

Research Paper
Cleft lip and palate

Healthcare use and direct medical costs in a cleft lip and palate population: an analysis of observed and protocolized care and costs

I. Apon^a, N. van Leeuwen^b,
S. Polinder^c, S.L. Versnel^d,
E.B. Wolvius^a, M.J. Koudstaal^a

^aDepartment of Oral and Maxillofacial Surgery, Erasmus University Medical Center, Rotterdam, the Netherlands; ^bMedical Decision-Making Section, Department of Public Health, Erasmus University Medical Center, Rotterdam, the Netherlands; ^cHealth Technology Assessment and Implementation Section, Department of Public Health, Erasmus University Medical Center, Rotterdam, the Netherlands; ^dDepartment of Plastic and Reconstructive Surgery, Erasmus University Medical Center, Rotterdam, the Netherlands

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Abstract. This study was performed to describe observed healthcare utilization and medical costs for patients with a cleft, compare these costs to the expected costs based on the treatment protocol, and explore the additional costs of implementing the International Consortium for Health Outcomes Measurement (ICHOM) Standard Set for Cleft Lip and Palate (CL/P). Forty patients with unilateral CL/P between 0 and 24 years of age, treated between 2012 and 2019 at Erasmus University Medical Center, were included. Healthcare services (consultations, diagnostic and surgical procedures) were counted and costs were calculated. Expected costs based on the treatment protocol were calculated by multiplying healthcare products by the product prices. Correspondingly, the additional expected costs after implementing the ICHOM Standard Set (protocol + ICHOM) were calculated. Observed costs were compared with protocol costs, and the additional expected protocol + ICHOM costs were described. The total mean costs were highest in the first year after birth (€5596), mainly due to surgeries. The mean observed total costs (€40,859) for the complete treatment (0–24 years) were 1.6 times the expected protocol costs (€25,198) due to optional, non-protocolized procedures. Hospital admissions including surgery were the main cost drivers, accounting for 42% of observed costs and 70% of expected protocol costs. Implementing the ICHOM Standard Set increased protocol-based costs by 7%.

Keywords: Cleft lip; Cleft palate; Health care costs; Patient reported outcome measures; Physicians' practice patterns.

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In 2006, the publication of the book *Redefining Health Care: Creating Value-Based Competition on Results* by Porter and Teisberg initiated a paradigm shift in healthcare, and achieving high value for patients became the overarching goal of healthcare delivery, in which value is defined as health outcomes per dollar spent.^{1,2} As one of the value-based healthcare (VBHC) pilot centers in the Netherlands, the cleft team at Erasmus University Medical Center started measuring outcomes in patients with cleft lip and palate as part of their routine care. In 2016, the Standard Set for Cleft Lip and Palate (Standard Set), developed by the International Consortium for Health Outcomes Measurement (ICHOM),³⁻⁵ was implemented for this purpose. The Standard Set includes clinician-reported outcomes and patient characteristics, and incorporates the patient's perspective on health via multiple patient-reported outcome measures (PROMs) related to appearance, speech, and facial and psychosocial functioning.^{3,5,6} These domains are of special interest for patients with a cleft, because this congenital facial disorder causes feeding difficulties and impairs facial growth, articulation, dentition, and psychosocial health.⁷⁻⁹ Besides the addition of various PROMs to the treatment protocol, the Standard Set introduced an extra cleft team visit at 22 years, and additional audiological and speech examinations.^{3,5,6}

So far, research on VBHC initiatives in cleft has mainly focused on patient outcomes and how to measure and improve them. Medical costs as part of the VBHC equation have rarely been investigated. Most studies have focused only on specific parts of the cleft treatment with a relatively short follow-up period, such as surgical interventions, or have described costs at a highly aggregated health insurance level.¹⁰⁻¹⁵ Also, research has shown that treatment protocols for cleft vary widely, both nationally and internationally,^{16,17} but there are no published studies that have explored to what extent treatment protocols correspond with actual care provided. Furthermore, the Standard Set was developed to be implemented in routine care globally, but its adoption has been hindered by the belief that implementation will increase medical costs considerably,¹⁸ even though this assumption has not been investigated yet.

