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Orthodontic Treatment of an Adult Patient with Left-Sided Cleft Lip and Palate and a Congenitally Missing Lateral Incisor

Leczenie ortodontyczne pacjenta dorosłego z lewostronnym rozszczepem wargi i podniebienia oraz wrodzonym brakiem zęba siecznego bocznego

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A – concept; **B** – data collection; **C** – statistics; **D** – data interpretation; **E** – writing/editing the text; **F** – compiling the bibliography

Abstract

Persons affected with cleft lip and/or palate require interdisciplinary treatment from birth to adolescence. The functional, aesthetic and psychosocial consequences of the condition affect both the patients and their families. The condition often reduces the patients' quality of social life and burdens them with psycho-emotional issues. The present paper discusses conservative treatment of an adult patient with a left-sided cleft of the lip, the alveolar process and the palate as well as hypodontia of tooth 22. Treatment was conducted with low-friction fixed appliances and a palatal expander. The duration of active treatment was 24 months. In the course of therapy, the patient underwent a surgical reduction of the oronasal fistula. Space was regained for missing teeth, which were restored with fixed prostheses following the completion of active orthodontic treatment (**Dent. Med. Probl. 2013, 50, 1, 96–105**).

Key words: cleft palate, interdisciplinary treatment, surgical closure of oronasal fistula, low-friction system.

Streszczenie

Osoby dotknięte rozszczepem wargi i/lub podniebienia od chwili narodzin do osiągnięcia dojrzałości wymagają leczenia interdyscyplinarnego. Czynnościowe, estetyczne oraz psychospołeczne konsekwencje tego zaburzenia mają wpływ zarówno na samych pacjentów, jak i na ich najbliższych. Choroba często objawia się obniżeniem jakości życia społecznego oraz problemami natury psychoemocjonalnej osób dotkniętych rozszczepem. Przedstawiono zachowawczy sposób leczenia dorosłej pacjentki z lewostronnym rozszczepem wargi, wyrostka zębodołowego i podniebienia oraz hipodoncją zęba 22. Leczenie było prowadzone aparatami stałymi cienkołukowymi w systemie niskiego tarcia wraz z aparatem grubołukowym do ekspansji szczęki. Leczenie aktywne trwało 24 miesiące. W czasie terapii wykonano zabieg chirurgicznego zmniejszenia przetoki ustno-nosowej. Odtworzono przestrzenie dla brakujących zębów, które zostały uzupełnione stałymi pracami protetycznymi po zakończeniu aktywnego leczenia ortodontycznego (**Dent. Med. Probl. 2013, 50, 1, 96–105**).

Słowa kluczowe: rozszczep podniebienia, leczenie zespołowe ortodontyczno-protetyczne, chirurgiczne zamknięcie przetoki nosowo-ustnej, system niskiego tarcia.

A complete cleft of the lip and palate is the most frequently occurring congenital malformation of the head and neck region and is the result of an incomplete development of the lip and/or palate [1–4]. It occurs more frequently in males and is more likely to affect the left side [1].

The cleft arises between the 5th and 12th week of gestation, resulting from a lack of union between tissues of the lip and/or palate [2]. Many factors can affect normal embryonic development and induce the emergence of a cleft [4].

The developmental anomalies involve both the soft and the hard tissues. Severe cases may impair breathing, feeding, speech and hearing. The condition may also seriously affect facial aesthetics [1, 5].

Patients with cleft lip and palate display a three-dimensional maxillary deficiency caused by an embryologic defect of the oral cavity, palate and/or pharyngeal tissues. Dental development is also affected by multiple anomalies such as: supernumerary teeth or agenesis, hypoplastic or dysmorphic teeth, impaction, rotation or ectopic eruption of teeth. Lateral incisors are missing in 10–20% of patients with deciduous dentition and 3–50% of those with permanent dentition. The most frequent abnormalities are malocclusions, which are both skeletal and dental in nature. These can include posterior crossbite, rotated maxillary incisors, lingually inclined or positioned incisors, excess space for lateral incisors in the cleft area, midline deviation and asymmetries [6].

