



## Evaluation of pain perception in patients of 0-5 months of age with cleft lip and palate subjected to pre-surgical orthopedics

### *Evaluación de la percepción del dolor en pacientes de 0 a 5 meses de edad con labio y paladar hendido sometidos a tratamiento de ortopedia prequirúrgica*

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#### ABSTRACT

**Introduction:** While it is considered that newborns are not able to verbalize feelings and express pain during pre-surgical orthopedic treatment, it is necessary to identify a primary measure of pain in pediatric patients with cleft lip and palate. **Objective:** The objective of the study was to assess pain during pre-surgical orthopaedic treatment using the FLACC scale, which assesses the expression of the face, legs, activity, scream and compression, and to determine the reliability of this scale in these patients. **Material and methods:** Six independent observers assessed pain through video recordings of 20 children aged 0 to 5 months during three stages or moments of time: 1. During feeding technique; 2. While taking impressions of the alveolar ridges; 3. During the placement of the pre-surgical orthopaedic appliance. This scale was scored at each stage during the procedure. During the evaluation of the impression taking and placement of the appliance the values were compared with the feeding technique using the paired Wilcoxon test. Intraclass correlation coefficients were also used to assess the reliability of the scale. **Results:** Statistically significant increases in scale scores were found during impression taking ( $p < 0.000$ ) and appliance placement ( $p < 0.000$ ) compared to the feeding technique. The reliability of the scale was rated as good (0.71-0.90). **Conclusions:** Pain was evident according to the FLACC scale during impression of the alveolar ridges and placement of the appliance prior to the patient's cheiloplasty. In addition, the scale showed good reliability, which provides preliminary support for considering alternative or pharmacological procedures to complement the patient's treatment.

#### RESUMEN

**Introducción:** Puesto que se considera que los recién nacidos no son capaces de verbalizar los sentimientos y expresar el dolor durante el tratamiento prequirúrgico ortopédico, es necesario identificar una medida primaria del dolor en pacientes infantiles con labio y paladar hendido. **Objetivo:** El objetivo del estudio fue calificar el dolor durante el tratamiento de ortopedia prequirúrgica utilizando la escala de FLACC, que evalúa la expresión de la cara, las piernas, la actividad, el grito y la compresión, además de determinar la fiabilidad de dicha escala en estos pacientes. **Material y métodos:** Seis observadores independientes evaluaron el dolor a través de video grabaciones de 20 niños de 0 a 5 meses durante tres etapas o momentos de tiempo: 1. En la técnica de alimentación; 2. En las impresiones de las crestas alveolares; 3. En la colocación del aparato ortopédico prequirúrgico. Dicha escala se puntuó en cada etapa de tiempo durante el procedimiento. Durante la evaluación de la toma de impresión y colocación del aparato ortopédico, los valores se compararon con la técnica de alimentación, utilizando la prueba de Wilcoxon pareada. También se emplearon coeficientes de correlación intraclass para evaluar la confiabilidad de la escala citada. **Resultados:** Se determinó que hubo incrementos estadísticamente significativos en las puntuaciones de la escala durante la toma de impresión ( $p < 0.000$ ) y la colocación del aparato ortopédico ( $p < 0.000$ ) en comparación con la técnica de alimentación. La confiabilidad de la escala fue catalogada como buena (0.71-0.90). **Conclusiones:** El dolor fue evidente de acuerdo con la escala de FLACC durante la toma de impresión de las crestas alveolares y la colocación del aparato ortopédico previos a la queiloplastia de los pacientes. Además, la escala mostró una buena confiabilidad, la cual brinda un apoyo preliminar para considerar procedimientos alternativos o farmacológicos para complementar el tratamiento del paciente.

**Key words:** Cleft lip and palate, FLACC scale, pain.

**Palabras clave:** Labio y paladar hendido, escala de FLACC, dolor.

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## INTRODUCTION

Cleft lip and palate (LPH) is a congenital defect of the structures of the mouth. It is a cleft in the lip and/or palate<sup>1</sup> that occurs in 1 out of every 750 live newborns and hinders feeding, language and social adaptation functions, as well as impacting the psychological aspect.<sup>2</sup> When talking about pain in the paediatric patient, anatomical and physical reasons should not only be taken into account: it is known that the perception of pain, in addition to being a biological phenomenon, is also influenced by psychological and child environment aspects that affect and modulate the nociceptive sensation.<sup>3-5</sup>

The assessment of pain during preoperative orthopaedic procedures in children with cleft lip and palate is an issue that should be evaluated in terms of outcome and treatment effectiveness.

