

The contemporary approaches to alveolar bone defect management in children with cleft lip and palate. A review

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Abstract

The cleft lip and palate belong to the triad of the most common defects in humans. Secondary maxilla bone plasty, as a stage of complex rehabilitation, was first described in the 1970s. Its principle was to restore the dentition and connect the jaw fragments, and move and stabilise the existing teeth in a new position by bone grafting of the alveolar process and implantation of bone tissue (or bone substitute material) into the defect. The approaches to congenital defects of the alveolar process plasty of the upper jaw with congenital facial defects differ significantly in different countries and clinics in terms of timing, type, and processing of bone substitute material, as well as the level of filling of the alveolar defect. Comprehensive rehabilitation of such patients includes alveolar process osteoplasty, which is a promising direction in the development of maxillofacial surgery and surgical dentistry, and it contains some poorly understood and unexplored aspects.

Introduction

According to data from various international organisations and national healthcare systems, congenital clefts of the lip, alveolar process, and palate (CLP) belong to the triad of the most common human congenital defects. In the maxillofacial area, such defects are generally the most frequent. As professional publications indicate, the birth rate of children with congenital maxillofacial defects is increasing every year worldwide. According to the WHO, the daily birth rate of such children worldwide reaches 220, and annually over 80,000 cases. In Ukraine, children with congenital defects are born in the country with a frequency of 1 : 600–700 births; as of 2015 this figure was 1 : 700, and in 2021 it was 1 : 600, which indicates a growing trend. In some works we can see cited data that the approximate birth rate of such children worldwide is 1 case per 700 newborns, and there is also a tendency towards growth. In each of the countries of the world, attempts are still being made to coordinate and regulate the procedure and scope of medical rehabilitation for such patients and ensure their full recovery. In some countries, the provision of medical care to this contingent of patients has also been regulated at the state level. However, the medical and social aspects of the rehabilitation of such patients in many countries of the world are not fully resolved [1–4].

The birth of a child with a cleft lip and palate is accompanied by a number of medical and social prob-

lems, which, according to many authors, determine the need for continuous improvement of rehabilitation methods for these patients. The medical and rehabilitation measures that such a patient should receive is complex, multi-stage, and long-term. In Ukraine, the current rehabilitation protocols for children with CLP include the content and elements of guideline 00158, which was based on the principles of evidence-based medicine [1, 4–6].

As practice shows, approximately 60% of children with congenital clefts of the upper lip and palate have a residual alveolar process defect [7], and for complete rehabilitation of such a patient, that defect must be closed. Therefore, surgical intervention to reconstruct the clefts of the alveolar process of the maxilla in children with CLP has found many like-minded supporters. The interest of domestic and foreign authors in this direction is also associated with the fact that by eliminating the alveolar process defect, not only is the anatomical integrity of the upper jaw restored, but several other important tasks are also solved. In particular, the stability of the non-fused fragments of the upper jaw is recorded, and support for the bone and cartilage structures of the nose is ensured, which allows exclusion of the subsidence of soft tissues of the lip. Achieving such even intermediate results creates favourable conditions for the development of not only the upper jaw but also the entire middle facial zone, contributing to the stabilisation of orthodontic treatment results, and creating conditions for orthognathic surgery [8, 9].

One of the problems for children with congenital CLP (and maxillary alveolar process) is dysfunction of the upper lip muscles, manifesting not only in the form of changes in the lips' architecture, but also in the form of changes in the muscular tone of the *m. orbicularis oris*, both at rest and during activity (including parafunctions). Such a situation may have an additional adverse effect on the development of malocclusions and secondary deformities, as well as change the effectiveness of orthodontic treatment of patients [10–14].