Studies show differences in the prevalence of clefts, which correlate with race, geography and gender [7, 8]. Conversely, no correlations were found for prevalence and socioeconomic, historical or seasonal factors [7].

The literature suggests that the world occurrence of clefts ranges from 0.8 to 1.7 per 1000 live births [2–5, 8]. The highest rates are found in Asian populations, the lowest in African Americans [7–9].

Cases of cleft lip with or without cleft palate occur more frequently in males, while isolated cleft palate is more likely in females [7–9]. Boys develop more severe forms of clefts than girls [9].

Historically, classifications of oral clefts were numerous. Currently, a widely used classification is one by Kernahan and Stark, published in 1958.

As a result of many studies it has been found that the aetiology of clefts is multifactorial with a varying genetic and environmental impact [4, 9]. Heredity is the main causative factor.

Clefts of the primary and/or secondary palate are an element of over 200 specific genetic syndromes. Isolated palatal clefts make up a component of over 400 syndromes. Syndromic clefts comprise between 5 and 7% of all clefts [7].

Since the origins and the timing of the development of the primary and secondary palate are distinct, the clefts of these structures can be divided into isolated cleft of the primary palate, isolated cleft of the secondary palate, and combined cleft of the primary and secondary palate.

Genes play an important role in facial development; however, environmental factors seem equally important.

Many factors have been shown to contribute to the risk of cleft development. They include maternal exposure to: tobacco smoke, medication (anaesthetics, chemotherapeutics, anticonvulsants, steroids and interferon), alcohol, nutrient deficiencies (folic acid, group B vitamins, zinc), viral infections, environmental pollutants (e.g. agricultural chemicals), hyperthermia, X-rays and ionising radiation.

The best research data are available for detrimental effects of the first four teratogenic factors [4, 7, 9].

Case Report

Diagnosis

Patient A.U., aged 32, presented at our orthodontic office with the complaint of "unacceptable dentition". The patient was found to have had a complete left-sided cleft of the upper lip, the alveolar process and the palate. History revealed that the defects of the upper lip and palate were surgically repaired in infancy, albeit the methods of treatment and their sequence remain unknown due to missing documentation.

The patient's features were characterised by a flattened tip of the nose, a surgical scar of the left part of the vermillion, an altered geometry of Cupid's bow as well as a collapsed subnasal region (Fig. 1).

An intraoral exam revealed a narrow maxilla; bilateral partial posterior crossbite; hypodontia of tooth 22; prior extractions of teeth 16, 26, 36, 46; a dentoalveolar leftward shift of the maxillary midline by 3 mm with a marked angulation of the teeth towards the cleft; an upward cant of the left sector of the maxillary occlusal plane; pathological abrasions of teeth; an oronasal fistula; and a thin bony biotype in the area of the lower incisors (Fig. 2). The patient also had hypernasal speech.

Her panoramic X-ray revealed an endodontically treated tooth 27 and tooth 47 treated by amputation as well as a mildly reduced alveolar bone level (Fig. 3).

Her lateral skull X-ray displayed a retrognathic profile, skeletal Class 1, a tendency to skeletal open bite, as well as retroclined upper and lower incisors.