The diagnosis of pain during any type of treatment during this period is controversial as it is sometimes difficult to determine whether it is psychological or anatomical. However, some scales have been developed to evaluate it, such as FLACC (Face, Legs, Activity, Cry, Consolability), a tool developed by Merkel and colleagues<sup>6</sup> to measure postoperative pain in children between two months and seven years of age, which is considered valid, reliable and feasible to use in a variety of settings, including: minor non-invasive procedures; ear, nose or throat operations; pain from surgery, trauma, cancer or other disease processes; pain suffered by critical patients; and postoperative pain in children with cognitive impairment.<sup>6</sup> The scale is scored in the range 0-10, where 0 represents no pain. It has five criteria with a score of 0, 1 or 2. The five scores are summed and the severity of the pain is determined on the basis of the pain scale 0-10.<sup>2,6-8</sup>

Whenever possible, measurement of pain should be used in combination with self-report. The objective of this study was to evaluate the perception of pain in pediatric patients in the pre-surgical orthopedic stage by means of the FLACC scale in patients with cleft lip and palate from 0 to 5 months, in order to determine the reliability of the FLACC scale as a pain assessment instrument with the participation of different observers.

## MATERIALS AND METHODS

### Sample selection

A longitudinal and prospective observational study was performed in patients aged 0 to 5 months, for which a sample of 20 research subjects was formed, in this case 14 girls and 6 boys. The sample included

12 children with unilateral cleft lip and palate-5 with right cleft lip and palate and 7 with left cleft lip and palate-. In addition, 8 bilateral cleft lip and palate were also included in the sample. The average age was  $5.75 \pm 4.58$  weeks and all of them were patients who came to the Dentistry and Orthodontics service of the «Dr. Manuel Gea González» General Hospital to begin pre-surgical orthopedic treatment in preparation for cheiloplasty.

The sample size was calculated using the Biostatistics program (Glantz, Stanton A., San Francisco CA, USA), with an alpha level of 0.05, calculating a test power at 80%, which yielded a sample of 17 patients. However, for this study, the sample was made up of 20 patients from whom a file was compiled, in addition to obtaining the informed consent signed by the parent or guardian.

### Description of the procedure for the FLACC evaluation

The mother or father entered the Stomatology and Orthodontics service with the baby. The patient was placed in the dental chair in order to record his/her behavior during the first 4 minutes. It was then evaluated according to the FLACC scale.

An alginate impression of the alveolar processes of the maxilla was then taken. During this procedure, the infant was re-recorded for 4 minutes to observe behavioral changes.

The orthodontist proceeded to place the pre-surgical orthopedic appliance in the mouth, in addition to incorporating a nasoalveolar molding. The baby's behavior was again videotaped for another 4 minutes. A week later, the patient had his appliance checked and the nasoalveolar molding was activated. Afterwards, the patient came every 15 days until the cheiloplasty was performed (*Figure 1*).

Six observers who assessed pain perception using the FLACC scale in three stages reviewed the recordings: 1) feeding technique; 2) impression taking; 3) placement of the pre-surgical orthopedic plate.

### Information gathering

The cases were assigned to the study groups in a sequential manner, as the patient arrived at the dentistry service.

An Excel database that included the results of each observer was developed. An algologist, a pediatrician, a psychologist, a plastic surgeon, an orthodontist, and the patient's mother participated. They rated before, during, and after each procedure with a scale