A better understanding of healthcare utilization patterns and medical costs during the complex and long treatment period for cleft lip and palate is essential for the following reasons: (1) to adapt care pathways adequately and efficiently, (2) to determine the 'value' of cleft lip and palate care based on the VBHC equation,^{1,2} and (3) to lead negotiations between health insurance companies and hospitals towards fair pricing for future payment strategy transformations, such as bundled payments.²

Therefore, the aim of this study was two-fold. Firstly, to describe the total healthcare use and direct medical costs of care for patients from 0 to 24 years old with a unilateral cleft lip and palate, and to compare this to the expected costs based on the treatment protocol. Secondly, to explore the additional protocol-based costs after implementation of the Standard Set.

Methods

This retrospective cohort study was conducted from the perspective of a Dutch academic hospital (Erasmus University Medical Center, Rotterdam). The data collected were recorded as part of routine care and were extracted from the patient's electronic health record (EHR) or the institution's information systems. Research ethics approval was granted by the Institutional Review Board of the Erasmus University Medical Center, Rotterdam, The Netherlands (MEC-2016-156).

Study population

Patients with a unilateral cleft lip and palate (UCLAP) between 0 and 24 years of age, treated by the cleft team at Erasmus University Medical Center between January 1, 2012 and December 31, 2019, were eligible. The UCLAP phenotype was chosen because this is the most complex and severe entity of cleft, and the unilateral variant is more common than the bilateral form.⁷ There were no exclusion criteria since the aim was to obtain a patient population representing real practice. Eligible patients were identified through the Zorgmonitor (English: healthcare monitor), a secured platform linked to the patient's EHR, for the collection of outcome data within the Erasmus University Medical Center.⁴ From all

identified patients with UCLAP, a group of 40 patients was randomly sampled to match the real patient population as closely as possible. This number was chosen for feasibility reasons, due to the time-consuming and labor-intensive nature of collecting and sorting all of the data.

Study parameters

First, the volume of cleft-related healthcare services delivered to the patients was counted. Healthcare services included medical consultations, diagnostics, and surgical procedures with hospital admissions. A detailed overview of the parameters collected is presented in [Table 1](#). Due to privacy legislation, requesting any type of information on externally performed cleft-related treatments, such as speech and language therapy or dental and orthodontic care, was not allowed.

Second, all observed direct medical costs were calculated using the following formula: costs = volume of healthcare service × price of the healthcare product.¹⁹ The prices of healthcare products were collected from the hospital's financial information systems in Euros and were based on the 2019 price allocations ([Table 1](#)). Prices for the years 2012–2018 were adjusted for inflation according to the Dutch price index percentages ([Supplementary Material Table S1](#)).²⁰

Third, since cleft care is highly protocolized, two treatment protocols followed by the cleft team at Erasmus University Medical Center were outlined to estimate care use and medical costs in the scenario where a patient solely followed one of the protocols: (1) the treatment protocol applicable before 2016, hereafter named the 'protocol', and (2) the treatment protocol expanded by the implementation of the outcome measures of the Standard Set, hereafter named 'protocol + ICHOM' (2016 onwards, and ongoing). Important additions to the protocol by the Standard Set included various psychosocial PROMs and extra audiological and speech testing (e.g. pure-tone, percentage of consonants correct) around the age of 8, 12, and 22 years, and an extra cleft team visit at the age of 22 years.^{3,5,6} The volume of care and costs based on the protocols do not include optional, non-protocolized surgeries and treatments due to complications. Details of both treatment protocols are presented in [Fig. 1](#).

Table 1. Overview of the healthcare services collected and the related mean rounded price allocations in 2019, as used in this study.