However, despite the long history of solving the problem of treating children with CLP, to this day, specialists do not have a single opinion about the timing of surgical intervention and methods of surgical treatment, as well as the need to use osteoplastic material and its selection. By solving the listed problems, which have been repeatedly raised in foreign and domestic scientific literature, it would become possible to achieve an optimal anatomical, cosmetic, and functional result in the full rehabilitation of patients. According to orthodontists and orthognathic surgery specialists, the very presence of an alveolar process defect at the level of its basal part plays a significant role in the mechanisms of upper jaw deformation in children with CLP, leading to a violation of the shape and size of the basal section of the upper jaw, which further negatively affects the stability and sustainability of orthodontic treatment results. To date, many approaches and methods for each stage of eliminating the alveolar process defect of the upper jaw in children with congenital facial defects are known. Quite often, they are associated with the choice of technique and ideal material for bone tissue autotransplantation (or implantation of other materials), as well as patient age. In addition, during treatment, several questions arise; firstly, about the expediency of using such auxiliary interventions as orthodontic expansion of the upper jaw, and secondly, when it should be carried out – before or after transplantation [14–17].

The pronounced variability of alveolar process defect anatomy in children with congenital defects additionally creates a problem of classification and visualisation. According to known classifications, as a rule, the anatomical basis of the jaw is taken into account, so CLP is divided into unilateral or bilateral, obvious or hidden, complete or partial. At the same time, the alveolar process defect of the upper jaw can be either complete one- or bilateral clefts, or a partial cleft lip. It is also worth noting that in children with CLP after surgical interventions (cheiloplasty, veloplasty, and palatoplasty), secondary cicatricial deformations are present: upper lip, nose, upper lip, and hard and soft palate. Deformations are distinguished by type and severity. Also, the type of cleft affects the severity of deformation, namely the presence of an alveolar process defect and its size. The bone basis of the alveolar process of the upper jaw is the foundation for all soft tissue structures surrounding it [6, 7, 16, 18].

Purpose of the study

The study aims to explore the historical development of modern approaches to the management of congenital maxillary alveolar process defects in children with cleft lip and palate.

Modern diagnostic measures for planning and evaluating the effectiveness of alveolar process defect elimination in the maxilla in children with cleft lip and palate

For diagnosis, planning, and assessment of surgical outcomes, the most popular diagnostic methods include orthopantomogram, targeted radiograph, 3DCT, and cone-beam computed tomography (CBCT). In 1996, Rosenthal compared the effectiveness of targeted radiography and CT in assessing the preservation of the bone graft after alveolar ridge defect plastic surgery of the maxilla. The study lasted 6 months post-operation in 14 patients with cleft lip and palate. The research confirmed that using targeted dental radiographs to evaluate the size of the preserved graft is justified. With targeted dental radiography, we can determine the height of the bone bridge of the graft and the interdental septum, while the patient is exposed to significantly less radiation compared to CT and CBCT. In 2005, Trindade *et al.* noted that orthopantomogram and targeted radiography allow determination of the following: the planar size of the defect before surgery, the amount and distribution of bone at the transplantation sites, the height of the interdental septum formed during tooth eruption, and the graft's reaction to orthodontic treatment. Feichtinger *et al.* pointed out that orthopantomogram and dental radiography are unsuitable for planning alveolar ridge bone grafting and evaluating the bone graft for significant reasons: image magnification and distortion, overlapping of adjacent anatomical structures, limited number of identifiable landmarks, and positioning issues [19–21].

However, two-dimensional imaging of a three-dimensional structure has many inaccuracies, and using such diagnostic methods makes it impossible to assess the volume of the alveolar ridge defect of the maxilla. CBCT with a significantly lower radiation dose than conventional CT provides more accurate images of the maxillofacial region. Currently, there are several programs (HOROS and others) that enable analysis of the patient's CT and determination of the alveolar ridge defect volume. All this collectively allows for proper planning of the surgical procedure. Therefore, the assessment of alveolar ridge bone grafting results is based on clinical, photogrammetric, and radiological data. Clinically, the outcome of alveolar ridge bone grafting is evaluated based on parameters such as the presence of a secondary defect (oro-nasal communication) at the main defect site or the depth of periodontal pockets around erupting teeth on the cleft side (canine or lateral inci-