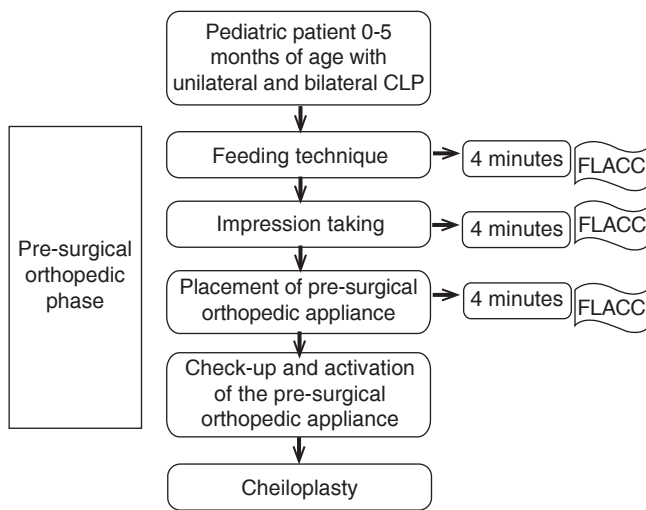
of 1 to 10. Subsequently, the result was reclassified considering the categories of relaxed, mild, moderate, and severe.

Before analyzing the video recordings, the evaluators were calibrated by the principal investigator. None of them were present during the preoperative orthopedic stage. The videos of 20 patients were collected and evaluated by the six observers

mentioned above for each of the patients with cleft lip and palate.

The FLACC scale scores were obtained in the three times mentioned above that represent the steps of preoperative orthopaedic stage: during the feeding technique, impression taking and placement of the preoperative orthopaedic appliance.

The recorded observation period was 4 minutes, the time needed to record the FLACC scale values.<sup>6</sup> The evaluators looked at each recording of the total of 60. The pain scores were recorded individually. On average, each of the observers analyzed six videos per day, and the principal investigator collected and ordered the scale assessment information for each observer (Figure 2).



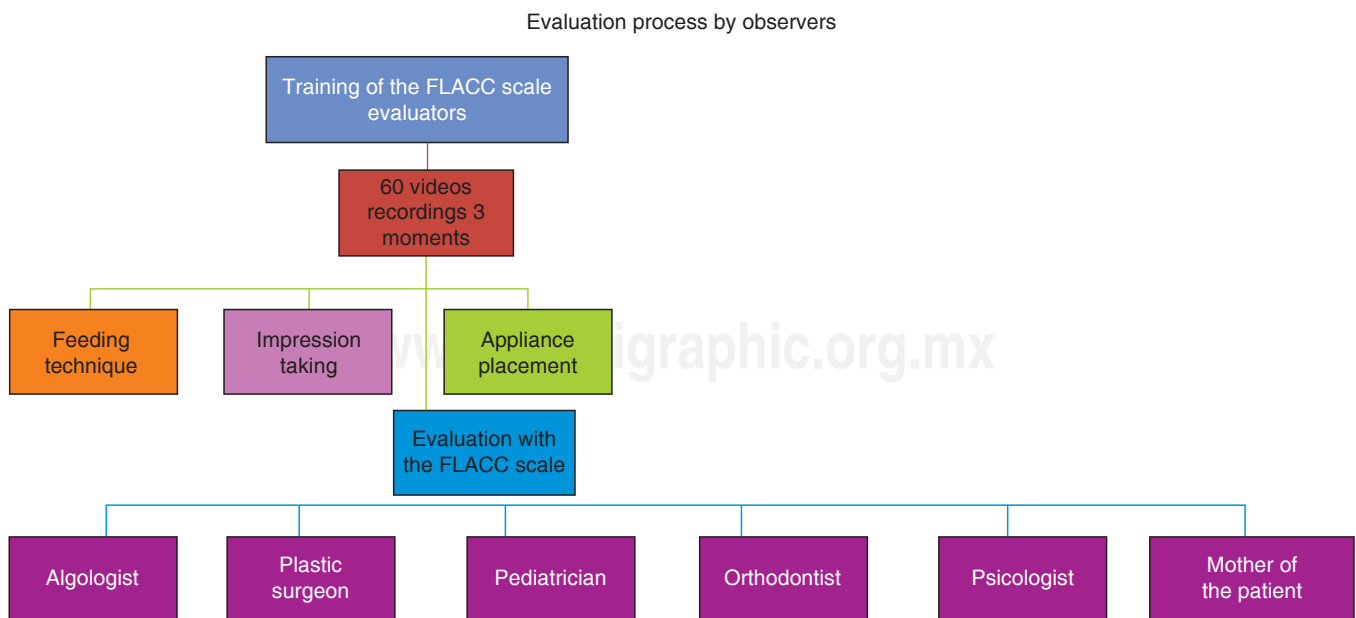
**Figure 1.** Flowchart during the FLACC evaluation. It shows the 4 stages of pre-surgical orthopaedic treatment up to the completion of the cheiloplasty. The FLACC evaluation was applied at all stages.

