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TIMING OF CLEFT LIP AND PALATE REPAIR*

CZAS OPERACJI ROZSZCZEPÓW WARGI I PODNIEBIENIA

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Abstract

The opinions presented below concerning the choice of the timing of the cleft lip and palate repair have been presented on the basis of the literature and own experience. The comments and attempts to make an assessment reflect both direct contacts with representatives of numerous centres conducting interdisciplinary treatment of patients with clefts, as well as 35 years of the first author's experience gained in the course of treating 5,500 cases of primary clefts, besides own patients and numerous post-cleft deformities.

Key words: cleft lip and palate, timing of operations, late results of treatment

Streszczenie

W oparciu o piśmiennictwo i doświadczenia własne, przedstawiono poglądy na wybór czasu przeprowadzania operacji zespalandia rozszczepów wargi i podniebienia. W komentarzach i próbach oceny uwzględniono zarówno bezpośrednie kontakty z przedstawicielami wielu ośrodków prowadzących zespołowe leczenie pacjentów z rozszczepami, jak również 35-letnie doświadczenia własne pierwszego autora poczynione w trakcie leczenia ponad 5500 pierwotnych rozszczepów, nie licząc własnych i kierowanych z innych placówek zniekształceń porozszczepowych.

Słowa kluczowe: rozszczepy wargi i podniebienia, czas operacji rozszczepów, odległe wyniki leczenia

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Apart from cleft morphology and the surgeons' skills, the effects of treating cleft defects depend on numerous factors, such as the choice of operative methods, timing and sequence of surgical repair, as well as widely understood interdisciplinary treatment. One of the most disputable elements is the timing of treatment, around which a lot of controversy, emotions and differing opinions have accumulated, which are not only of medical nature.

Despite increasingly common prenatal diagnostics, the birth of a child with a cleft defect is a real shock to the parents and closest relatives, one which turns into striving for the quickest comprehensive treatment, though the repair of the most visible lip cleft is the priority. This is perfectly understandable, therefore the frustration of those involved is hardly surprising when frequently they

cannot obtain clear answers to what in their opinion is the simplest question, i.e. what and when should their child be operated on? However, contrary to popular belief, there are no simple answers.

Followers of the traditional approach begin treatment with the lip operation when the child is eutrophic (ca. 6 months old), whereas the cleft palate is repaired at the age of 2 to 3 years. In the past, relatively late treatment resulted from problems of anesthesiological nature and was due to the risk of causing midfacial underdevelopment. That approach was, and still is, quite reasonable, as the younger the organism, the more sensitive it is to traumas and the harmful, deforming effect of scarring, which may lead to severe growth disorders. Although the disorders do not result exclusively from the early

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operation, as other factors usually overlap, e.g. rough surgery, complications or using harmful surgical methods, yet this is still an important argument for delaying the operation. In practice that means attempting to ensure better maxillary development, unfortunately at the cost of delayed speech development. The speech quality has tremendous significance for normal functioning in society and therefore, in the opinion of speech therapists and paediatricians, it should be regarded among the priorities when developing the so-called treatment protocols. The increasing pressure of the phoniaticians is usually supported by parents who, understandably, strive for the quickest possible improvement of the physical appearance and repair of the cleft malformations.

Excluding the earliest attempts, characterised by a high mortality rate, the cleft lip repair performed during the first 48 hours after birth was initiated by Desai (1), who operated on 300 children. However, he did not present any evidence to justify his approach except for a general positive assessment, and the method did not find many followers. The fears and doubts were mainly related to the potential harmfulness of surgical trauma, and to avoid that, the concept of adhesive operations was suggested in the 1950s. The method, popularised by Millard (2), consisted in a simple lip adhesion procedure followed by proper correction after 3 months. Despite expectations, the proposed solution neither turned out to be simple, nor did it offer sufficient protection against iatrogenic complications, which, combined with a two-stage treatment, considerably reduced its popularity.

In 1966, Wilhelmsen and Musgrave (3) introduced the "rule of ten", i.e. 10 pounds of body weight, 10 weeks old and 10 mg/dl of haemoglobin. The rule translated into earlier lip cleft repair, which in specialised centres with adequate anaesthetic and paediatric staff, turned out to be justified and safe.

