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ABSTRACT

Background and Objectives. Congenital talipes equinovarus (CTEV) is one of the most common congenital defects of the musculoskeletal system, with an incidence ranging from 0.9 to 7 cases per 1000 live births. Deformity in CTEV does not resolve on its own, and if proper treatment is not provided, it will worsen as the patient reaches adulthood and cause side effects such as pain and long-term dysfunction. The classification of CTEV deformities is an important component of evaluation before therapy and is useful in

assessing progress in therapy. In this study, we retrospectively investigated the association between clinical outcomes and ininitial Dimeglio score before therapy, itial Pirani score before therapy, and patient age at presentation.

Materials and Methods. This study was an analytical study with a retrospective case-control design. The subjects in this study were patients with CTEV who were treated with the Ponseti method.

Results. Based on the classification by age implemented by the researchers, 12 participants (30.8%) were in the ≥ 12 months age group, followed by 10 (25.6%) in the 2–7 months age group, 9 (23.1%) in the < 2 months age group, and 8 (20.5%) in the 8–11 months age group. Further, statistically, initial Dimeglio score, initial Pirani score, and patient age at presentation had a moderate correlation with treatment outcome assessed by ACT score.

Conclusions. This study indicates an association between low initial Dimeglio score, low initial Pirani score, and early age at presentation with quality of life and high ACT score.

Keywords: Pirani score, Congenital talipes equinovarus, Dimeglio score

Abbreviations:

CTEV – Congenital talipes equinovarus,

ACT – Assessing Clubfoot Treatment,

SPSS – Statistical Package for the Social Sciences,

INTRODUCTION

Congenital talipes equinovarus (CTEV) is one of the most common congenital defects of the musculoskeletal system, with an incidence rate ranging from 0.9 to 7 cases per 1000 live births [1]. Deformity in CTEV does not resolve on its own, and if proper treatment is not provided, it will worsen as the patient reaches adulthood and cause side effects such as pain and long-term dysfunction [2].

The goals of CTEV treatment are to provide the patient with a leg that is functional, pain-free, and cosmetically acceptable [3,4]. Classifying CTEV deformity is an important component of the pre-therapy evaluation, as well as a useful tool for assessing the progress of therapy [2]. Currently, there are two universally accepted CTEV classification systems, the Pirani classification and the Dimeglio classification. Both have high rates of intra-observer and inter-observer reliability, good clinical relevance, and can be easily applied in clinical practice [5].

In previous decades, surgeons performed various surgical methods on patients with CTEV to restore the normal anatomical shape of the leg, but long-term results have shown that the operation causes many complications, including pain and stiffness [6]. Current methods of addressing CTEV have evolved from operative methods to conservative therapy [5].

The Ponseti method has been recognized worldwide as an effective method for treating CTEV; it is also a cost-effective technique that can be performed anywhere. In 2014, as many as 113 of the 193 United Nations member states had adopted this method [6]. The Ponseti method includes manipulation and serial placement of a circular cast until the desired initial correction is achieved, followed by Achilles tendon tenotomy and the use of an orthosis to maintain the correction and prevent the recurrence of deformities [6,7].

The wider application of the Ponseti method to include older patients with CTEV also raises the question of whether the patient's age at initiation of treatment is a factor affecting the outcome of therapy [8]. The extent of the relationship between age at presentation and success of correction with the Ponseti method is a research subject that has not achieved concrete conclusions [7].

Age at presentation is known to have a direct influence on the duration of treatment and the costs required for therapy. These factors are closely related to motivational and financial feasibility, especially in developing countries including Indonesia, which is one of the countries that has adopted the Ponseti method as a technique for treating patients with CTEV.

In this study, we retrospectively investigated the association between clinical outcomes and initial Pirani score before therapy, initial Dimeglio score before therapy, and patient age at presentation. If differences were found between these factors, the correlation between factors and the significance of correlations were also analyzed.

MATERIALS AND METHODS

Study Design

This study was an analytical study with a retrospective case-control design. The subjects in this study were patients with CTEV who were treated with the Ponseti method. This research was conducted at RSUP Dr. Wahidin Sudirohusodo hospital and the network in Makassar. Patient data from 2019 to 2023 were collected. Data collection was conducted from September 2020 to November 2024. The target population in this study were all patients with CTEV in Indonesia who met the inclusion and exclusion criteria. Ethical approval for this study was obtained from Research Ethics Committee, Faculty of Medicine, University of Hasanuddin, Makassar, Indonesia. Number: 322/UN4.6.4.5.31/PP36/2023.