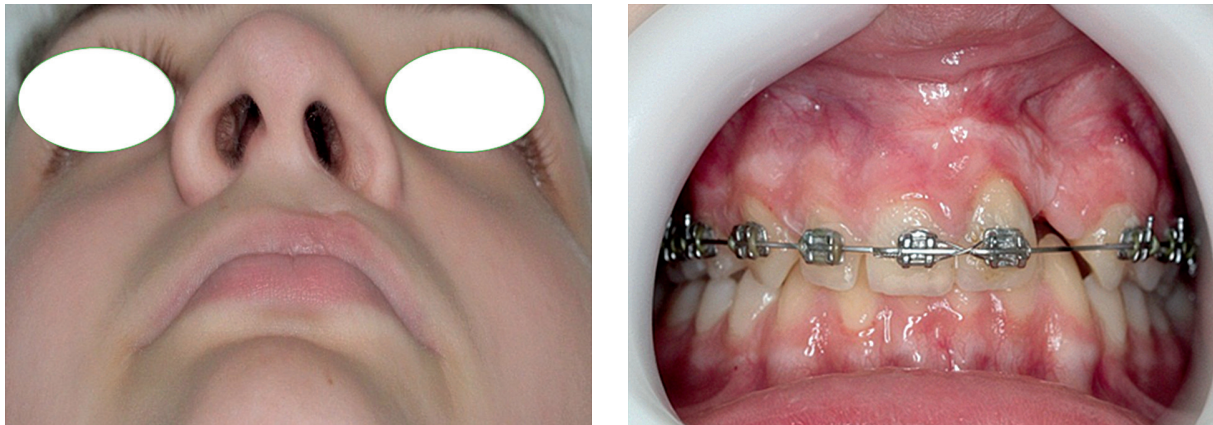


Figure 1. Intraoral photo and deformation of nasolabial complex in a patient with a small volume of alveolar process congenital defect ($V = 1.0 \pm 0.5 \text{ cm}^3$) (from own clinical archive)

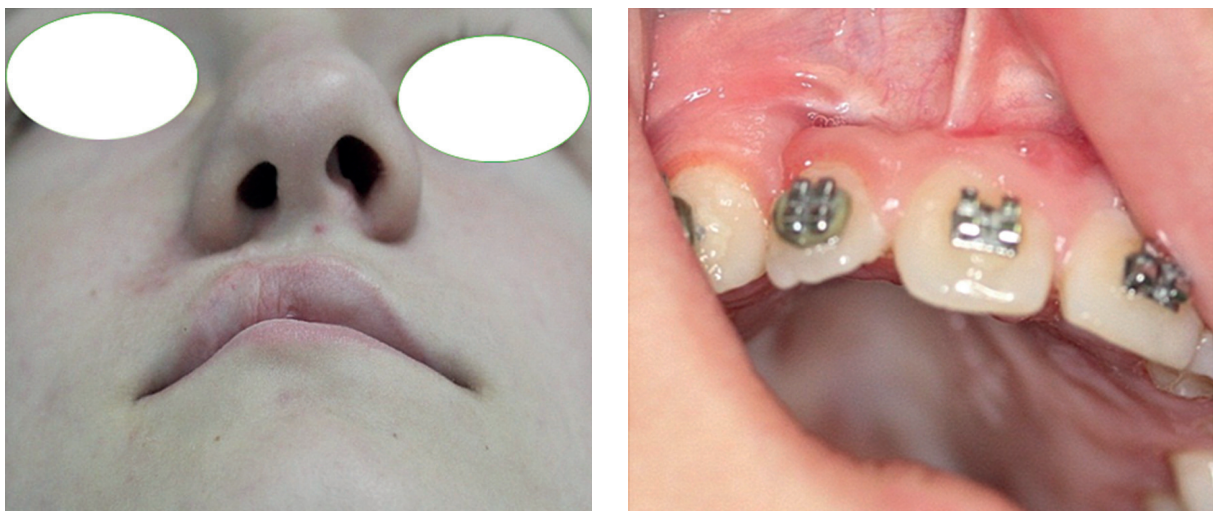


Figure 2. Clinical presentation of a patient with a medium volume of alveolar process congenital defect ($V = 1.0$ to $1.5 \pm 0.5 \text{ cm}^3$) (from own clinical archive)

sor). One of the most important goals of reconstructing the alveolar ridge defect is to ensure bony support for the nasal wing on the cleft side (Figures 1–3). Therefore, photogrammetry is used to assess the quality of alveolar ridge bone grafting. It evaluates changes in the position of the nasal wing before and after surgery and compares the healthy side with the cleft side [20, 22–24].