Healthcare services			Mean price
Medical consultations	Protocolized consultations	Cleft surgeon	€89
		Ear, nose, throat specialist	
		Orthodontist	
		Dentist (per 5 min)	
		Speech therapist	
	Optional, non-protocolized consultations	Specialized nurse	€165
		Social care worker	
		Psychologist	
		Screening CLEFT-Q (by psychologist)	
		Anaesthesiologist	
Diagnostic procedures	Medical imaging	Geneticist	€30
		Pediatrician	
		Psychosocial care during hospitalization	
		Medical photographs	
		Dental models	
	Audiological testing	Skull-profile X-ray photograph	€37
		Panoramic photograph	
		Tympanometry	
		Oto-acoustic emission	
		Tone audiometry (including pure-tone)	
Other procedures	Psychological examination	€195	
	Speech/language examination		
	Percentage of consonants correct (PCC, by speech therapist)		
	Naso-endoscopy		
	Closure of cleft lip		
Surgical procedures	Primary procedures	Closure of soft palate	€3038
		Closure of hard palate	
		Alveolar bone grafting	
		Pharyngoplasty	
		Grommet placement (per side)	
	Secondary or optional, non-protocolized procedures	Lip revision	€1415
		Septorhinoplasty	
		Le Fort I osteotomy	
		Implants	
		One day hospital admission	
Hospitalization		€795	

Observed healthcare use and costs data were linked to the year in which the service was delivered, or costs were made. General information on sex, age, adoption status, and presence of a genetic syndrome was also collected.

Statistical analysis

The observed healthcare use (including optional surgeries and treatments due to complications) was counted and the related medical costs were calculated. As it was not feasible to measure a 24-year care trajectory for each patient, the patients were followed up to 8 years within the study period. Observed mean care use and costs per person-year for

each year of the treatment trajectory were calculated and totaled to obtain overall healthcare use and costs of a full treatment trajectory from 0 to 24 years of age.

Consequently, the care and costs of the full treatment trajectory were broken down into six important phases based on the Standard Set time-points for outcome measurements: 0–4 years (no additional outcome measurements), 5–7 years, 8–10 years, 11–13 years, 14–21 years (no additional outcome measurements), and 22–24 years of age.⁶

Subsequently, mean observed costs were compared to expected costs based on the protocol to treat patients with

UCLAP. Further, the expected additional costs of protocol + ICHOM were described. All statistical analyses were performed using IBM SPSS Statistics version 24.0 (IBM Corp., Armonk, NY, USA).²¹ An overview of the terminology used in this paper is presented in [Table 2](#).

Results

In total, 40 patients with UCLAP contributed 301 observed person-years. Twenty-seven (67.5%) patients were male and 13 (32.5%) were female; five (13%) were adopted and four (10%) were diagnosed with a genetic syndrome.

Healthcare use

[Table 3](#) highlights the observed healthcare per age phase. The highest mean number of medical consultations was provided to patients in the 8–10 years ($n = 7$) and 11–13 years ($n = 8$) age ranges. The mean number of diagnostic procedures was highest in the age groups of 5–7 years ($n = 4$), 8–10 years ($n = 4$), and 11–13 years ($n = 4$). The highest mean number of surgical procedures performed was during the age ranges of 0–4 years ($n = 1$) and 8–10 years ($n = 1$), with the highest number of surgeries in the first year after birth ($n = 2$) and a mean total of 10 surgical interventions over the course of 24 years ([Supplementary Material Fig. S1](#)). An overview of observed counts per person-year for medical consultations and diagnostic procedures can be found in [Supplementary Material Figs. S2 and S3](#), respectively.