Treatment Goals

I. Face: to increase the projection of the subnasal area and to increase the surface area of the vermillion.

Fig. 1. Pre-treatment extraoral photographs

Ryc. 1. Fotografie zewnątrzustne pacjentki przed leczeniem

Fig. 2. Pre-treatment intraoral photographs

Ryc. 2. Zdjęcia wewnątrzustne przed leczeniem

Fig. 3. Pre-treatment panoramic X-ray

Ryc. 3. Zdjęcie pantomograficzne sprzed leczenia

II. Oral cavity:
- to improve size, form and proportion of dental arches,
- to normalise overbite and overjet,
- to render dental and facial midline coincident,

Fig. 4. Intraoral photographs on the day of bonding

Ryc. 4. Zdjęcia wewnątrzustne w dniu montażu aparatów stałych

Fig. 5. Treatment course between month 3 and month 7

Ryc. 5. Przebieg leczenia między 3. a 7. miesiącem

- to correct crossbites,
- to prepare conditions for prosthetic rehabilitation of teeth 16, 22, 34', 46,
- to restore normal sizes, proportions and shapes of upper incisors,
- to improve shapes of lower incisors and eliminate paragingival black triangles,
- closure of the oronasal fistula,
- speech rehabilitation.

Appliances and Methods Used

1) upper and lower thin archwire fixed appliance (Damon Mx),
2) banded Hyrax screw for the upper arch,
3) transpalatal arch,
4) disocclusion,
5) upper incisor build-up with composite,
6) stripping of lower incisors,
7) masking veneer for tooth 22,
8) surgical closure of oronasal fistula,
9) retention,
10) post-orthodontic prosthetic rehabilitation,
11) speech-language therapy,
12) vermillion geometry correction with hyaluronic acid filler.

The plan to enlarge the envelope of the upper arch and to upright the roots of the teeth adjacent to the cleft put the patient at risk of fistula widening as well as sustaining gingival recessions due to the presence of a thin bony biotype in the lower arch. Due to the patient's age and the size of the cleft, the prognosis regarding complete closure of the fistula was guarded. Having been informed of the potential risks and foreseeable complications, the patient accepted the proposed management plan.

Treatment Time

The time of active orthodontic treatment was estimated at 24 to 36 months. The time of retention splint wear was planned for 2 years.

Course of Treatment

Following interproximal reduction of the lower incisors, we bonded an upper and lower low-friction fixed appliance along with a Hyrax screw mounted on rigid wires.

For the first two months, the patient was treated with CuNiTi thin round archwires (Fig. 4). She was instructed to perform slow activation of the Hyrax screw (once every 2 days).

Between the third and seventh month of treatment, we applied rectangular arches, gradually increasing their thickness (Fig. 5). We focused on regaining space for tooth 22 and levelling the occlusal plane with vertical elastics. The space for tooth 22 was masked with a composite pontic bonded to tooth 21. In the meantime, space was being regained for teeth 16 and 34', as planned. Owing to pulpitis of tooth 47, we abandoned the plan to regain space for tooth 46.

In accordance with our concerns, gingival recessions emerged at an advanced stage of space regaining for teeth 22 and 34'. This resulted from the flaring of upper and lower incisors as well as the angulation of roots 21 and 23 towards the cleft. This process tended to progress (Fig. 6).

Fig. 6. Recessions emerging around teeth 21, 23, 32

Ryc. 6. Recesje dziąseł przy zębach 21, 23, 32

By the ninth month of treatment, we reconstructed the shape, size and proportions of the patient's upper incisors. The widening of the crowns became feasible thanks to the space gained by their flaring (Fig. 7).

The surgical closure of the residual palatal fistula was performed by raising and elongating a mucoperiosteal flap (Fig. 8). Healing of the wound was uneventful. In effect, the size of the fistula was significantly reduced and the patient's comfort improved.

By the 24th month of treatment, we debonded the patient and initiated the retention phase as well as the post-orthodontic prosthetic rehabilitation.

Owing to pulpitis of tooth 47 and associated complications, we decided to debond the patient despite the fact that her lower roots had not been positioned parallel to each other. The decision was also prompted by the finding of early apical resorptions of lower incisors and of tooth 12 (Fig. 9).