The accessible population in this study were all patients with CTEV who met the inclusion and exclusion criteria treated in the Dr. Wahidin Sudirohusodo hospital and the network in Makassar. The inclusion criteria were patients previously diagnosed with CTEV who have received Ponseti method and foot orthosis treatment. The exclusion criteria were patients with non-idiopathic CTEV (artrogryphosis, syndromic, neuromuscular, etc.), patients with a history of prior operative treatment, patients who did not follow a regular schedule of serial circular casts (Ponseti method) and patients recurrent

Statistical analysis

Data analyzed by using SPSS Version 22.0 (IBM Corp., Armonk, NY). Pearson correlation and double linear regression tests were used to estimate the correlation between variables.

RESULTS

Based on the inclusion criteria, there were 78 patients with CTEV who were treated with the Ponseti method at Dr. Wahidin Sudirohusodo Makassar hospital and the network between 2010 and 2018. Of these, 14 patients were excluded due to having other congenital abnormalities, and 9 patients were excluded because they did not follow the schedule for wearing a circular cast regularly. Of the remaining 55 patients, 16 patients

could not be included in the study due to incomplete medical record data; the remaining 39 patients were included in the study.

Patient Characteristics

Of the 39 patients (65 feet) studied, 26 patients had bilateral CTEV (66.7%) and 13 patients had unilateral CTEV (33.3%). In the unilateral cases, 9 had right foot involvement (23.1%) and 4 had left foot involvement (10.26%). Males comprised 25 (64.1%) and females comprised 14 (35.9%) of the 39 patients (Table 1).

Age at presentation

The age range of patients when initiating treatment (age at presentation) was 2.5 months (youngest) to 36 months (oldest), with a mean age of 8.7 ± 8.4 months. The patients were separated into the following age group categories (Table 1):

- < 2 months old age group: 9 patients (23.1%), 15 feet (23%)
- 2–7 months old age group: 10 patients (25.6%), 18 feet (27.7%)
- 8–11 months old age group: 8 patients (20.5%), 11 feet (17%)
- ≥ 12 months old age group: 12 (30.8%), 21 feet (32.3%)

Initial Pirani score

Pirani scores at the start of treatment (initial Pirani score) for the 65 involved feet ranged from 2 to 5.5, with a mean Pirani score of 4.02 ± 0.82. In the 25 bilateral cases, there were 9 cases in which Pirani scores differed between left and right feet (Figure 1).

Initial Dimeglio score

Dimeglio scores at the start of treatment (initial Dimeglio score) ranged from 9 to 18, with an average Dimeglio score of 13.16 ± 2.6. In the 25 bilateral cases, there were 4 cases with different left and right foot Dimeglio scores (Figure 2).

Assessing* Clubfoot *Treatment (ACT) Score

ACT score ranged from 4 to 12, with a mean of 7.46 ± 1.80. ACT scores can be interpreted based on the following categories: minimal improvement (ACT score 0–7), moderate improvement (ACT score 8–10), and good improvement (ACT score 11–12) (Figure 3).

Correlation test

After applying the Pearson correlation test for each variable separately, the following results were obtained (Table 2):

- Patient age at presentation had a moderate correlation with treatment outcome assessed by ACT score (r = -0.697) (Figure 4).
- Initial Pirani score had a moderate significant correlation with clinical outcomes assessed by ACT score (r = -0.665; P = 0.000) (Figure 5).
- Initial Dimeglio score had a moderate significant correlation with clinical outcomes assessed by ACT score (r = -0.574; P = 0.000) (Figure 6).

DISCUSSION

The study included participants ranging in age from 0.3 months to 36 months, with a mean age of 8.7 months. The participants were classified into different age groups, with the majority falling12-month age group. The researchers used this classification for descriptive purposes but did not use it in the analysis of its correlation to clinical outcomes based on ACT score. Instead, they performed correlation testing by comparing each patient according to age and ACT score, and statistical testing and analysis were carried out on an individual patient basis to avoid bias between patients of different ages.