Observed costs and comparison with expected costs

The mean observed total costs for the treatment period from birth until 24 years (including optional, non-protocolized treatments due to complications) (€40,859) were 1.6 times the expected costs based on the protocol (€25,198). Mean total costs for observed care per patient based on a maximum of 8 years of follow-up were €11,809 (range €2616–33,323), with 50% of patients within the interquartile range (€6513–14,831). This distribution was similar for the adopted and syndromic patients together. Observed mean costs per person-year were €1702. The highest mean observed costs were made in the first year after birth and at

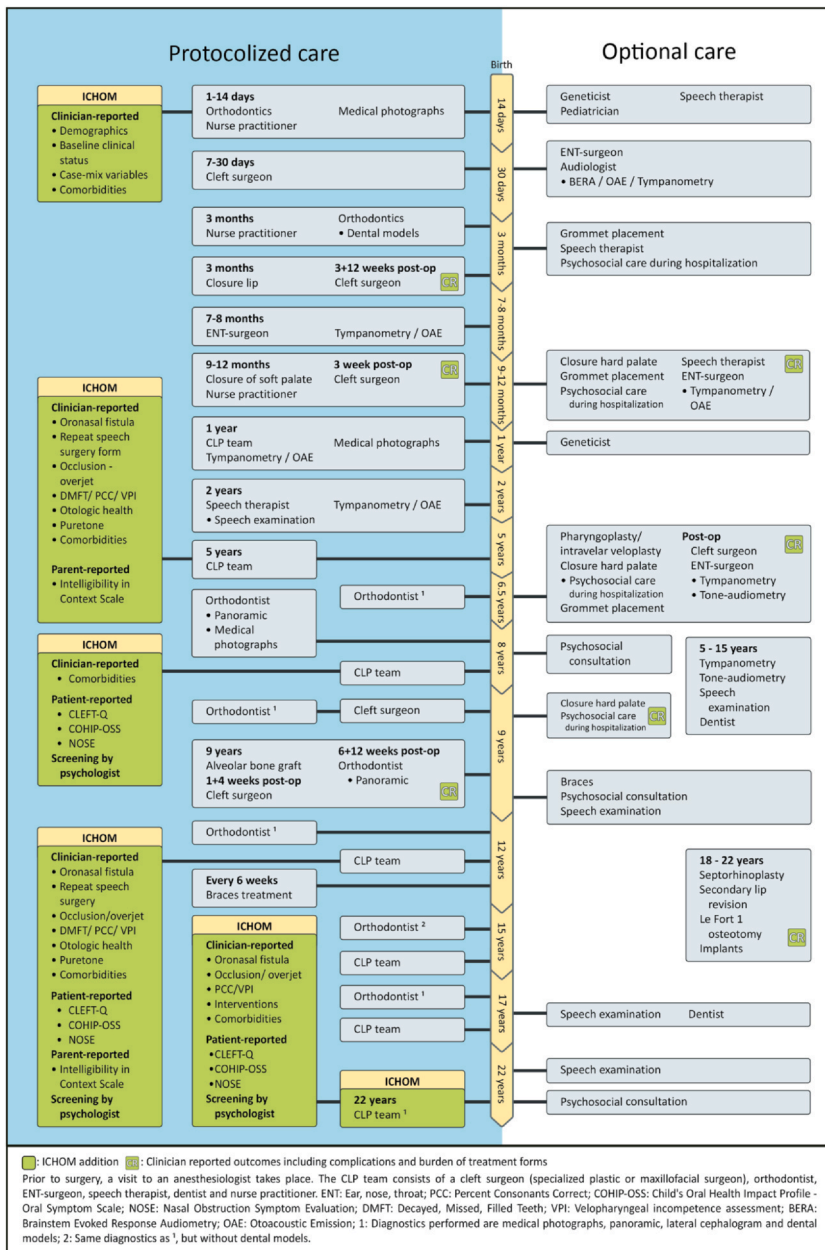


Fig. 1. Overview of treatment protocols for the treatment of UCLAP at the Erasmus Medical Center. Light blue boxes in the blue field indicate the 'protocol' and green boxes indicate the additions of the Standard Set, which together with the light blue boxes make the 'protocol+ICHOM'. The white field presents optional, non-protocolized treatments offered after diagnosing specific problems or needs.

the age of 11 years (€5596 and €3454, respectively; ratio 1:0.6). Clustering the data into age groups of 0–4 and 11–13 years, observed costs were €2681 and €2383, respectively (ratio 1:0.8) (Supplementary Material Fig. 4). Surgeries including hospital admissions accounted for 42% of total observed costs.