**Ethical considerations**

All the procedures of this study were carried out in accordance with the provisions of the Regulations of the General Health Law on Health Research and with the approval of the Research and Research Ethics Committee of the General Hospital «Dr. Manuel Gea González». Informed consent was obtained from the parents and/or guardians of each of the pediatric patients participating in the study.

**Statistical analysis**

We performed descriptive statistics of the sociodemographic variables with central tendency,



**Figure 2.** Flowchart of the FLACC scale evaluation during the different procedures.

dispersion, proportion and precision measurements at a 95% confidence interval. For the evaluation of pain using the FLACC scale, the following were used: the average, the t-Student test, the Shapiro normality test and the Wilcoxon non-parametric test. The reproducibility of the method was determined using 95% intraclass correlation coefficients at random and two-way analysis of variance (ANOVA) using the statistical package Stata 11.2 (USA).

## RESULTS

The present study showed female predominance in a ratio of 7 to 3 over male; the average age was  $5.75 \pm 4.58$  years; the type of fissure that predominated was unilateral in 60%.

### Impression of the patient's fissure

Regarding the parameters established by the FLACC scale, the results showed that the six observers obtained a median of 0. However, the percentile of 75% evaluated by the mother or the psychologist reached a value of 6, which showed a general agreement on the absence of pain during the feeding technique during which the patient is relaxed and comfortable according to the FLACC scale score.

The median of observers –66%, (n = 4)– determined moderate pain during impression taking and appliance placement while 33.3% of observers (n = 2), in this case surgeon and allergist, rated pain as severe. In the feeding technique stage, when pain is evaluated

according to the type of fissure, it was observed that, in bilateral cleft lip and palate, as well as in right and left unilateral cleft lip and palate, there is an agreement of 100% absence of pain.

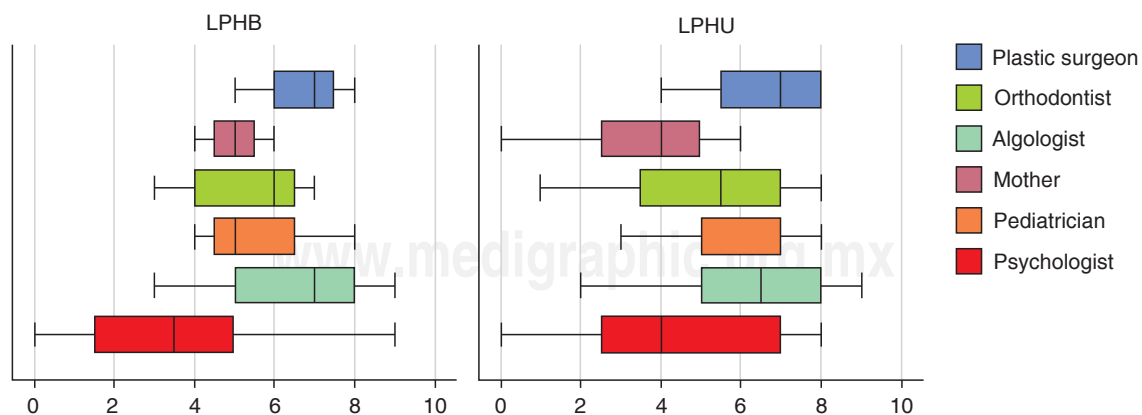
During the bilateral cleft lip and palate impression, 50% of the observers (n = 3) agreed that there was moderate pain; the algologist and the plastic surgeon –33.3%, (n = 2)– qualified it as severe pain. Only the psychologist –16.6%, (n = 1)– evaluated it as mild pain. In right or left unilateral cleft lip and palate, 66.6% of the observers (n = 4) agreed that there was moderate pain; the plastic surgeon and the pediatrician –33.3%, (n = 2)– evaluated it as severe pain (*Figure 3*).