Radiologically, the results of alveolar ridge bone grafting are assessed by measuring the height of the interdental septa at the defect edges and using various classifications. Abyholm *et al.* were the first to propose radiological determination of the intra-alveolar septum height. One of the most well-known classifications for assessing the quality of secondary alveolar ridge grafting is the Bergland classification, also known as the Oslo classification system, from 1986. This classification divides bone height in the defect area into four types: type I (approximately normal height), type

II ($\frac{3}{4}$ of normal height), type III (less than $\frac{3}{4}$ of normal height), and type IV (absence of bone graft). Kindelan *et al.* proposed a classification to evaluate the quality of secondary alveolar ridge grafting (defining four classes): I (more than 75% bone fill), II (50–75% bone fill), III (less than 50% bone fill), and IV (no formed bone bridges). In 2003, Hynes and Earley suggested a modification of the Oslo measurement system. They performed an average radiological observation of alveolar ridge grafting over 4.5 years. They performed $3 \times 4 \text{ cm}$ periapical dental radiographs, measuring the occlusal level, basal level, and the total height of the newly formed bone in the alveolar ridge defect, evaluated using the Oslo system. The height of the bone graft was compared with the expected normal interdental alveolar bone tissue on corresponding films [9, 15, 24, 25].

Long *et al.* studied the contours of bone tissue in 46 clefts, with an average follow-up of 3 years; they



Figure 3. Intraoral photo and deformation level of nasolabial complex in a patient with a huge volume of alveolar process congenital defect ($V > 1.6 \pm 0.3 \text{ cm}^3$) (from own clinical archive)

identified a series of measurement ratios obtained directly from radiographs, and they could detect patterns in the formation of bone bridges. Their measurements include the amount of bone graft, the length of the proximal and distal segments of the piriform aperture, the cutting edge from the healthy side of the root, the position of the alveolar crest, and the size of the largest coronary attachment of the bone from both sides. Witherow *et al.* analysed radiographs of 87 cleft areas using an 8-point scale to describe the position of the bone graft relative to the roots in the cleft area (their scale can be used in variable dental occlusion if the root length is divided into four parts, assuming radiographs are directed through the cleft line). Additionally, depending on the position of the bone bridges across the cleft, radiographs were classified into one of six groups (A to F). Nightingale *et al.* compared three methods of radiographic analysis proposed by Bergland *et al.*, Kindelan *et al.*, and Witherow *et al.* They found that none of the three radiographic scales demonstrated superior reproducibility over the others, that each scale appeared more reproducible in variable dental occlusion, and that neither occlusal nor periapical radiographs proved more useful during the assessment of alveolar ridge grafting success [26–28].

Development of surgical techniques for restoring defects of the maxillary alveolar process in CLP

Secondary bone grafting of the maxillary alveolar bone, as a stage of comprehensive rehabilitation for children with congenital maxillofacial defects, was first described in the 1970s. The principle was to restore the dental arch and connect jaw fragments,

move and stabilise existing teeth in a new position by bone grafting of the alveolar process, and transplanting bone tissue (or bone-substituting material) into the defect, with the expectation of their integration and replacement with native bone tissue. Therefore, it is important for tooth movement and stabilisation that the alveolar process defect be closed. In modern conditions, two surgical techniques are used to solve this problem: bone grafting of the alveolar process and periosteal plasty. The main goal of bone grafting or periosteal plasty in patients with CLP is to close the alveolar process defect and create its continuity, so that permanent teeth can erupt in a more physiological position and be successfully moved using orthodontic appliances and devices. Reconstruction of the maxillary alveolar process also serves to connect the maxillary segments and prevent growth disorders [1, 2, 8, 9, 29].

