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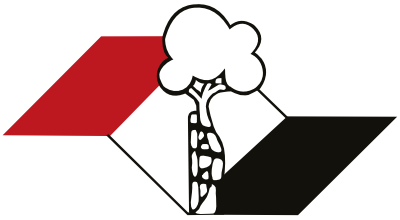

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SUMMARY

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Original Article

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ANALYSIS OF INTRINSIC FOOT MUSCLE STRENGTH IN DIABETIC PATIENTS WITH AND WITHOUT DIABETIC PERIPHERAL NEUROPATHY COMPARED WITH HEALTHY CONTROLS

ANÁLISE DA FORÇA DA MUSCULATURA INTRÍNSECA DO PÉ EM PACIENTES DIABÉTICOS COM E SEM NEUROPATIA PERIFÉRICA DIABÉTICA, COMPARADOS A CONTROLES SAUDÁVEIS

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ABSTRACT

Objective: to evaluate the strength of the intrinsic foot muscles in diabetic patients with and without diabetic peripheral neuropathy, compared to healthy controls. Methods: a prospective experimental study was conducted with 63 patients divided into three groups: diabetics with peripheral neuropathy (20), diabetics without neuropathy (23), and healthy controls (20). Muscle strength was measured using a digital handheld dynamometer, assessing plantar flexion of the hallux and toes in two positions (neutral and maximum plantar flexion). Statistical analyses were performed using R software, with α=0.05 and β=0.2. Results: the groups had a mean age of 66 years. Variables with statistically significant differences ($p < 0.05$) included Right Hallux – Neutral Position, All Toes (Right) – Neutral Position, All Toes (Right) – Plantar Flexion, and Left Toes – Plantar Flexion. Diabetic patients with neuropathy showed lower medians and interquartile ranges in these variables, indicating reduced muscle strength compared to controls. Conclusions: this study explored the strength of the intrinsic foot muscles in diabetic patients, using dynamometry as an assessment tool. Although no conclusive evidence was found, it is suggested that dynamometry may be useful in the early detection of muscle weakness in diabetic neuropathy. Expanding the study with a larger sample and more data is necessary to validate and refine the preliminary conclusions, contributing to the monitoring of muscle deterioration and the effectiveness of treatments. ***Level of Evidence II; Prospective[d] comparative study[e].***

Keywords: Diabetes Mellitus; Manual Dynamometry; Muscle Strength; Diabetic Neuropathies; Diabetic Foot; Disease Prevention.

RESUMO

Objetivo: avaliar a força da musculatura intrínseca dos pés em pacientes diabéticos com e sem neuropatia periférica diabética, comparados a controles saudáveis. Métodos: estudo experimental prospectivo realizado com 63 pacientes divididos em três grupos: diabéticos com neuropatia periférica (20), diabéticos sem neuropatia (23), e controles saudáveis (20). A força muscular foi medida utilizando dinamômetro manual digital, avaliando a flexão plantar do hálux e dos artelhos em duas posições (neutra e flexão plantar máxima). Análises estatísticas foram realizadas com software R, utilizando α=0,05 e β=0,2. Resultados: os grupos apresentaram média de idade de 66 anos. Variáveis com diferenças estatisticamente significativas ($p<0,05$) incluíram 'Hálux D - neutro', 'Todos D - neutro', 'Todos D - flexão plantar', e 'Artelhos E - flexão plantar'. Pacientes com neuropatia diabética apresentaram medianas e intervalos interquartis inferiores nessas variáveis, indicando uma redução na força muscular comparada aos controles. Conclusões: este estudo explorou-se a força da musculatura intrínseca do pé em pacientes diabéticos, utilizando-se a dinamometria como ferramenta. Apesar de não terem sido encontradas evidências conclusivas, sugere-se que a dinamometria pode ser útil na detecção precoce de fraquezas musculares em neuropatia diabética. A ampliação do estudo, com uma amostra maior e mais dados, é necessária para validar e refinar as conclusões preliminares, contribuindo para o monitoramento da deterioração muscular e a eficácia de tratamentos. ***Nível de Evidência II; Estudo[e] prospectivo comparativo[d].***

Descritores: *Diabetes Mellitus; Dinamometria Manual; Força Muscular; Neuropatias Diabéticas; Pé Diabético; Profilaxia.*

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INTRODUCTION

An epidemic of diabetes *mellitus* (DM) is ongoing. In 1985, it was estimated that there were 30 million adults with DM in the world, this number jumped to 135 million in 1995, reaching 173 million in 2002.[1] In 2019, the International Diabetes Federation (IDF)[2] estimated that 9.3% of the global population, aged 20 to 79, had been diagnosed with the disease, this percentage will reach 10.4% by 2040, i.e. 642 million diabetic patients.[3]