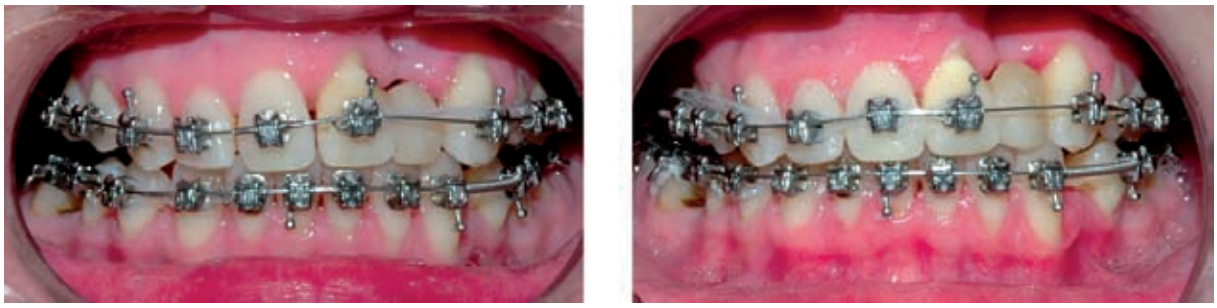

Fig. 7. Preparation for the restoration of shape and size of the upper incisors and the completed composite build-ups

Ryc. 7. Przygotowanie do odbudowy kształtu i wielkości zębów siecznych górnych oraz rekonstrukcje z materiału złożonego

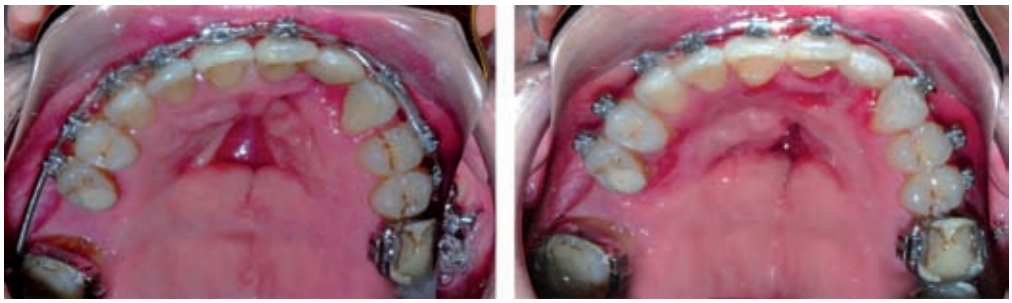

Fig. 8. Photographs of the residual opening before and after surgical management

Ryc. 8. Otwór resztkowy przed i po zabiegu chirurgicznym

Fig. 9. The presence of apical resorption of the lower incisors and of tooth 12; a deepening bony pocket around tooth 47

Ryc. 9. Cechy resorpcji szczytów korzeni zębów dolnych siecznych oraz zęba 12, pogłębiająca się kieszonka kostna przy zębie 47

For retention, we applied a bonded retainer made of braided stainless steel wire to the upper central incisors and an Erkodur retention splint with a masking veneer for tooth 22 as well as a similar lower retention splint with a masking veneer for tooth 34'.

One month after debonding and the stabilisation of occlusion, the patient received final prosthetic restorations. In the upper arch, tooth 22 and the bony hiatus were restored with a Maryland bridge made up of a porcelain crown and fibreglass wings bonded to teeth 11, 21 and 23 as well as a gingival replacement made of pink porcelain (Fig. 10). It should be underscored that the fibreglass wings were not only a source of support for the bridge itself but also a fixed retention element for the anterior dentition.

In the mandibular arch, the space regained for tooth 34' was filled by an adhesive bridge, with a design conceived to spare the abutment teeth. The bridge was supported by a fibreglass element bonded to tooth 33 and an inlay anchorage in an appropriately prepared caries lesion on the occlusal surface of tooth 34 (Fig. 11).

Treatment Results

The records taken following the completion of treatment showed that the majority treatment goals had been achieved (Fig. 12). Features were improved, as analysed in several views. The projection of the subnasal area was enhanced, the oral aperture widened and the size of the vermilion enlarged.