Bone transplantation for residual (secondary) defects of the maxillary alveolar process was first performed in the late 1950s. Such interventions belong to the technically most complex surgical techniques in the maxillofacial area. For a long time, the main difference between treatment protocols in different clinics was the timing of bone grafting. For example, primary bone grafting, introduced by Nordin and Johansson in 1955, served to reconstruct the alveolar process defect in childhood or early childhood. Due to frequent reports of midface growth disorders resulting from these operations and less successful bone formation in the defect area, this method was gradually replaced by primary periosteal plasty. In the 1960s, Tord Skoog performed paediatric periosteal plastic surgery simultaneously with primary lip plastic surgery. The “non-bone plastic” technique in-

volved using local periosteal flaps that were moved to the defect area to close it during cheiloplasty, thereby eliminating the alveolar process defect and creating continuity between the maxillary segments. This technique did not involve the use of bone tissue or bone-substituting materials. However, many clinics abandoned this method due to the accumulation of negative experience and data on insufficient bone tissue formation to support teeth adjacent to the non-fusion. Moreover, it was suggested that using the periosteum necessary for periosteal plastic could further harm jaw growth due to trauma, especially if such manipulation is performed in childhood or early childhood. As a solution to the shortcomings of primary bone plastic and periosteal plastic, the use of autotransplantation of spongy bone tissue to the non-fusion of the alveolar process of the upper jaw during mixed dentition, before the eruption of the permanent canine, was introduced, which was later called secondary bone plastic [8, 30–32].

Another approach to eliminating defects of the alveolar process of the upper jaw was bone plastic surgery performed during the eruption of permanent teeth, after the completion of orthodontic treatment, called tertiary or late bone plastic. Bone tissue transplantation in tertiary plastic became a tool for ensuring prosthetics and periodontal rehabilitation of the patient, as well as for closing persistent oronasal fistulas. However, tertiary (or late) bone plastic cannot replace the loss of bone tissue located near the teeth adjacent to the cleft. Therefore, in modern conditions, secondary bone plastic of the alveolar cleft, first described by Boyne and Sands in 1972, has become a common operation. Such plastic is performed at the stage of mixed dentition in combination with orthodontic treatment. The optimal age period for performing this type of osteoplasty is the period from 8 to 11 years, when the root of the permanent canine is already formed to 1/3 of its length [2, 5, 29, 33–35].

During such a period, alveolar ridge bone plastic surgery of the upper jaw ensures the eruption of the canine through the segment of the former cleft of the process, provides bony support for teeth adjacent to the non-union, maintains arch width, and stabilises the upper jaw arch. It also serves as a support for removing any residual oronasal fistula and can improve facial symmetry by providing some support to the nasal base and nasolabial contour. In 1990, Millard and Latham introduced a variant of periosteoplasty combined with active preoperative orthopaedic treatment. Gingivoperiosteoplasty is performed on alveolar segments that are in direct contact as a result of preoperative orthodontic treatment. Millard and Latham emphasised that their method differs technically from that practiced by Skoog in that minimally incised and less invasive local flaps are used to cover the reduced size of the alveolar ridge defect. However, long-term follow-up of operated patients performed

by Berkowitz *et al.* revealed delayed maxillary growth during adolescence [5, 18, 32, 36].

In 2021, Ma *et al.* evaluated the success of gingivoperiosteoplasty using the Millard-Latham technique compared to bone plastic surgery in patients with unilateral and bilateral clefts using a radiographic assessment scale. Other researchers who developed this approach found that the quality of bone tissue in the reconstructed cleft zone was lower in patients who underwent gingivoperiosteoplasty, and that most operated patients required repeat bone plastic surgery [37]. In 1982, Friede and Johanson performed a study that showed that patients who underwent alveolar ridge bone plastic surgery in early childhood developed severe retrognathia and significant vertical growth of the upper jaw. Such surgical intervention was sharply criticised by many surgeons [38].

In 1987, Ross published the results of a study on the effects of different treatment methods on the growth of the upper jaw. According to his data, early periosteoplasty and alveolar bone plastic surgery negatively affect the growth of the upper jaw. Early secondary bone plastic surgery, performed at ages 4–9 years, has the same effect on the development of the upper jaw as in early childhood, and the operation performed at age 9 years does not have a negative impact on the transverse growth of the upper jaw. Such data allowed for the restriction of the age period for performing this type of osteoplasty [39].