About two-thirds of these individuals with DM live in developing countries where the epidemic is more intense, with an increasing proportion of people affected in younger age groups coexisting with the problem that infectious diseases still represent.[3]

The latest data from IDF DIABETES ATLAS[2] show that Brazil ranks 5th among the countries with the highest prevalence of diabetes: 16.8 million people with diabetes. It is estimated that the prevalence of the disease in the country is 7.6%, with half of these people not aware of having the problem.[4] It is estimated that this prevalence will increase by almost 50% over the next 25 years.[5] These figures are worrying, especially because the disease raises mortality by more than 50% compared to the general population of Brazil.

According to the World Health Organization, diabetic foot is characterized by infection, ulceration and/or destruction of deep tissues, associated with neurological abnormalities and varying degrees of peripheral vascular disease in the lower limbs.[6] Peripheral neuropathy, defined as the presence of symptoms and/or signs of peripheral nerve dysfunction in people with diabetes after excluding other causes, affects almost 50% of patients after 10 years of disease, being the most common complication of diabetes *mellitus*.[3] Peripheral arterial disease, along with neuropathy, are responsible for the "Diabetic Syndrome", which includes ulcer, Charcot arthropathy, infection and can lead to amputation[7].

Peripheral vascular disease is the main factor related to the evolution of a diabetic foot ulcer, and it should be diagnosed through a clinical examination of the feet, evaluating the color, skin temperature, pulse palpation and measurement of ankle pressure.[7]

About 40% to 60% of non-traumatic amputations of lower limbs occur in diabetic patients, with 85% of these preceded by foot ulcers.[7] Skin and soft tissue infections represent a significant impact for both diabetic patients and healthcare systems. Diabetic patients often face longer hospitalization times, high rates of clinical failure, increased readmission rates, and higher mortality rates compared to nondiabetic patients.[8]

When analyzing the investments in the area, it is estimated that around US$760 billion are spent globally annually on DM and its complications, and there are projections that by 2030 the value will exceed US$825 billion.[9]

This study aims to investigate possible changes in the strength of the intrinsic musculature of the foot in diabetic patients using dynamometry. The aim is to test the hypothesis that the loss of strength can serve as an early marker for complications associated with diabetes, such as deformities, ulcers and amputations. This approach aims to offer a strategy to monitor the deterioration of muscle strength and evaluate the effectiveness of the treatments applied. The relationship between muscle weakness monitoring and treatment effectiveness is based on the assumption that early detection of muscle weakness can reflect the progression of diabetic complications and thus allow for more effective adjustments in treatment to prevent adverse outcomes.

MATERIALS AND METHODS

This project is part of the research "Analysis of the clinical-epidemiological profile of patients undergoing outpatient follow-up in the Department of Orthopedic and Traumatology of University Hospital Prof. Edgard Santos-UFBA", approved with CAAE number 13790619.6.0000.0049.

This prospective experimental study was conducted at the Ambulatory Hospital Complex Professor Edgard Santos, with the aim of comparing diabetic patients, with and without diabetic peripheral neuropathy, with non-diabetic patients. The study exclusively included adult women, aged between 60 and 70 years, recruited from the services of Endocrinology and Orthopedics.

• Inclusion Criteria: The choice to include women between the ages of 60 and 70 was motivated by two main factors:

1. Homogeneity of the age group: the age range from 60 to 70 years was selected to ensure the homogeneity of the group in terms of age, minimizing the impact of aging-related variables on the results.
2. Female focus: only women were chosen to be included to reduce the variability associated with gender differences in the prevalence and clinical presentation of diabetic peripheral neuropathy. Studies indicate that women in this age group are more susceptible to diabetes-related complications, justifying the focus of this study.

• Non-Definition of Type of Diabetes (DM I or II): The type of diabetes (I or II) was not specified among the participants, as the central objective of the study is to investigate the presence and absence of diabetic peripheral neuropathy as the main variable. Regardless of the type of diabetes, peripheral neuropathy can develop, and the study's interest lies in comparing outcomes related to this condition.

• Duration of Disease Progression and Degree of Control: Although the duration of diabetes progression and the degree of glycemic control are important factors in the progression of diabetic complications, including peripheral neuropathy, these aspects were not directly controlled in this study. The inclusion of patients who have reached the age of 60-70 years without taking into account these variables is intended to reflect a broader and generalizable clinical scenario.

