

The Social Cost of Nasal Sequelae in Patients with Cleft Lip and Palate in a Peruvian University Dental Clinic

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Objective: To estimate the social cost of nasal sequelae in patients with cleft lip and palate (CL/P) treated in a Peruvian university dental clinic.

Methods: This is a cross-sectional study. The sample consisted of thirty patients over 18 years of age with nasal sequelae due to CL/P. This study conducted a partial economic analysis from a social perspective, by using the following questionnaires: ENAHO-Peru to assess sociodemographic variables, the Instituto de Salud del Niño - San Borja care guidelines and its tariff schedule to estimate health care costs, the Rhinoplasty Outcome Evaluation (ROE) scale to assess quality of life about the nasal sequela, and the “willingness to pay” (WTP) technique to calculate the monetary cost of the sequela from the patients’ perspective. Costs in soles, where the exchange rate was: 1 USD = S/3.878.

Results: A total of 30 CL/P patients, who agreed to participate, were evaluated. Most of the patients were male (70.0%), aged 18–39 years (73.0%), single (93.4%), students (73.4%), and had secondary education (50.0%). Additionally, most were attended with Seguro Integral de Salud (46.7%), most had no daily income (50.0%), and the average quality of life score was 8.1. The highest direct cost reported was dental treatment (S/5756.89 ± S/359.22) and hospitalization (S/5013.60 ± S/880.15), statistically significant ($p < 0.05$). The highest indirect cost was reported for absenteeism with a mean of 5288.6 ± 1280.23 ($p < 0.05$). Regarding direct, indirect, and intangible costs in the treatment of nasal sequelae due to CL/P, the lowest median (S/6000.00) was found in intangible costs, with significant differences ($p < 0.05$).

Conclusion: About half of the social cost (12,000 Peruvian soles = 3094 US dollars) was assumed by the patients, a prohibitive cost considering that most of them come from low socio-economic backgrounds.

Plain Language Summary:

- Cleft lip and palate (CL/P) cause a disproportionate economic burden. Despite advances in surgery, these patients very often develop sequelae.
- No health insurance in Peru covers rhinoplasty, as it is considered a surgery to improve the image and not necessarily function.

This research estimated that the patients with CL/P bear almost half of the social cost. This could have implications for state insurance mechanisms that could improve coverage of surgical procedures aimed at improving the sequelae of CL/P, which would reduce the social cost borne by patients.

Keywords: cleft lip, cleft palate, social cost, sequela, health economics

Introduction

Globally, cleft lip and palate (CL/P) accounts for more than 90% of congenital malformations in humans.^{1,2} This malformation causes a disproportionate economic burden in low- and middle-income countries.¹ Despite advances in surgery and timely multidisciplinary management of CL/P, these patients very often develop sequelae.^{3,4}

The most important from an aesthetic point of view is the nasal sequelae.^{5–7} These sequelae vary due to the type and width of the fissure and are affected by the type and width of CL/P, but also by the age of the patient and the surgical technique used.^{8–10} As it is in the center of the face, the nasal sequela generates a great psychological toll on the patient that ultimately results in a loss of quality of life.^{11–13} Given that facial aesthetics are of universal importance, especially between the ages of 18 and 30, most adults who have undergone CL/P surgery seek additional treatments, mainly for the nose, such as rhinoplasty.^{6,7}

On the other hand, the correct allocation of economic resources is made according to a cost analysis of a given product or service. Costs will always depend on the context and the decision-maker at the time.¹⁴ Costs can be direct, indirect, and intangible. Direct costs are associated with the health systems' own resources, such as public or private health care. Indirect costs are related to the valuation of the use of the patient's or companion's lost time (transfer, waiting, recovery, etc.) in terms of their productivity and salary. Intangible costs are related to the loss of the patient's quality of life. However, the social cost of a disease is a measure of economic analysis (cost-benefit) that includes the sum of tangible and intangible costs, which arise during a pathology and affect all individuals in society.^{14–16}