Based on the protocol, expected mean costs per person-year were €1050, and the highest costs were expected in

the first year after birth (€11,728) and at 9 years of age (€6237) (ratio 1:0.5), while no costs were expected at 11 years of age (Fig. 2). Surgical procedures including hospital admission accounted for 70% of total expected costs.

The addition of the Standard Set to the treatment protocol resulted in an increase of €1686 (7%) on total expected costs (€26,884). The additional team visit at 22 years accounted for 3% point of this increase.

Discussion

This study investigated healthcare utilization and costs of a long and complex treatment period for UCLAP and assessed the impact of the implementation of the ICHOM Standard Set on medical costs. In general, large variations between patients, between costs based on observed healthcare use and costs based on treatment protocols for cleft along 24 years of care were assessed; costs for the full treatment period from birth until young adulthood were 1.6 times the expected costs based on the treatment protocols. This is mainly due to multiple diagnostic and surgical procedures such as speech therapy, grommet placement, or septorhinoplasty that are not performed routinely but are offered after diagnosing specific needs or wishes. Even though it is known that procedures such as orthognathic surgery, dental implant placement, or septorhinoplasty benefit the countenance of patients as perceived by laypersons,^{22,23} the need to perform these procedures and their timing primarily depends on the patient's feelings, concerns, and wishes. Therefore, these procedures are defined as 'optional' in the local treatment protocol. Currently, the use of PROMs in clinical practice helps to identify areas of concern and act as a conversation starter during routine medical consultations to discuss the patient's worries and wishes and related possible interventions.¹⁸

The high number of medical consultations and surgical procedures, including closure of the lip and soft palate, performed during the first year after birth are probable causes for the high expenditure. Previous micro-costing research by Abbott et al.^{12,24} presented similar results, with costs ranging from \$35,826 to \$56,611 for various subtypes of cleft lip and palate during the first 18 months in life, and costs ranging from \$10,426 to \$16,115 in patients with cleft lip. The majority of these first year of life costs stemmed from inpatient care, i.e. surgical procedures and hospital admissions.^{12,24} Additionally, Boulet et al.²⁵ reported that mean expenditures for children with a cleft in the USA decreased with increasing age, starting at \$95,819 for infants and decreasing to approximately \$5054 at 7–8 years of age. Of note, the costs of cleft treatments described by Boulet et al. and Abbott et al. are much higher than those presented here from Erasmus University Medical Center.^{12,24}

Table 2. Overview of terminology with descriptions, as used in this paper.

Terminology	Description
Healthcare services/use (cleft-related)	Medical consultations, diagnostic procedures, surgical procedures, and hospital admissions, as registered by the cleft team at Erasmus University Medical Center; see Table 1 for more details
Observed costs	Calculated with the formula of costs = volume of observed healthcare service utilization × price of the healthcare product
Expected costs	Calculated with the formula of costs = volume of expected healthcare services based on the treatment protocol × price of the healthcare product
Protocol	The treatment protocol for cleft lip and palate that was employed before 2016
Protocol + ICHOM	The treatment protocol for cleft lip and palate employed from 2016 onwards, including additional consultation and diagnostics introduced by the local implementation of the ICHOM Standard Set for Cleft Lip and Palate

ICHOM, International Consortium for Health Outcomes Measurement.

These differences may be explained by the fact that (1) data on costs were collected from (private) health insurance companies, (2) costs were not limited to cleft-related healthcare, (3) treatment protocols differ between hospitals, and (4) healthcare costs in general might be higher in the USA than in The Netherlands, due to differences in healthcare organization and insurance strategies.