### Appliance placement

During appliance placement on the lip and cleft palate bilaterally, 50% of the observers (n = 3) agreed that they had moderate pain and the other 50% (n = 3) rated it as severe pain. As in the impression taking, the psychologist and the mother showed lower pain perceptions than the other observers.

In right or left unilateral cleft lip and palate, 83.3% of the observers (n = 5) agreed that moderate pain was present and only the plastic surgeon -16.6%, (n = 1)-evaluated it as severe pain. The dispersion of the data is shown in *figure 4*.

The results obtained by the six observers were analyzed together through the Shapiro-Wilk normality test, which resulted in an abnormal distribution. Nonparametric tests were used to normalize the sample (*Table I*).



**Figure 3.** Evaluation of pain using the FLACC scale by type of fissure according to the observer during the impression taking in the patient's mouth: on the left side, the dispersion data are shown for patients with bilateral cleft lip who rate severe pain at 33%, moderate at 50% and mild at 16% during the impression taking in the patient's mouth. On the right side, data are presented for patients with unilateral right and/or left cleft lip, where severe pain was rated at 33.3% and mild pain at 63.6%.

The median reported by observers for impression taking and appliance placement was significantly different compared to the feeding technique (Table II).

Reliability of the FLACC scale in pain assessment in patients with cleft lip and palate

The ranges of the intraclass correlation coefficient showed results with a good degree of reliability (0.71-0.90)<sup>9</sup> during the feeding technique (0.784); Impression taking (0.835) and placement of the orthopaedic appliance (0.822) when using the FLACC scale by the six observers during the different procedures (Figure 5).

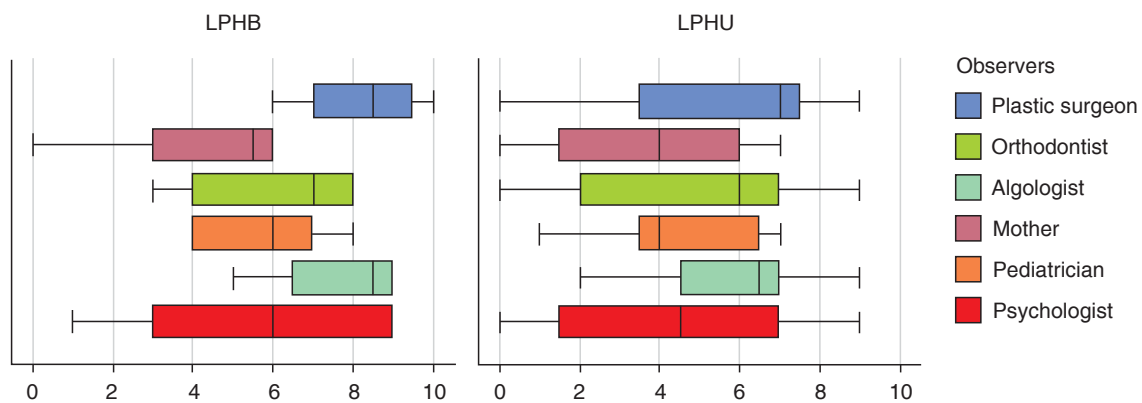
The reliability of the evaluation of the FLACC scale according to the values of the Intraclass Correlation

Coefficient (ICC) is shown, which was good (0.71-0.90) (Table III).

**DISCUSSION**

The purpose of this study was to assess pain perception using the FLACC scale in 20 pediatric patients aged 0 to 5 months with cleft lip and palate during the preoperative orthopaedic stage. The evaluation was performed by six independent and blinded observers using video recordings at three points during the preoperative orthopaedic phase: during feeding technique, impression taking and orthopaedic appliance placement.

To date no published evidence has been found of the reliability or validity of FLACC scale tests in