In Peru, there are several public and private institutions committed to the care of patients with CL/P. However, rhinoplasty is not currently included in the optimal surgical chronology for CL/P patients because it is usually considered a “cosmetic procedure”. In addition, there is a resource-constrained healthcare environment, where the correct allocation of financial resources plays a key role in decision-making.¹⁴

There are no specific data on monetary estimates of the cost to society of the nasal sequelae of CL/P. An estimate of the monetary value of these patients will be useful in raising awareness and sensitization of society and the relevant authorities for the correct decision-making in health policies in the medium and long term. For these reasons, the aim was to estimate the social cost of nasal sequelae in patients with CL/P treated in a Peruvian university dental clinic.

Materials and Methods

Cost Study

For this study, the cost of illness method was used to estimate the cost of CL/P sequelae, which takes into account the actual cost without considering the outcomes of the interventions.

Participants

The sample consisted of thirty patients with nasal sequelae due to CL/P attended at the University Dental Clinic of the Universidad Peruana Cayetano Heredia (UPCH) between January 2016 and December 2020, who met the following inclusion criteria: post-operative patients with CL/P, both sexes, over 18 years of age and who had some nasal sequelae. The study was previously approved by the Institutional Research Ethics Committee (CIEI) - UPCH, registration No. 102777, R-E071-15-20.

Data Collection

Before the study, permission was requested from the statistics department of the University Dental Clinic of the Faculty of Stomatology-UPCH to access the database of patients diagnosed with CL/P in the period mentioned. The information was collected prospectively using a telephone survey of all participants who met the selection criteria, who answered the survey voluntarily and with prior informed consent. Since data collection was carried out using a telephone survey, all nasal alterations reported by patients after primary CL/P surgery were classified as sequelae.

Costs (direct, indirect, and intangible) were expressed in soles. To calculate direct costs, information was obtained on the frequency of use and the cost of each of the resources paid according to the INSN-SB clinical practice guideline on CL/P (2017).¹⁷ Direct costs include surgery, hospitalization, medical consultations, laboratory tests, and imaging for each patient. This cost information was extracted from the INSN-SB (2019) tariff published in RD N° 063/2019/INSN-SB,¹⁸ considering the rates for public agreement. To calculate the indirect or opportunity costs, the INSN-SB care guide was also used, which specifies the average number of days of hospitalization, days of outpatient care, days with limited activity, and patient absenteeism, to then make a monetary valuation for each patient. Indirect costs were related to the monetary valuation of the

work activities of patients or accompanying persons. Finally, the valuation of intangible costs was carried out using the WTP technique, which was measured using the “open question” method.¹⁹

Instruments

A questionnaire consisting of three sections was used: the first section comprised questions related to socio-economic variables, the second section included questions related to quality of life and the last section on intangible costs. The questions in the first section were based on the validated ENAHO-Peru (2017)²⁰ questionnaire. The questions in the second section were the same as those of the ROE scale “Rhinoplasty Outcome Evaluation” Spanish version (2013).^{12,21} The ROE questionnaire is a validated, reliable, and acceptable instrument used to measure quality of life specific to the nasofacial condition. It consists of 6 Likert-scale questions involving social aspects of nasal deformity, nasal aesthetics, nasal function, and awareness of nasal deformity.^{12,21,22} The questions in the third section were related to direct, indirect and intangible costs, the latter being obtained with the methodology called “willingness to pay” (WTP) which is validated for use in health economics and frequently used to express the maximum amount a consumer would pay to purchase a certain good or a user to have a certain service.^{23,24} Respondents were asked in an open-ended way what would be the maximum value they would be willing to pay - in Peruvian soles (S/) of the year 2021 - to have no evident nasal sequelae due to CL/P. When subjects were not willing to pay, it was recorded as zero.