High-quality corrective treatment is a key requirement to reduce defects and improve function of the lower extremities. The last few decades have seen an increase in the use of the Ponseti method to correct CTEV [6]. This method involves the simultaneous correction of the three components of CTEV through serial manipulation and fixation. Ponseti initially suggested that the technique can be started in the first week of life, is effective when it is started before 9 months of age, and can still provide satisfactory results when started before 24 months of age [9]. The effectiveness of the Ponseti method for patients under 24 months of age has been demonstrated in multiple studies [1].

Many groups have reported different success rates with the Ponseti technique due to cultural, economic, and health care differences. It is believed that manipulation should start as soon as possible. However, several investigators have shown that even patients with CTEV who begin treatment late can have success with the Ponseti method. The relationship between immediate treatment at an early age and successful correction and recurrence rate is not certain [9]. We therefore first investigated whether age at presentation influenced correction and recurrence rates.

The Pearson correlation test in this study showed a significant relationship between the initial percentage of patients receiving therapy and the clinical outcome ($p < 0.005$), with a Pearson correlation of -0.697, indicating that the younger the age when treatment was initiated, the higher the ACT score and the better the patient's clinical outcome.

The results of this study are in line with the research conducted by Liu et al. (2018), which had similar results; patients were divided into several age groups (I: < 28 days, II: 28 days–3 months, III: 3–6 months), and patients who started therapy earlier achieved relatively better results compared to older children [10].

The recommendation that CTEV should be treated immediately after birth has been widely accepted. Ponseti also suggested that initial treatment should be started in the first few weeks of life, taking advantage of the improved viscoelastic properties of the newborn's connective tissue. Until recently, however, the upper age limit for Ponseti management was not clear. Some authors have reported that CTEV occurring at an older age can also be successfully managed with the Ponseti method [10].

Different results were shown by Alves et al., who compared the outcomes of patients undergoing Ponseti method therapy based on their age at therapy initiation. The study compared two age groups (I: < 6 months, II: > 6 months), and there was no significant difference between the two groups in clinical outcomes. The difference in results may be due to differences in methodology, as this study used a retrospective design [11].

Based on previous studies, the optimal time to recommend therapy with the Ponseti method is unclear [12]. The findings of the current study suggest that carrying out the correction as soon as possible achieves better results with minimal complications.

Classification of severity of CTEV deformity is important for evaluating, monitoring, and predicting the effects of therapy; a standardized CTEV classification system is thus required, has clinical relevance, and should be easy to use [1]. Today, the Dimeglio and Pirani scoring systems have been adopted as the most widely used and universally accepted classification systems. Both have high rates of inter-observer and intra-observer reliability, good clinical relevance, and are easy to use in clinical practice [7].

Although both the Pirani and Dimeglio scoring systems have excellent inter-observer and intra-observer coefficients, their clinical uses are different and can be complementary. The Dimeglio score evaluates reducibility, while the Pirani score evaluates the morphological aspects of the feet. The Dimeglio classification system focuses on the corrections obtained after performing a light reduction force on foot deformities, while the Pirani scoring system assesses the physical appearance of the feet [5].

The results of the Pearson correlation test in this study indicate a significant relationship between final outcomes and both the Pirani score (correlation coefficient value, r = -0.655; P = 0.004) and the Dimeglio score (r = -0.594; P = 0.000), where both were negatively and significantly correlated. Thus, the higher the initial Pirani or Dimeglio score, the worse the ACT score and the worse the clinical outcome. Both results showed a strong correlation between initial score and clinical outcome.

Several previous studies have attempted to correlate the Dimeglio and Pirani scores to outcomes of the Ponseti method. Another study showed that there was a significant relationship between the incidence of relapse after the Ponseti method and the initial Pirani score. The incidence of relapse was greater with higher Pirani scores [12]. Fan et al. had similar results in their study comparing Pirani and Dimeglio scores at the start of therapy to the occurrence of relapses. Clinically, there was a significant difference, in which patients with higher initial Dimeglio and Pirani scores were more likely to relapse while those with lower Pirani and Dimeglio scores had no relapses. However, this study was not statistically significant, possibly due to a small sample size [13].

Although the Dimeglio and Pirani scores are effective for clinically assessing CTEV, these systems still have a certain element of subjectivity. Differences in clinical evaluation by different observers are frequent and are described in some studies. In addition, published studies have not found a statistically significant relationship between initial clinical examination and treatment outcomes [14].