In the further evolution of preoperative orthopaedic techniques, Grayson introduced nasoalveolar moulding combined with gingivoperiosteoplasty in 1999. Although Millard's works contain arguments for early union of the maxillary dental arch combined with lip repair and primary gingivoperiosteoplasty to reduce the future need for alveolar bone plastic surgery [40]. Today, the majority of practicing surgeons believe that at age 9 years, the growth of the anterior part of the upper jaw in length and width ceases. The further development of the mid-face occurs through downward and forward growth of the maxilla. After performing bone plastic surgery, there is an opportunity for teeth to erupt through the graft and for further growth of the upper jaw.

Types of bone grafts and their effectiveness for alveolar process recovery in children with CLP

Autogenous bone grafts can be obtained from extraoral or intraoral sites, with spongy autograft – from iliac or tibial bones, and cortical-cancellous blocks – from skull bones and mandibular symphysis. During secondary alveolar ridge plastic surgery, one of the most common grafting materials is cancellous bone taken from the anterior part of the iliac crest. The advantages of this method are that the material can be harvested in large quantities. During the op-

eration, the team should include two surgeons and two operating tables: one creates the surgical bed, and the other harvests the autograft [1, 5, 7].

According to Pinholt and Kerwin, cancellous iliac bone has significantly more advantages over cortical-cancellous blocks. The authors state that revascularisation of the autograft generally occurs within 3 weeks and is associated with a high potential for osteoprogenitor (stem) cells and pluripotent cells. These cells participate in early-stage osteogenesis following bone grafting. Literature indicates that the main drawback of iliac crest bone grafts is complications at the donor site. Postoperative pain is felt in the iliac crest area, and children are unable to move for several days. In such cases, most patients report pain of such intensity that the thigh area is more bothersome than the oral cavity. The duration of hospitalisation and postoperative rehabilitation is quite long. However, the use of this type of autograft remains the gold standard for many authors. The risk of resorption of such autografts can reach up to 40.0% within a year, which has prompted surgeons to seek other autogenous bone harvesting sites [41–44].

The tibia is considered an alternative harvesting site because it also contains cancellous bone tissue. In 1908, Lexer performed the first surgical intervention to eliminate the defect of the alveolar process of the maxilla using a cortical-cancellous graft from the tibia. These grafts were used in orthopaedics and later in reconstructive craniofacial surgery during orthognathic procedures and cleft repairs. Initially, this type of graft was used only in adults before it was adopted for children [45, 46].

Taking cancellous bone from the epiphysis of the tibia before its complete growth poses a significant risk of damaging the growth centre. In 1999, the technique for harvesting autografts in children was described. However, in this case, the amount of cancellous bone is less than from the iliac crest. For an adult, the volume is limited to 25 ml. In the case of a child, the volume of harvesting is even smaller, so sometimes it can be performed from both legs if a significant volume of bone is needed. After such intervention, patients are asked not to play sports for 3 months. Therefore, due to the possibility of injury to the growth zone, this type of transplant is not the first choice [47–50].

In 2003, Rosenthal and Buchman performed an experimental study in which they compared bone grafts of cortical and cancellous iliac bone (endochondral origin) with grafts of the mandible (membranous origin) in rabbits. Four defects of the required size were made in the rabbit's skull: the first was filled with cortical iliac bone, the second with cancellous iliac tissue, the third with cortical bone of the mandible, and the fourth left empty as a control (healing under a clot). The animals were removed from the experiment at 3, 8, and 16 weeks for histological exami-

nation of the results. Interestingly, the authors noted that the best result was obtained using cancellous iliac tissue [19].

It is worth emphasising that the bones of the skull have a single embryological origin, and the membranous process of ossification is similar to that of the alveolar bone. The authors recommend using bone autografts from the bones of the skull in reconstructive maxillofacial surgery to eliminate defects of the alveolar process in certain cases. They are usually used to fill the upper part of the alveolar process defect and the *apertura piriformis*. This conclusion was reached by several authors – such as Sadove, Kortebein, and La Rossa – in their studies, showing better results when using cancellous iliac bone tissue compared to transplants from the skull [43, 51, 52].