After clinical evaluation and application of inclusion and exclusion criteria (exclusion of patients with foot deformities, Charcot foot, prior history of amputation or diabetic foot ulcer), 63 patients were selected and divided into three groups:

1. 20 diabetic patients with diabetic peripheral neuropathy;
2. 23 diabetic patients without diabetic peripheral neuropathy;
3. 20 non-diabetic patients, no foot and ankle pathologies.

The selection of the 63 individuals was made from a larger group of patients followed in the services of Endocrinology and Orthopedics, ensuring the representativity of the groups and the validity of the results.

In this sense, the clinical evaluation in the outpatient clinic, in which the Term of Free and Informed Consent (TCLE) was presented for eligible patients. Therefore, after reading the TCLE for the participants and making the clarifications, the signing of the same, as well as the completion of a questionnaire with basic cadastral data was carried out.

The physical examination included a clinical evaluation and tactile sensitivity tests in diabetic patients using the Semmes-Weinstein 10g monofilament.[10] Six points were evaluated in the plantar region of the right and left feet: hallux, 3rd and 5th toes (plantar region of the distal phalanges) and 1st, 3rd and 5th heads of the metatarsus. The patient's inability to feel the 10g filament at one or more of these points indicated loss of protective sensitivity (LPS), which classified the patient in the group of diabetics with peripheral neuropathy. Diabetic patients who did not experience loss of sensitivity were included in the group of diabetics without peripheral neuropathy. To ensure the accuracy of the results and the reliability of the comparison, similar tests were performed in the group without diabetes and without foot and ankle pathologies.

• Control and description of deformities: During the physical examination, a check was performed to identify and describe any

deformities in the feet, both in diabetic patients and in healthy adults. Deformities were recorded, such as clutched feet or other anatomical alterations, which could interfere with monofilament sensitivity. This documentation helped ensure that the detected loss of sensitivity was not attributable to pre-existing structural deformities.

• Muscle Strength and Dynamometer Test: The contraction force of the intrinsic musculature of the feet was measured using a digital manual dynamometer Bticx® and an adapted wooden platform (Figure 1). The wooden platform provided a stable base during the measurement and ensured the consistency of the tests. The footer on the platform, which mimics the movement axes of the lateral fingers and the hallux, has been designed to facilitate the correct positioning of the feet and ensure that the force applied is measured accurately and repeatably. The dynamometer was positioned to evaluate the plantar flexion of the metatarsophalangeal joints of the hallux and the 2nd, 3rd, 4th and 5th arthritis, as well as the total strength of all arthritis of the right and left feet.

The method of measuring intrinsic muscular strength of the foot using manual dynamometer was described and validated by Xu et al.[11] and Błażkiewicz et al.[12] Additionally, Ribot-Ciscar et al.[13] discuss the application of similar techniques to diabetic patients. To ensure that only the strength of the intrinsic foot muscle is measured, the test was designed to minimize the influence of the leg muscle, with the platform and the dynamometer adjusted to isolate the strength of the foot muscles.

The dynamometer used in our study is the Bticx® handheld digital dynamometer, is certified for use in strength tests, has precise caliber cable and is used to measure grip strength in a variety of applications.

The recording of the contraction force was performed in two moments: with the ankle in neutral position and in the position of maximum plantar flexion (Fig. 2 and 3). The illustration subtitles were elaborated to provide a clear and reproducible description of the methods and equipment used, ensuring that other researchers can accurately replicate the procedure.

After data collection, the analysis was performed, in which the variables between the groups were compared; an $\alpha = 0.05$ and $\beta = 0.2$ were used. The statistical analysis methodology was performed using the *R programming language.* A definition of normality was made through graphical analysis and Shapiro-Wilk test. For the descriptive analysis, the quantitative variables with normal distribution were represented by their mean and standard deviations, those of non-normal distribution per quartile ($Q_{1/4}$, median, $Q_{3/4}$).

Source: Author.

Figure 1. Digital manual dynamometer and an adapted wooden platform.