Various parameters influence the clinical outcomes of CTEV therapy, and thus far there has not been a clear consensus on the outcome of therapy. Research conducted by Dyer et al. found a significant correlation between initial Dimeglio and Pirani scores and the need for an Achilles tendon tenotomy procedure [15].

The study conducted by Gao et al. showed that Pirani and Dimeglio scores had limitations in determining the outcome and prognosis of therapy, particularly in the early phase of therapy with the Ponseti method [16]. However, several studies have shown that initial Dimeglio and Pirani scores are strongly associated with the need for Achilles tenotomy and the number of casts used during therapy [14,17].

To the best of the researchers' knowledge, this study is the first to simultaneously analyze the relationship between the three variables of age at presentation, Pirani score, and Dimeglio score and clinical outcomes measured by ACT score after correction of CTEV using the Ponseti method.

This study was retrospective, with data collected from the patients' medical records, not based on direct clinical observations, so it was strongly influenced by the level of accuracy and reliability of the available data. Our study took samples from several hospitals, so the Pirani and Dimeglio scores were not determined by the same clinician, though this is not a significant limitation because both the Pirani score and the Dimeglio score have high inter-observer reliability rates.

Although there are specific treatment guidelines according to the Ponseti method, the decision to discontinue the use of circular casts and initiate the use of foot abduction orthoses is fully determined by the doctor in charge of the patient. Because more than one doctor treated the patients included in this study, differences in judgment and decision-making could potentially be a factor causing bias in this study. There is a possibility that clinical procedures were an intermediate factor that directly influenced the outcome of the Ponseti method, as differences in perceptions of the treating doctors may affect the casting procedure and have an impact on the patient's outcome.

CONCLUSION

This study contributes to the data on the evaluation of clubfoot treatment in low-resource settings. The ACT score, which includes a physical observation of the foot and

parent-reported outcome measure, was used to determine clinical outcomes. Patients with early age at presentation and low initial Pirani and Dimeglio scores achieved higher ACT scores, indicating a good clinical outcome. This study indicates an association between early age at presentation, low initial Pirani score, and low initial Dimeglio score with high ACT score and quality of life.

CONFLICT OF INTEREST: None declared

AUTHOR'S CONTRIBUTIONS: MT, JL, NM, and GVS (idea, planning, availability, materials, gathering and processing of data, interpretation and analysis, literature search, and writing of manuscripts). JL, NM, and GVS (idea, design, supervision, evaluation and interpretation, and literature search). MIK and CR (idea, design, supervision, evaluation and interpretation, and literature search). MF (Concept, Design, Analysis and Interpretation, Critical Review)

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INFORMED CONSENT: Informed consent was obtained from all parents/guardians in the study.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE: The study was approved by the Research Ethics Committee, Faculty of Medicine, University of Hasanuddin, Makassar, Indonesia. Number: 322/UN4.6.4.5.31/PP36/2023.

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TABLES

Table 1. Patient characteristics by age group, sex, and foot involvement

Age (months)	Number of patients n (%)	Sex		Bilateral n (%)	Unilateral		Number of feet n (%)
		Male n (%)	Female n (%)		Right n (%)	Left n (%)	
< 2	9 (23.1)	7 (17.9)	2 (5.1)	6 (15.3)	3 (7.6)	0 (0)	15 (23)
2–7	10 (25.6)	8 (20.5)	2 (5.1)	8 (20.5)	1 (2.5)	1 (2.5)	18 (27.7)
8–11	8 (20.5)	3 (7.6)	5 (12.8)	3 (1)	3 (7.6)	2 (5.1)	11 (17)
≥ 12	12 (30.8)	7 (17.9)	5 (12.8)	9 (23)	2 (5.1)	1 (2.5)	21 (32.3)

Table 2. Correlation of Dimeglio score, Pirani score, and age at presentation with ACT score

Variable	ACT score		
	Correlation		
	N	Coefficient (r)	P-value
Age (months)	65	-0.697	0.004a
Pirani score	65	-0.665	0.005a
Dimeglio score	65	-0.574	0.007b

