals with Poland syndrome post-natally, was first seen at the pediatric dental set-up two years back. He was an outcome of full-term gestation and pregnancy was not supervised. The pregnancy and delivery were not adversely eventful. The child was the first and the only son of the couple in a well-educated nuclear family setting of a non-consanguineous marriage. There was no family history of congenital anomalies. The mother complained a history of poor 'latch-on' and suckling reflexes during breast-feeding. Developmental milestones were normal.

2.2. *Physical examination*

Physical examination reveals that his whole of right side body looks smaller compared to his left side body, though the child has a good mental status and IQ levels.

His right hand and right leg exhibit detectable hypoplasia of the phalanges of the ring fingers, thumbs and toes, resulting in abnormally short fingers (brachydactyly). In addition, there was webbing (syndactyly) of the ring and little fingers. His right arm was slightly shorter than the other arm. His chest X-ray reports right anterior chest wall deformity [Fig. 1].

He has normal cardiovascular system. Weight, height and the remainder of the findings were normal.

The child's behavior on the rating scale can be graded as Frankl's definitely positive.

2.3. *Oral clinical examination*

Unilateral facial paralysis was over-ruled by checking for pupillary and corneal reflexes which were normal. Jaw opening was assessed by three finger test.

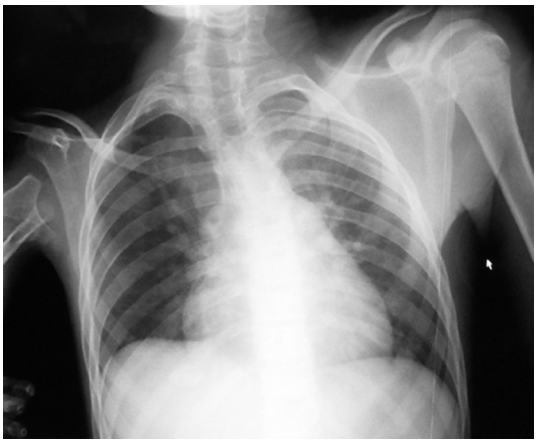

Fig. 1 – Chest X-ray showing right anterior chest wall deformity.

2.3.1. *Hard tissue findings*

Mixed dentition with erupting 16, 26, 36, 46 and 31, 41. There was absence of dental caries on any of the teeth. The occlusion of primary molars was of flush terminal plane. Deep bite was noticed causing a slight trauma to the erupting 31 and 41 teeth.

2.4. *Radiographic examination*

The gonial angle, condylar heads, skeletal pattern of growth all seemed to normal on the left side and the right side exhibiting mild hyploplasia [Fig. 2].

2.5. *Findings in masticatory ability and performance and efficiency*

Nordic Orofacial testing questionnaire was given to the parents to assess the child's chewing and swallowing pattern. The child exhibited unilateral chewing habit, exclusive with left side.

2.5.1. *Masticatory efficiency*

Initially the soft food such as peanut and hard food such as raw carrot was given to the child to chew upon. As these tests seemed vague it was decided to determine the efficacy by analyzing fuchsine concentration in a solution obtained from chewed granules measuring the absorbance at 546 nm in a calorimetric Spectrophotometer. It was concluded that masticatory time was 32 s and the masticatory cycle frequency was very slow.

2.5.2. *Oral stereognostic ability*

Due to impaired motor activity of the tongue, the Oral stereognostic ability was observed as improper. The various forms used in this study were: square shape form, triangle shape form, star-shaped form and circle shape form. The test forms were fabricated from heat cured acrylic resin by conventional technique. The test forms used were 2–3 mm in length. For identification purpose, similar but 5–6 times oversized test forms were fabricated paper cardboard and displayed in front of the child.

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