Statistical Analysis

Descriptive analysis was performed by calculating frequencies for qualitative variables, measures of central tendency and dispersion for quantitative variables. For the bivariate analysis for qualitative variables and to evaluate the association, Fisher’s exact test was used; for quantitative variables, the normality of the data and homogeneity of variances were previously evaluated; in case of non-compliance, non-parametric tests were used to find differences between two groups, the Mann–Whitney *U*-test and the Kruskal-Wallis *H*-test were used to compare three or more groups, with a significance level of 0.05 for the hypothesis contrast. Statistical analysis was conducted using STATA statistical software version 16 (StataCorp LLC, Texas, USA).

Results

A total of 30 CL/P patients, who agreed to participate, were evaluated. Most of the patients were male (70.0%), aged 18–39 years (73.0%), single (93.4%), students (73.4%), and had secondary education (50.0%). Additionally, most were attended with Seguro Integral de Salud (46.7%), most had no daily income (50.0%), and the average quality of life score was 8.1. The description of the sociodemographic characteristics, type of sequelae, and daily admissions with nasal sequelae due to CL/P of the sample are shown in [Table 1](#).

In the direct economic costs in soles for treatment of nasal sequelae due to CL/P, it found that the high cost reported was dental treatment (S/5756.89 ± S/359.22), it was statistically significant ($p < 0.05$); followed by hospitalization costs (S/5013.60 ± S/880.15). Meanwhile, regarding the indirect economic costs in soles for treatment of nasal sequelae, the highest cost was reported for work absenteeism (S/5288.6 ± S/1280.23), it was statistically ($p < 0.05$); these findings are shown in [Table 2](#).

When each patient was asked how much they would be willing to pay to have no obvious nasal sequelae due to CL/P, 12 patients answered S/9001 to more and 7 answered S/0 to 3000. [Table 3](#) shows the direct and indirect costs for the treatment of nasal sequelae of CL/P according to type of sequela. No significant differences were found in these costs in the manner of the type of sequela ($p > 0.05$).

Regarding direct, indirect, and intangible costs in the treatment of nasal sequelae due to CL/P, the lowest median (S/6000) was found in intangible costs, and significant differences were found between these costs, as shown in [Table 4](#).

Discussion

This research estimated the social cost of nasal sequelae in CL/P patients evaluated in a university dental clinic in Peru between 2016 to 2020, finding that the patients bear almost half of the social cost.

The nasal sequelae of CL/P are a clear example of the unwillingness of the health sector to provide these patients with access to an improvement in their quality of life and unfortunately affects the lower socioeconomic sectors of the

Table 1 Sociodemographic, Economic, and Quality of Life Characteristics in the Sample

Characteristics	n (%)
Sex	
Female	9 (30.0)
Male	21 (70.0)
Age	
18 to 29 years	22 (73.0)
30 to 59 years	8 (27.0)
Marital status	
Single	28 (93.4)
Cohabitant	1 (3.3)
Married	1 (3.3)
Occupation	
Student	22 (73.4)
Self-employed	1 (3.3)
Employee	4 (13.3)
Other	3 (10.0)
Education level	
Incomplete Primary	2 (6.7)
Incomplete Secondary	7 (23.4)
Complete Secondary	8 (26.6)
Incomplete Technical Higher Education	9 (30.0)
Complete Technical Higher Education	2 (6.7)
Incomplete University Higher Education	1 (3.3)
Complete University Higher Education	1 (3.3)
Type of Insurance	
Seguro Integral de Salud (SIS)	14 (46.7)
Seguro Social de Salud (ESSALUD)	11 (36.6)
None	5 (16.7)
Daily income with nasal sequelae due to CL/P (In soles)	
None	15 (50.0)
200.00	2 (6.7)
125.00	2 (6.6)
75.00	3 (10.0)
30.00–50.00	8 (26.7)
Quality of Life Questionnaire Score	8.1 ± 2.6

Notes: Mean ± Standard Deviation. Daily income in Peruvian soles in 2021, where the exchange rate was 1 USD = S/3.878.

population the most. No health insurance in Peru covers cosmetic rhinoplasty, as it is considered a surgery to improve the image and not necessarily function. However, these patients with nasal sequelae have an affected image, and their faces can hardly fulfill an adequate social function when interacting.