The one- or two-stage operation in wide bilateral clefts has remained a contentious issue. Although one-stage lip repair is regarded more rational and easier, yet in fear of causing excessive maxillary pressure, some surgeons prefer the two-stage operation, which makes it possible to obtain a more natural protrusion of the lower part of the lip and early columella elongation (3, 4) (fig. 1).

Reluctance to operate palate clefts early has been much stronger, as such operations are considered to be the main reason for iatrogenic deformities and maxillary growth disorders. This applies in particular to operations consisting in mucoperiosteal flap plasty according to methods of Langenbeck, Veau, Kilner and Wardill (6-9), leading to reduced blood supply, bone exposure and healing of extensive wounds by granulation. The above was confirmed by the publication by Ross et al. (10), who, based on the data from American and Canadian centres, found indications for orthognathic operations in 25% patients with complete unilateral cleft lip and palate. The above was also confirmed through experimental research by Kim et al. (11).

The introduction of the so-called sparing surgical methods gave an impulse for earlier cleft palate operations, which created chances of less harmful surgery and justified, in a sense, changes induced in relation to

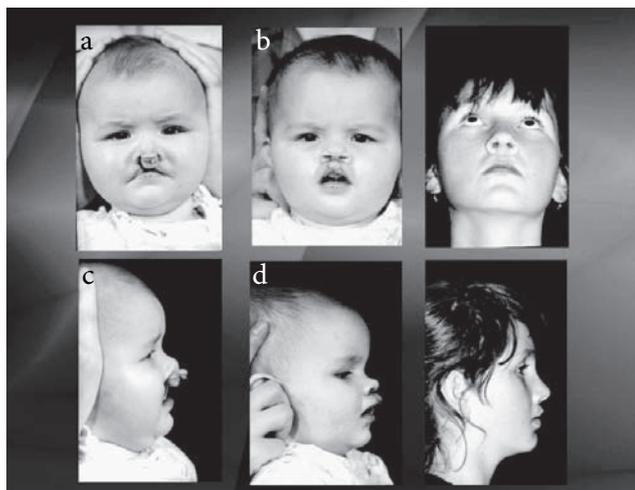


Fig. 1. A, B. Bilateral lip and palate cleft C, D – late result of two-stage lip suture combined with columella elongation by means of the author's method (K. K.), no correction surgeries.

Ryc. 1. A, B. Obustronny rozszczep wargi i podniebienia. C, D – Odległy wynik dwuetapowego zeszytce wargi w połączeniu z wydłużeniem słupka sp. autora (K.K.), bez jakichkolwiek operacji korekcyjnych.

earlier operation. The Schweckendiek's method (12) was among the most popular ones. It consisted in soft palate closure at the age of 6 months followed by hard palate closure when the patient was 14 years old, which was to facilitate quick development of speech and prevent jaw underdevelopment.

Despite logical assumptions, it turned out that the soft palate closure using Schweckendiek's method was only slightly less traumatic than the operation of the entire palate, and the speech of patients forced to wear a hard palate obturator was found highly unsatisfactory by most of the authors (13-16). There were also positive assessments, e.g. by Schweckendiek's son (17) or Perko (18), who performed veloplasty using the V-Y technique at the age of 18 months and hard palate closure at the age of 5-6 years.

Farina in 1958 and Davies in 1966 initiated the one-stage lip and palate operations in children over 1 year old and at the age of 5-6 months (19, 20). Unfortunately, the data are too scarce to evaluate that method, because apart from the justification for the adopted approach (Farina) (19) and the description of the surgical technique (Davies) (20), the remaining data describe only a few months of observation. Three short, subsequent publications by Kaplan et al. focus on 8-10 years of observation of patients who underwent one-stage surgery at the age of 3-4 months, yet the articles provide only general information (21-23). Honigmann, who treated 6 month-old children, presented a one-stage surgical technique, however the observation period (1-2 years) was too short and made it impossible to evaluate the method (24).

Among rare publications on early, one-stage cleft lip and palate operations, the work by Dudkiewicz et al. (25) offers observations supported by orthodontic

measurements. As the author informs, the method discussed has been successfully used since 1980, i.e. the period is long enough to present treatment outcomes based on large material and long – term observations. However, the reports published every now and then concern exclusively the treatment results in children at the maximum age of 9-10 years (26-29). As a result, the importance of such publications is diminished and explanations attributing the lack of the late results to some modifications of surgical methods do not sound convincing. This makes it impossible to evaluate these methods, as the tendency to slow down midfacial growth with time is observed (30) – which means that the parameters analysed in a ten-year-old will deteriorate after the patient has ceased to grow, i.e. after the patient is over 18-20 years old.