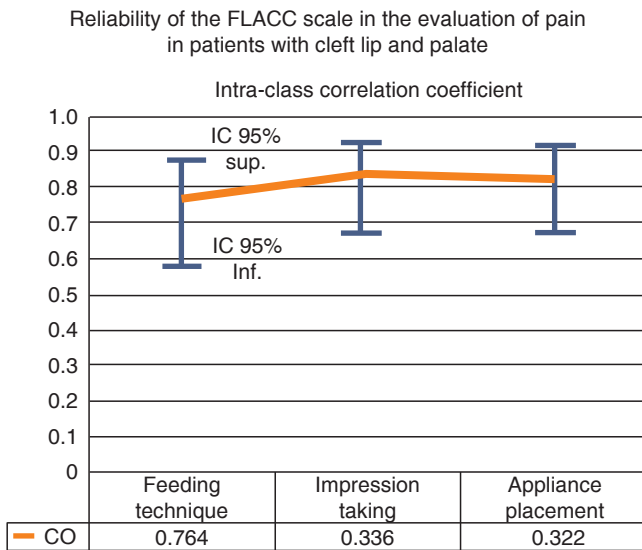
**Figure 4.** Pain assessment using the FLACC scale by type of fissure according to the observer during orthopaedic appliance placement. For the BCLP group 50% showed severe pain and 50% moderate pain, for the UCLP group 83.3% moderate pain and 16.3% severe pain.

**Table I.** Comparison of different procedures during the assessment.

	Obs	W	V	Z	Prob > z
Feeding technique	120	0.82519	16.821	6.324	0.00000
Impression taking	120	0.95918	3.928	3.065	0.00109
Orthopedic appliance placement	120	0.97112	2.779	2.29	0.01101

**Table II.** Wilcoxon range test for paired data between the feeding technique and impressions in which a statistically significant difference was reported \*\*p < 0.000000. The Wilcoxon range test for paired data between feeding technique and appliance placement also showed a statistically significant difference \*\*\*p < 0.0000.

	Mean	S.D.	Median	Min	Max	p value
Feeding technique	0.641667	1.424845	0	0	6	*
Impression taking	5.241667	2.355026	6	0	9	0.0000****
Orthopedic appliance placement	5.491667	2.625012	6	0	10	0.0000****



**Figure 5.** Evaluation of the reliability of the FLACC scale during the feeding technique, impression taking and orthopaedic appliances placement in the mouth.

**Table III.** Reliability of the FLACC scale assessment.

ICC value	Reliability
> 0.90	Very good
0.71-0.90	Good
0.51-0.70	Moderate
0.31-0.50	Mediocre
< 0.30	Bad or inexistent

patients with this type of condition, age and procedure performed. However, in other clinical instances it has been applied, such as the Pediatric Intensive Care Unit (PICU), the Post-Anaesthesia Care Unit (PACU), the Surgery and Trauma Unit and the Children's Oncology Unit.<sup>6</sup> Babl et al. also used it in the Emergency department,<sup>7</sup> while Johansson et al. used it in the Pediatric Intensive Care Unit (CIP).<sup>10</sup>

The results of this research showed that the presence of pain was evident in both impression taking and appliance placement. Pain may be evident in this type of procedure, which can be compared in some ways to a procedure that occurs when immunization is given to children.<sup>6</sup>

On the other hand, although the minimal clinically significant difference on the FLACC scale has not been well established,<sup>10</sup> the overall median obtained from the 120 assessments made by the six observers during impression-taking and orthopaedic appliance placement is significantly greater than that obtained during the feeding technique.

In this context, it is important to mention that the differences were more pronounced in patients with bilateral cleft lip and palate, a fact that is possibly explained by the greater manipulation and innervation of the affected area.<sup>11</sup> Although the reliability of the FLACC scale for assessing pain in pediatric patients aged 0 to 5 months with cleft lip and palate during the preoperative orthopaedic stage obtained in this study is consistent with other studies<sup>10-15</sup> that reported the reliability and validity of the scale for assessing pain in patients of different characteristics and ages, authors such as Manworren and Hynan<sup>6</sup> showed that this scale is an appropriate pain assessment tool for pre-verbal patients in surgery, trauma, cancer or other pathological processes.

A limitation of this study is that the sample size was small, although sufficiently adequate to meet the purpose of the research. Further study in these patients may provide a better understanding of the behavioral changes that best describe the pain during the procedure.

## CONCLUSIONS

Assuming the limitations of the present study, the following is concluded:

- It was demonstrated that there was pain during impression taking and appliance placement prior to lip cheiloplasty.
- The need to accompany the observer's report with behavioral measures was determined, as well as to consider alternative and/or pharmacological management, which should be multi and interdisciplinary during the treatment of pre-surgical orthopedics in children with cleft lip and palate.

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