The most common type of nasal sequelae found were nasal deviation, lack of nasal tip projection and asymmetry of one nostril. These results are similar to those found in some studies.^{9,10,25} Many patients had unilateral CL/P, which explains why there was a nasal septal deviation, asymmetry of the nostril affected by CL/P and lack of nasal tip projection. Other research also points to unilateral CL/P as the most frequent form of presentation.^{9,10,25,26}

Dental treatment not only improves oral function but also nasal aesthetics and function; this is achieved through orthodontics, maxillary expansion, or dental prostheses performed on patients with nasal sequelae after primary CL/P surgery. The most basic dental treatment in these patients includes orthodontics, which can correct malocclusions and dental misalignments that lead to facial asymmetries and worsen nasal appearance. In addition, maxillary expansion

Table 2 Direct and Indirect Economic Costs for Nasal Sequelae Treatment Due to Cleft Lip and Palate

Economic Costs (In Soles)	Mean $\pm$ SD	p
Direct costs for		0.001 ^a
Consultation	143 $\pm$ 18.64	
Medicines/supplies	379.11 $\pm$ 81.03	
Laboratory/tests	227.33 $\pm$ 47	
Dental treatment	5756.89 $\pm$ 359.22	
Hospitalization	5013.6 $\pm$ 880.15	
Transportation	382.67 $\pm$ 10.01	0.0001 ^b
Indirect costs for		
Family expenses	54 $\pm$ 0	
Work absenteeism	5288.6 $\pm$ 1280.23	

Notes: ^aKruskal–Wallis *H*-test, $p < 0.05$. ^b Mann–Whitney *U*-test. Costs in soles, where the exchange rate was: 1 USD = S/3.878.

Table 3 Direct and Indirect Economic Costs in Soles for Nasal Sequelae Treatment Due to Cleft Lip and Palate by Type of Sequela

Type of Sequela	Direct Cost		Indirect Cost	
	Mean $\pm$ SD	p*	Mean $\pm$ SD	p*
Nasal deviation + lack of tip projection + nasal fossa asymmetry	11535.1 $\pm$ 1017.1	0.370	4549 $\pm$ 1590.7	0.070
Lack of tip projection	12359.6 $\pm$ 3.5		6037 $\pm$ 0	
Lack of tip projection + alar retraction + short columella	12017.5 $\pm$ 907.6		5611.9 $\pm$ 1124.8	
Lack of tip projection + short columella	10800 $\pm$ 1350.3		4053 $\pm$ 1718.2	
Nasal deviation + nasal fossa asymmetry	12348.2 $\pm$ 26.6		6037 $\pm$ 0	
Nasal deviation + lack of tip projection	12358.6 $\pm$ 2.4		6037 $\pm$ 0	
Total	11902.6 $\pm$ 892.9		5342.6 $\pm$ 1280.2	

Notes: *Kruskal–Wallis *H*-test, $p > 0.05$ not significant. Costs in soles, where the exchange rate was: 1 USD = S/3.878.

Table 4 Total Cost of Nasal Sequelae Treatment Due to Cleft Lip and Palate

Costs	Mean	SD	Median	IQR	Min	Max	p
Direct	11902.6	892.93	12,357.6	46	9959.2	12,363.56	0.001*
Indirect	5342.6	1280.23	6037	0	3061	6037	
Intangible	6766.7	4492.78	6000	6000	0	18,000	
Total	24011.9	3925.31	23,730.38	5625.6	13,020.2	31,144.2	

Notes: *Kruskal–Wallis *H*-test, $p < 0.05$ significant. Costs in soles, where the exchange rate was: 1 USD = S/3.878.

widens the nasal cavity and thus improves breathing and nasal shape. Finally, stabilization of the dental arch through orthodontics facilitates additional surgical interventions.^{10,25,26}

The highest direct cost was for dental treatment and hospitalization. Being a pathology affecting the lower third of the face, much of the treatment involves oral rehabilitation and orthodontics, which would explain the reason for the high cost. Hospitalization is also related to primary lip or palatal surgery. Almost the entire indirect cost was due to the absenteeism of family members. These data are in line with the background found about the social cost found in different pathologies.^{27–30} The monetary valuation of indirect costs is related to the income reported by patients and their direct relatives or companions. This figure could be higher over time due to the variability of income reported by patients or relatives, especially those working in the informal sector.