Judging from personal contacts and literature, the work of Malek has been best documented so far. Malek, together with Psaume, an orthodontist, proposed a reversed sequence of treatment, comprising surgery of the soft palate at the age of 3 months, followed by the operation of the lip and hard palate cleft at the age of 6 months (31). Due to the widely known insufficiency of tissue at the border of the hard and soft palate, Malek sutured in a small vomer flap, which, contrary to Pichler (32) and the first author of this work (33), was anteriorly pedicled and reversed by ca. 1250. The hard palate closure was single-layer, which, unfortunately, often contributed to the formation of fistulae.

The continuator of the method, de Mey et al., used it in the years 1981-1988, however later the single-stage treatment was taken up (34). In the next publication (35), the prospective study of the two groups of patients was presented. The authors performed two-stage operations on 34 patients at the age of 3 and 6 months. The control group consisted of 38 patients who underwent a single-stage lip and palate operation at the age of 3 months. Their craniofacial morphology was assessed twice – when the patients were 10 and 15 years old. The first evaluation showed relatively small differences between the groups, which concerned mainly the relation of the maxillary plane to the skull base. However, in comparison to healthy children, statistically significant smaller maxillary protrusion (<SNA), reduced posterior maxillary height in both groups (R-PMP) and reduced anterior maxillary height in children after the two-stage surgery was revealed. Unfortunately, the groups examined at the age of 15 were smaller (n=18 and 21) and were not compared to the patients who did not suffer from clefts. The measurement results were similar in both groups, yet, as the authors quoted state, it will be necessary to analyse the late treatment outcomes after the completion of skeletal growth, based on the analysis of the craniofacial morphology, tooth models, speech and hearing.

Taking the above into consideration, it is very probable that the views on the optimal timing for lip and palate cleft repair performed under the age of 1 year, especially as regards the one-stage procedure, will not become consistent for a long time yet. There is still an insufficient number of long-term reliable studies, especially of prospective nature. As nature abhors a vacuum and the temptation

to boast of success happens to be contagious, we witness a virtual flood of information based on heterogeneous and scarce material, observations conducted merely for a few years and fragmentary measurements, tailored to justify the adopted assumptions and confirm the rightness of a given approach. Among the most common, there is the practice of comparing treatment results with not always optimal methods or centres, whereas referring to the norm is much less frequent. And if such a reference occurs, it is usually to the disadvantage of the adopted treatment methods (36). Certainly, it may be attributed to the weakening of the primary growth potential in patients with clefts (37, 38), but it does not change the fact that a vast majority of growth, shape and function disorders are of iatrogenic origin and are considerably related with timing the operation. Skeletal disorders and maxillary hypoplasia should always be regarded as an evident failure and proof of inadequacy of surgical treatment. Therefore, we find it unacceptable to underestimate the midfacial underdevelopment by adopting a view that the maxilla may be later moved forward by means of the osteotomy or distraction.

Due to the lack of convincing evidence gathered in many centres in favour of the effectiveness of the early one-stage operation, many surgeons with satisfactory treatment results take the middle ground, in line with the principle *primum non nocere*. Most often it means that the cleft lip is operated on between the 3rd and 6th month of age and the cleft palate between the 1st and 2nd year of age, which provides conditions for normal feeding and speech, as well as reduces the harmful effect of the surgical trauma on the midfacial development. In some sense, these are attempts to find a happy medium, as they are often combined with methods considered “sparing”, such as adhesion procedures, the use of mucous flaps instead of mucoperiosteal flaps, the use of vomer flaps etc. (15, 18, 32, 33, 39) (fig. 2).

As own research has shown, the late treatment results of patients in whom the palatoplasty with extended vomer flap was performed at the age of 3-6 months did not considerably differ from the outcomes in patients operated at the age of 2-3 years (40). What is interesting, the above is true not only concerning orthodontic measurements (which favourably compare with the three best European centres examined by EUROCLEFT (41)), but also speech quality. Speech, which depends not only on early surgery but also, and fundamentally on its quality, i.e. the correct suturing of muscles combined with palate elongation (39, 42) and lack of fistulae, which are frequent in Malek's method (16%).