FIGURES

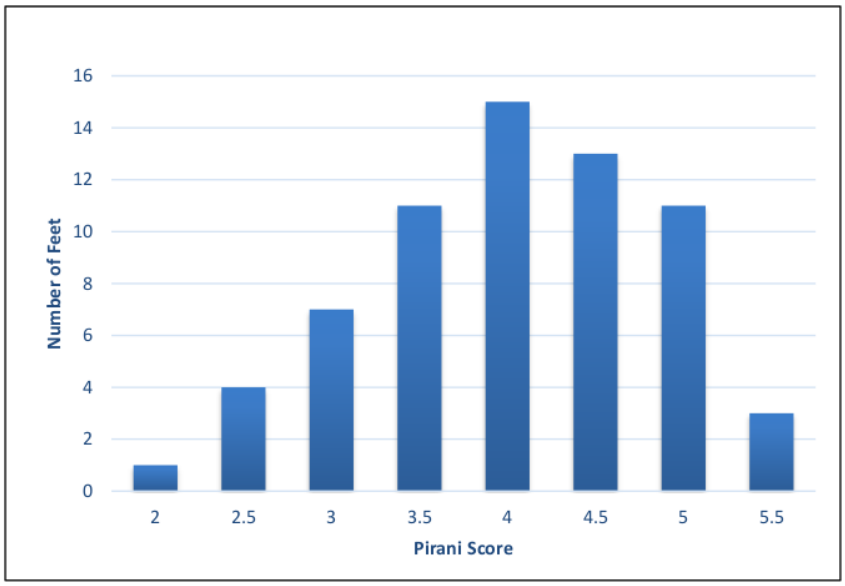

Figure 1. Distribution of the number of feet based on Pirani score

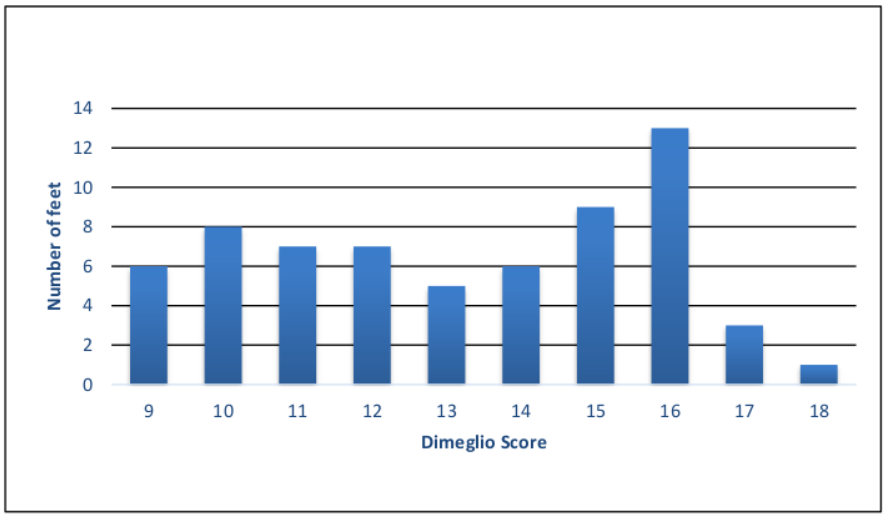

Figure 2. Distribution of the number of feet based on Dimeglio score

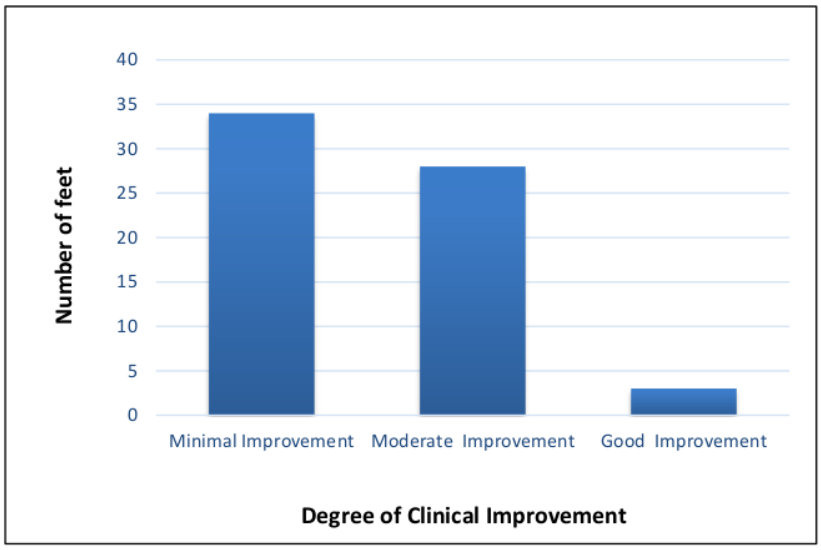

Figure 3. Distribution of the number of feet based on the degree of clinical improvement according to ACT Score