An unanticipated finding was that observed costs in the first year after birth were lower than expected based on the protocol, in contrast to the subsequent years in which costs were higher than expected. Interventions protocolized within the first year of life are sometimes spread over a longer period due to planning difficulties or late referrals. A similar pattern was seen at a later stage of care; higher expenditures were expected at 9 years of age based on the protocol, due to the alveolar bone grafting procedure and orthodontic treatments. In practice, the timing of alveolar bone grafting is dependent on the dental development status of the child, resulting in higher costs between 10 and 12 years.^{26,27} Evaluating costs of a long period of complex care might benefit from clustering multiple years, since the start of an intervention may vary and treatments such as orthodontics or speech therapy may continue for multiple months, or even years.

With the implementation of the Standard Set, additional speech, audiological and psychosocial

screening, and an extra cleft team visit were introduced.^{3,6,28} Aside from these clinical encounters, patients at 8, 12, and 22 years of age were asked to complete PROMs at home prior to their visit.^{5,6} The use of PROMs could provide insight into a patient's perspective on their functioning and well-being, and detect concerns or problems that would otherwise remain undiscussed. Tackling healthcare problems and improving quality of life early on could potentially reduce complication rates and treatment costs in the long run. Even though implementation of the Standard Set may lead to increasing expenditure, an increase in value and concomitantly cost-effectiveness can still be attained by improving patient outcomes. Due to the complex character of cleft lip and palate and the need for long-term care into young adulthood, patience is asked of clinicians, researchers, and policy-makers before the cost-effectiveness and potential value-improvement can be reliably examined. Meanwhile, measuring outcomes could be utilized to improve patient-centered care, shared decision-making, and local quality improvement endeavors.^{18,29}

This research highlights the heterogeneity in healthcare use and medical costs for patients with a cleft lip and palate. Care consumption and costs varied widely, with 50% of patients outside the interquartile range. Surgical procedures were expected to be responsible for 70% of medical care costs

based on the protocol; however the actual surgical costs were found to be much lower (42%), suggesting that additional consultations and diagnostics are more often needed than expected. Consequently, solely relying on cost estimates based on a cleft treatment protocol to reform payment strategies or to lead negotiations between hospitals and health insurance companies should be done with caution. Further, understanding the patterns of healthcare use aids in determining the most efficient treatment pathways. For example, knowledge of the clinician's consultation burden could guide reorganization of the cleft protocol and team; it might be needed for a specialist to be more (or less) often available for consultations, or at different time-points during the treatment trajectory. Further research is needed to specify predictors for the variability in healthcare consumption, such as cleft type, family circumstances, and socio-economic status, in order to target individuals in need of more extensive care and enabling risk stratification.³⁰ The methodology described in this paper could be a useful first step in mapping and gaining insights into healthcare use and medical costs at a local level.

A unique point of this study is the evaluation of healthcare utilization and costs for a challenging and complex treatment trajectory in cleft lip and palate patients, with a long follow-up time of 8 years to reconstruct a full treatment trajectory of 24 years. In

Table 3. Counts per person-year of medical consultations, diagnostic procedures, and surgical procedures for the various age groups.

	Age, years					
	0–4	5–7	8–10	11–13	14–21	22–24
Medical consultations	4	3	7	8	6	4
Diagnostic procedures	4	4	4	4	3	2
Surgical procedures	1	0	1	0	0	0

Treatment protocol	Age during care trajectory												
	0	1	2	3	4	5	6	7	8	9	10	11	12
"protocol" *†	€ 11,728	€ 717	€ 433	€ 0	€ 0	€ 626	€ 1647	€ 0	€ 717	€ 6236	€ 0	€ 0	€ 1155
"protocol+ICHOM" *†	€ 11,728	€ 717	€ 433	€ 0	€ 0	€ 883	€ 1647	€ 0	€ 795	€ 6236	€ 0	€ 0	€ 1490