We are aware that the declared lack of significant differences in speech quality between early and delayed surgery sounds almost like heresy (14, 15), and we find that verification based on more accurate research, more ample material and data from other centres applying similar methods would be recommendable. Nevertheless, based on the 35 years of experience of the first author (K.K.) and the material of the Hospital and Clinic of Plastic Surgery in Polanica Zdrój by 2008 comprising over 5,500 primary clefts, it seems that excessive haste, and in particular radical one-stage surgery, is not recommendable

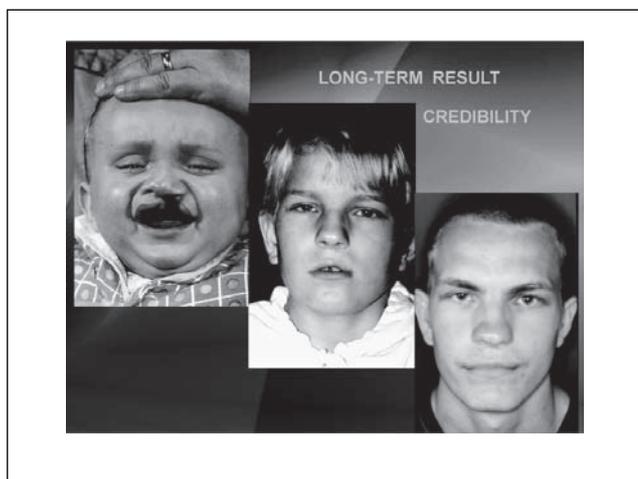


Fig. 2. A Unilateral lip and palate cleft before treatment. B, C – Early and late treatment outcome after the lip surgery at the age of 6 months and palate suture with the use of extended vomer flap at the age of 2 years (K. K.).

Ryc. 2. A Jednostronny rozszczep wargi i podniebienia przed leczeniem. B, C – Wczesny i odległy wynik leczenia po operacji wargi w wieku 6 miesięcy oraz zeszcyciu podniebienia z użyciem wydłużonego płata z lemiesza w wieku 2 lat (K.K.).

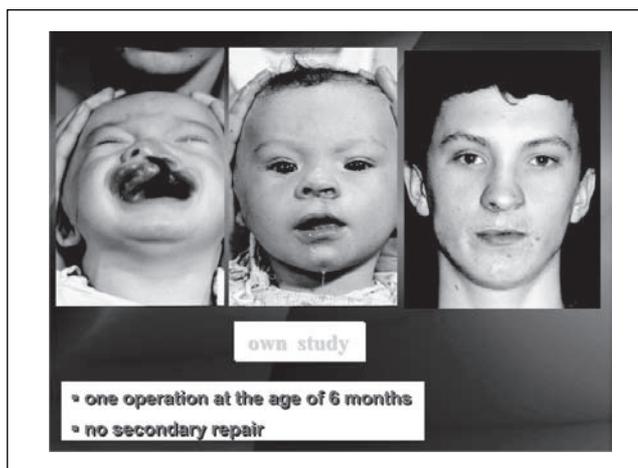


Fig. 3. A Unilateral lip and palate cleft. B – The condition after suture of the lip and palate cleft (with the use of a vomer flap) at the age of 6 months. C – Late treatment result without any correction surgeries.

Ryc. 3. A Jednostronny rozszczep wargi i podniebienia. B – Stan po zeszcyciu rozszczepu wargi i podniebienia (z użyciem płata z lemiesza) w wieku 6 miesięcy. C – Odległy wynik leczenia bez jakichkolwiek operacji korekcyjnych.

due to the risk of growth disorders occurring especially in the event of complications and rough surgery.

Although our own pilot trials conducted in the 1980s with respect to one-stage lip and palate cleft operation using vomer flaps were satisfactory (fig. 3), they were abandoned, as in two cases the risk of jaw underdevelopment was revealed. That decision, perhaps a premature one, was dictated by prudence relating to treatment and to consultation provided to thousands of patients with

cleft deformities and the belief that what matters in the long run are the final treatment outcomes, often having a decisive effect on the patients' whole life.

The frequency and gravity of correction surgeries are also of importance, because the need for re-operations occurring after one-stage procedures may render the benefits of early and radical operations not so self-evident. In addition, due to the greater area to be operated on and the fact that the operative time is 50% longer (35), this type of treatment should be confined to specialised centres and closely monitored.

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