Even if it is possible to take the required amount of bone material from the bones of the skull, the healing of donor areas may take longer, and be more difficult and less productive than on the iliac crest. The complication rate ranges from 0.25% according to Tessier (published in 2005) to 5.5% according to Jackson (published in 1986). According to the literature, complications when harvesting this type of autograft can be as follows: osteomyelitis of the skull bones, exposure of the dura mater, leakage of cerebrospinal fluid, extra- or subdural haematoma, and neurological complications. A scar remains on the head, which is quite difficult to hide with hair. Such areas of surgical intervention do not allow two surgeons to work simultaneously [23, 53–58].

It is also worth noting that the mandible has a single embryological origin with the maxilla. The first to describe the use of the mandibular symphysis to eliminate defects of the alveolar process of the maxilla were Bosker and van Dijk in 1980. They published their preliminary results on 25 cases in 1989. Also in 1994, Koole confirmed that the mandibular symphysis is an attractive donor site due to the ease of access, and that it gives good results of alveolar cleft transplantation. The advantages of this type of transplant are a single surgical site, an invisible scar, reduced postoperative pain, a short hospital stay [59–61].

Several authors (Freihofer, Koole, Park, Lee) described good results of using mandibular transplants compared to iliac bone transplants, but most of their assessments were performed in 2 dimensions and cannot be final. In addition, most authors report that the collected volume of bone on the mandibular symphysis is often insufficient for large or bilateral clefts. There is also a risk of injury to the roots of the teeth, which are located in the area of harvesting the autograft, especially when the transplantation is done at an early age [60, 62–66].

Rib bone tissue is the second most commonly used autogenous graft for alveolar ridge defect reconstruction. The rib is considered a practical donor site for autogenous bone for primary alveolar transplantation,

but incomplete graft integration into the alveolar bone, lack of bone reservoir, and the inability of teeth to erupt are the main disadvantages reported in the scientific literature on rib transplantation. In addition, the possibility of pneumothorax and persistent postoperative pain is not reported often enough. Ribs have no significant advantages over bone tissue taken from the iliac crest for alveolar defect reconstruction, which would justify the preference for using rib grafts at the stage of mixed dentition [67, 68].

It is also known that allografts (allogenic bone) have been proposed for alveolar ridge reconstruction because they do not have the disadvantages of autogenous bone grafts. This has advantages in limiting the time of surgery, providing a large amount of bone. Allografts have both osteoinductive and osteoconductive properties. Bone morphogenetic proteins (BMPs) provide osteoinduction and are released in response to osteoclastic activity to stimulate graft bioincorporation. The calcified collagen structure provides osteoconductive properties, forming a scaffold that facilitates bone deposition. To date, it is also not recommended to use bone substitutes (synthetic osteoplastic materials), such as hydroxyapatite, for alveolar ridge reconstruction in children before the completion of bone system formation, due to the fact that the bone tissue that forms in the defect area is very dense and does not allow the teeth around the defect to erupt. Such materials should only be offered to adult patients. Research data and clinical experience on the use of xenografts and bioceramics for alveolar ridge defect reconstruction are contradictory, and there is insufficient evidence in the scientific literature on this. Therefore, their use during alveolar cleft reconstruction is not recommended [2, 6, 62, 69–72].

Conclusions

Today, congenital cleft lip and palate belong to the triad of the most common defects in humans. Despite the long history of solving the problem of treating children with congenital defects of the alveolar process of the upper jaw, to this day, specialists do not have a single opinion on the timing of surgical intervention and methods of surgical treatment, as well as on the need to use osteoplastic material and its choice. If the above problems are solved, it would be possible to achieve optimal anatomical, cosmetic, and functional results in the full rehabilitation of patients.

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Ethical approval

The study was performed according to the guidelines of the Declaration of Helsinki and other legislation, and it was approved by the Bioethics Committee

of the State University “Uzhhorod National University” (Ukraine) (number 1/3, date of approval 2 November 2022).

Conflict of interest

The authors declare no conflict of interest.

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