Treatment protocol	Age during care trajectory												Total costs
	13	14	15	16	17	18	19	20	21	22	23	24	
"protocol" *†	€ 352	€ 0	€ 781	€ 0	€ 804	€ 0	€ 0	€ 0	€ 0	€ 0	€ 0	€ 0	€ 25,198
"protocol+ICHOM" *†	€ 352	€ 0	€ 781	€ 0	€ 804	€ 0	€ 0	€ 0	€ 0	€ 1015	€ 0	€ 0	€ 26,884

Fig. 2. Costs based on the 'protocol' and 'protocol + ICHOM' per time-point of care. *Costs based on 2019 prices only. †Costs of closure of the hard palate are divided over 0, 6, and 9 years due to varying surgical timings.

addition, exploring the additional costs due to the implementation of the Standard Set for cleft care has not been done before. In contrast to previous cost studies focusing on aggregated data, the present study presents healthcare use and costs per individual. The latter was possible because the sample size was relatively small, which at the same time limited the possibility of adjusting for potential confounders, such as adoption status or the presence of a genetic syndrome.

In-hospital pricing for services and interventions was used for the calculation of costs. These prices depend on both the total volume of care delivered and on a department's own preferences on how to attribute costs to healthcare items. For example, the costs for administrative personnel or the utilization of rooms for outpatient clinic visits can be attributed to a general overarching cost item within a department, or to one specific cost item such as a medical consultation. This approach results in price differences between years and between departments within one hospital, but potentially also between hospitals and countries. As a result, costs should be interpreted as estimates rather than exact numbers, and extrapolation of costs to other cleft care practices should be done with caution.

Further, this study only included healthcare use and direct costs from an academic healthcare provider's perspective. It was not possible to include costs such as patient out-of-pocket expenses, medication costs, costs of out-of-hospital treatments such as speech and language therapy, psychosocial care or dental and orthodontic care, travel costs, loss in work productivity of parents, and costs of administrative personnel.^{14,19} Also, the costs for the implementation of the Standard Set itself were not incorporated. Therefore, costs described in this paper are most

likely an underestimation of the true economic burden of cleft care.

In addition, this study was conducted in a specialized cleft center where various medical specialists work together in an integrated practice unit ('cleft team'). However, in some geographic areas, cleft care is not provided by such a coordinated team but rather by individual clinicians, limiting the generalizability of the study findings and hampering payment reform strategies.³¹

Performing a cost-effectiveness evaluation was hampered because outcome measures were not routinely collected in clinical practice before the implementation of the Standard Set. Consequently, no conclusions can be drawn as to whether the extra costs can be weighed against the effects of the implementation, for example in terms of patient satisfaction with treatment or better patient outcomes and quality of care. This issue deserves further study and could be a promising opportunity for centers that are planning to implement the Standard Set in their clinical practice.

In conclusion, a large variety of healthcare use and medical costs was found among patients with a UCLAP and throughout the cleft treatment trajectory, with the highest costs in the first year of life. Observed costs for the treatment from birth until young adulthood were 1.6 times the costs based on protocols, due to a wide range of secondary diagnostics and surgeries performed. Surgical procedures were found to be the main cost drivers, while the increase in medical costs due to the implementation of additional assessments, as defined by the ICHOM Standard Set for Cleft Lip and Palate, was 7%.

Ethical approval

Institutional Review Board of the Erasmus University Medical Center,

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Competing interests

None.

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Patient consent

Not required.

Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at [doi:10.1016/j.ijom.2023.08.007](https://doi.org/10.1016/j.ijom.2023.08.007).

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Correspondence to: Department of Oral and Maxillofacial Surgery
Erasmus University Medical Center
NA-building
room 2401
Postbus 2040
3000 CA Rotterdam
the Netherlands.
Tel:+31 10 70 341 38. Fax:
+31 10 70 368 44.
E-mails: inge.apon@gmail.com,
i.apon@erasmusmc.nl