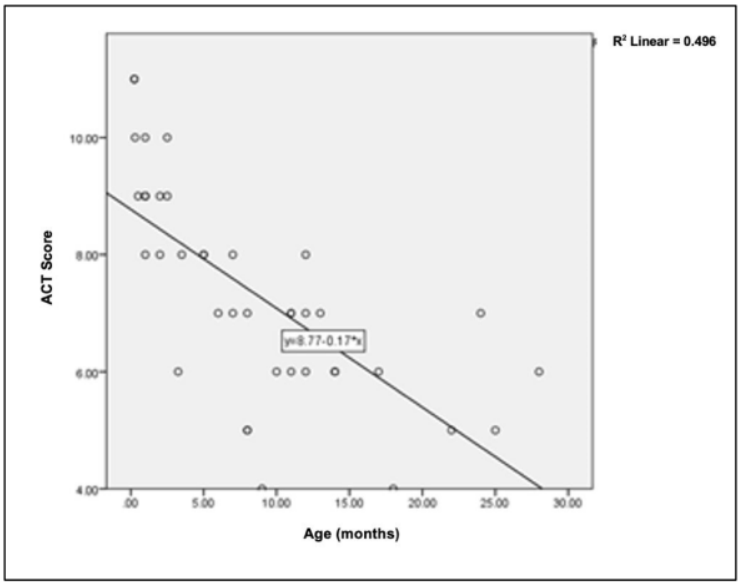

Figure 4. Relationship between age at presentation and ACT score

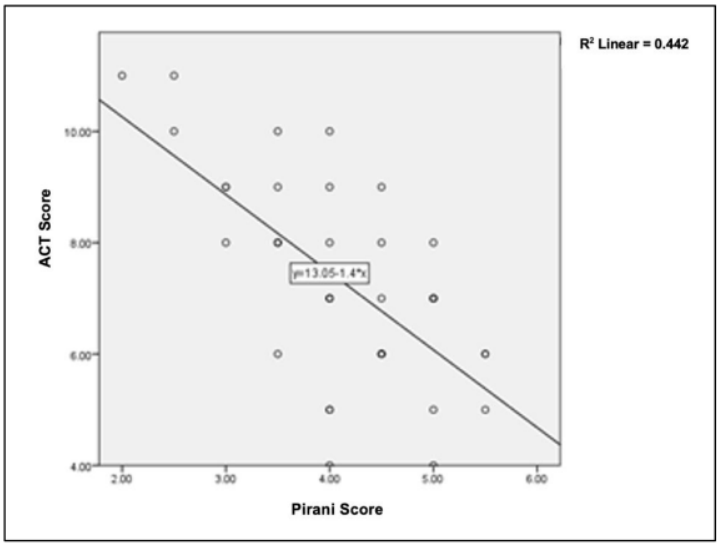

Figure 5. Relationship between initial Pirani score and ACT score

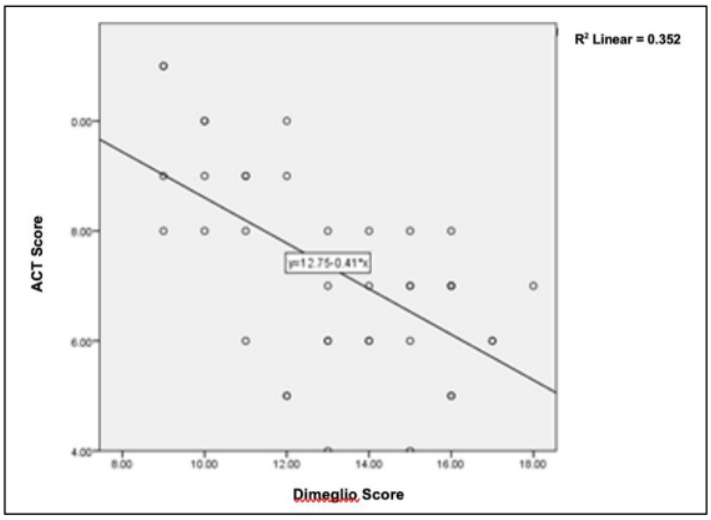

Figure 6. Relationship between initial Dimeglio score and ACT score