The achieved intraoral changes included intercuspidation of posterior teeth; enlargement of the dental arch envelope; normal forms and proportions of arches; normal overbite and overjet; coincident dental and facial midlines; crossbite correction; restoration of normal sizes, proportions and shapes of upper incisors; improvement of the shapes of lower incisors; elimination of perigingival dark triangles; and oronasal fistula reduction. The planned space gaining for teeth 16, 22, 34', 46 was only partly achieved due to pulpitis of tooth 47 and associated complications as well as early apical incisor root resorption. Due to the necessary early debonding of the patient, roots of the lower molars were not made parallel. The patient was referred for speech-language rehabilitation; however, we lack information whether she continues to attend sessions and what the results are.

Fig. 10. Intraoral frontal and upper occlusal views before and after ortho-prosthetic management

Ryc. 10. Zdjęcia zgryzu *en-face* oraz rzut łuku górnego przed i po leczeniu ortodontyczno-protetycznym

Fig. 12. Facial photographs before and after ortho-prosthetic management

Ryc. 12. Zdjęcia twarzy przed i po leczeniu ortodontyczno-protetycznym

Fig. 11. Intraoral occlusal views of the lower arch before and after ortho-prosthetic management

Ryc. 11. Zdjęcia łuku dolnego przed i po leczeniu ortodontyczno-protetycznym

Table 1. Comparison of pre- and post-treatment cephalometric indices

Tabela 1. Porównanie wyników badań cefalometrycznych przed i po leczeniu

Parameter (Wskaźnik)	Reference (Norma)	Pre-treatment (Przed leczeniem)	Post-treatment (Po leczeniu)
1+:SN (°)	104.0 ± 6.5	84.1	96.0
1+NPg (mm)	7.0 ± 2.5	1.0	4.2
1+:1– (°)	127.0 ± 8.5	154.6	131
1–:ML (°)	94.0 ± 7.0	79.0	90.9
1–APg (mm)	3.0 ± 2.0	–1.4	1.8

The positions of incisors changed, as reflected by cephalometric indices (Table 1).

Discussion

Contemporary cleft management programs emphasise interdisciplinary care provided by appropriately trained teams. These include paediatric surgeons, plastic surgeons, maxillofacial surgeons, paediatricians, orthodontists, otorhinolaryngologists, speech-language therapists, psychologists and social workers.

According to current trends, clefts are surgically closed as soon as possible. The continuity of the lip is restored at the age of about 3 months and that of the palate in the first half of the second year of life.

A recent paper reports a pioneering prenatal cleft closure, which evidences strides in surgical technique.

A three-dimensional underdevelopment of the maxilla is usually reflected by a crossbite involving one or more teeth on the cleft side. This phenomenon is mainly caused by the scarring, which follows surgical closure of the cleft lip and or cleft palate. The scar has a constricting effect, especially given the fact that it acts on a minor segment of the alveolar process [1]. According to modern surgical procedures, clefts of the hard palate are closed only partly. The restoration of full continuity may lead to scarring, which adversely affects the middle portion of the face [2]. The quality of surgery, assessed chiefly in terms of sparing technique, is critical for the success of cleft treatment [1].

The hallmark of our patient's case was a hypoplastic maxilla, manifest by the bilateral posterior crossbite, which we treated with a banded Hyrax screw. By this approach, we succeeded in enlarging the envelope of the upper dental arch.

Present-day practice tends to depart from concepts based on many years of removable appliance wear for the treatment of patients with cleft lip and palate. Factors, which deter clinicians, include imprecise treatment effects and the need for patient compliance. These factors lead to low efficacy and prolonged treatment time. Management using thin- and thick-archwire appliances is recommended at the stage of early permanent dentition, directly before the eruption of the permanent canines. The orthodontic treatment can be combined with an autologous bone graft placed in the cleft.