The difference found between the direct costs, which are higher than the indirect costs - which are those borne by the patient or the families - is not in line with the precedents reviewed.^{27–30} This result could be because most of the population was under 30 years of age. Another explanation could be the socioeconomic level, as CL/P affects more the lower-income sectors of the population. Therefore, the assessment of monthly income will vary according to occupation.

Many patients were willing to pay to have their nasal sequelae resolved. This figure is considerable for the socio-economic background of the patients and their occupation - mostly students who were financially dependent on their relatives. It is worth mentioning that all patients reported having undergone free CL/P surgery in public hospitals or surgical campaigns carried out by non-governmental organizations.

To the best of our knowledge, no studies were found that applied the described methodology to estimate the social cost of nasal sequelae - frequent in patients with CL/P. Furthermore, it emphasizes the need to correct this sequela to reverse the economic situation of sufferers. A simple understanding of the results will allow for improved resource allocation and decision-making. However, it will also raise the awareness of the different social actors, especially specialists, who can give due priority to these cases.

In the present study, the finding of a small population in the five-year registry of patients seen at the clinic suggests that it is a difficult population to capture. This could be related to a market failure, which under-records the nasal sequela and underestimates the burden of disease. The latter is measured by the effects of the disease from a medical point of view, ie, it expresses medical preferences, whereas in quality of life it is the patient who expresses his or her preferences. Although the study was only conducted in Lima and in a single health facility, which denotes an important limitation for the extrapolation of data at the national level, it is possible that these results could be useful for other facilities with similar conditions. Another limitation during the development of the study was the COVID-19 pandemic, which changed the survey methodology to a telephone survey and prolonged data collection. Although direct costs related to CL/P surgeries or treatment of other co-morbidities during CL/P were not considered, these results generate a minimum estimate of the resource costs borne by patients or their families.

Conclusion

About half of the social cost (12,000 Peruvian soles = 3094 US dollars) was borne by the patients. In addition, direct health costs, normally borne by health insurers, accounted for more than half of the social cost, and were mostly derived from dental treatment and hospitalization. Indirect costs related to the loss of productive time of patients or accompanying persons were half of the direct costs, but this loss increases according to occupation and socio-economic status.

This could have implications for state insurance mechanisms that could improve coverage of surgical procedures aimed at improving the sequelae of CL/P, which would reduce the social cost borne by patients.

Data Sharing Statement

Data may be made available by contacting the corresponding author.

Ethics Approval and Informed Consent

The study was previously approved by the Institutional Research Ethics Committee – Cayetano Heredia Peruvian University (Registration No. 102777, R-E071-15-20). Because the study was conducted during the COVID-19 pandemic when the Peruvian population was in mandatory social isolation (quarantine), oral consent was obtained from participants before conducting the telephone survey, which was approved by the institutional ethics committee. This reduced the risk of COVID-19 infection for participants. The confidentiality of the information obtained was guaranteed and used only for this study's purposes. The authors confirm that the guidelines outlined in the Declaration of Helsinki were followed.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Acknowledgement

This paper was uploaded as (English translation), “Social Cost of the Nasal Sequel in Patients with Cleft Lip and Palate, in the Teaching Dental Clinic of the Universidad Peruana Cayetano Heredia” as a thesis to the Universidad Peruana Cayetano Heredia repository in 2023.³¹

Disclosure

The authors declared that they have no conflicts of interest in this work.

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