If the graft is placed before the canine erupts, the tooth has an opportunity to traverse the transplanted bone. Thus, the canine may gain good periodontal support and the alveolar bone perimeter will approximate normal size. If graft placement is delayed to the time point where the eruption dynamics of the canine subside, the transplanted material is usually resorbed, hence, the procedure fails [8].

Patients with palatal clefts suffer from middle ear disease, and difficulties associated with eating, speech abnormalities, oronasal fistulae, dental abnormalities and aesthetic defects in their facial appearance. Maxillofacial surgeons can intervene to reconstruct deformed tissues, close oronasal fistulae, restore appropriate Eustachian tube function, improve swallowing and speech, as well as correct occlusion [2].

Current guidelines on cleft treatment in young patients recommend the early application of distraction methods to the maxilla, rather than conventional orthognathic procedures. This approach is recommended for patients with severe skeletal deformities. In such cases of moderate or severe maxillary hypoplasia, correction is achievable using complete or incomplete LeFort I osteotomy combined with distraction. Distraction osteogenesis is a biological process of new bone formation between two separated bony fragments. The process takes place gradually under the continuous pull of a distraction appliance. Thus, new bony tissue is formed systematically, while soft tissue expands and adapts to the new dimensions of bone [2]. The protraction of the maxilla with a distractor also gradually enlarges the pharyngeal space and allows for adaptation of the speech apparatus.

In adult cleft patients with major skeletal discrepancies, consideration is given to interdisciplinary orthodontic and surgical management. Orthognathic surgery corrects crossbites and improves facial features. With application of conventional surgical methods in severe maxillary hypoplasia, the degree of maxillary protraction is limited by rigid scarring of the lip and palate. If, following such a procedure, skeletal discrepancies remain, camouflage can be applied by performing a mandibular setback [8].

The assessment of our patient's occlusion, facial features and cephalometrics did not suggest a need for surgical correction. We were able to treat the case conservatively with satisfactory results.

The treatment of speech dysfunction in adult patients remains a challenge for speech-language therapists and phoniatricians, whether the patients are burdened with a cleft or not. Many years of abnormal articulation lead to habituation both in the patients and persons in their environment. This reduces the patients' motivation to consistently apply tedious exercises, which require patience and dedication. Our patient also presented a low level of motivation, failed to comply with her speech-language therapist's recommendations and eventually discontinued the process.

The reported patient is in the Polish age group, which at a young age did not have universal access to autologous bone graft reconstruction of the alveolar process. During fixed appliance treatment, the lack of bone in the cleft led to gingival and bony recessions around the teeth neighbouring with the fissure. The major bony defect was also a source of difficulties with prosthetic rehabilitation. The bonded bridge was therefore designed to include a fragment of pink porcelain to reconstruct the alveolar process.

There are many literature reports on orthoprosthetic treatment of adult cleft lip and palate patients. The most frequently applied solutions include removable prostheses, implants or luted porcelain bridges. In the reported case, we used a bonded bridge combined with an inlay to spare the abutment teeth from major grinding and avoid associated complications.

The treatment of adult cleft palate patients with presents a major challenge for the orthodontist. These patients have various combinations of malocclusions, dental as well as hard and soft tissue abnormalities. The cleft stigmatises the patient's face but also causes major mental and social burdens. Such patients require a holistic approach to therapy, which involves professional other than the orthodontist. Treatment plan frequently involve innovative methods, devices or procedures. Thanks to the current dynamic development of medical sciences and the advent of complex techniques, therapeutic teams can take on the difficulties which emerge and approximate treatment results achievable in non-cleft patients.

An appropriately planned and executed treatment, a good co-ordination of the interdisciplinary team, a compliant attitude of the patient and their family provides spectacular results both in terms of aesthetics and function. A satisfactory treatment result can restore self-esteem and improve social functioning for many patients who are burdened with a congenital malformation.

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