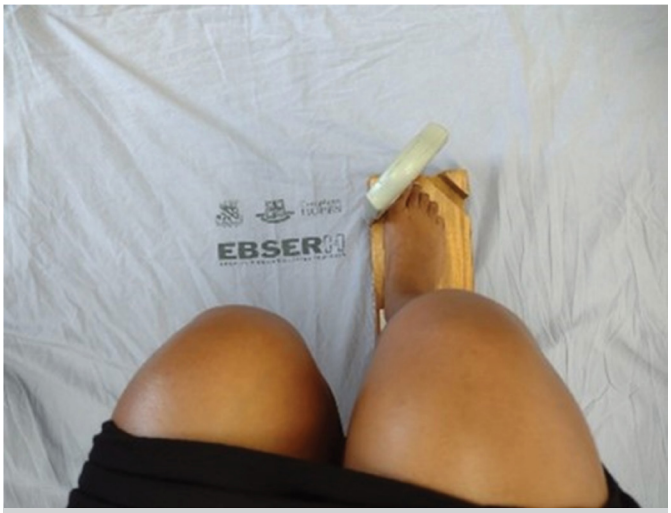

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Figure 2. Strength measurement, with ankles in neutral position.

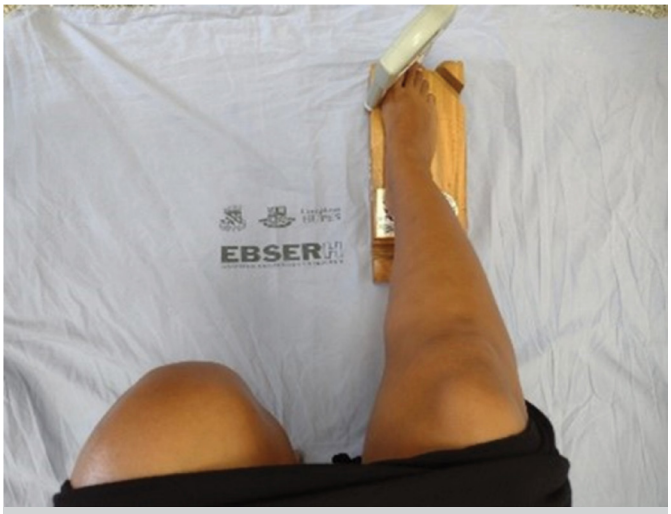

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Figure 3. Strength measurement, with ankles in maximum plantar flexion position.

Comparisons between groups were made through the Kruskal-Wallis test for non-normal distribution variables and ANOVA test for normal distribution variables.

RESULTS

After data collection, tabulation of the data was carried out for further analysis. Therefore, the definition of median and interquartile ranges can be observed in Figure 4.

The data obtained can be observed in Table 1, which contains the results obtained after clinical evaluation and physical examination of patients. The age variable is expressed in years; the rest correspond to the force in Newtons. The interpretation of the table takes into account the distribution for each group; normal variables present average and standard deviation (XX ± YY), non-normal numeric variables present median and lower and upper quartile (XX (Inf – Sup).

The results of the statistical tests are given by the column 'Value of p'. Values below 0.05 show statistically significant difference between groups, i.e., the groups present different results among themselves. Values greater than 0.05 tells us that the groups are statistically similar.

In addition, graphs of the type *Boxplot* were elaborated – which has its characteristics presented in Figure 5 for data analysis. Figures 6 to 9 were constructed in the *Boxplot* format, and used the variables of statistically significant value ($p<0.05$) – with the exception of the variable 'Hallux R - neutral' and 'Toes L – plantar flexion'.

DISCUSSION

Studies indicate that diabetes *mellitus* may result in loss of muscle strength, especially in the lower limbs, leading to a decline in physical function and reduction in the quality of life of patients.[14] This loss of strength is not limited to the lower limbs, but may also affect the strength of the hand, suggesting a diffuse loss of strength among diabetic patients.[15]

Intrinsic Foot Muscle Anatomy

The human foot is an adaptable structure, designed to adjust to surface and load variations, keeping efficient force transmission between the lower limb and the soil. The complex interaction of movements in the small joints of the foot allows the adaptation of the longitudinal arc during the walk, absorbing and reusing forces such as elastic energy.[16] The plantar aponeurosis and the windlass mechanism are essential for foot stiffness during walking and for efficient propulsion.[16] The intrinsic musculature of the foot, organized in four layers, plays a crucial role in the maintenance of the structural and functional integrity of the foot.[16,17]

Strength Analysis of Intrinsic Foot Musculature

Deficits in the strength of the intrinsic muscle of the foot are associated with various pathologies and compromise the balance of the lower limbs. Weakness or altered activation of this muscle can contribute to conditions such as cavus foot, plantar fasciitis, claw toe deformities, hammertoe, retraction of the medial longitudinal arch, hallux valgus, and post-medial ankle pain.[18] In diabetic patients, these deformities, combined with sensory neuropathy, increase the risk of plantar ulcers due to pressure in the affected areas.[19]

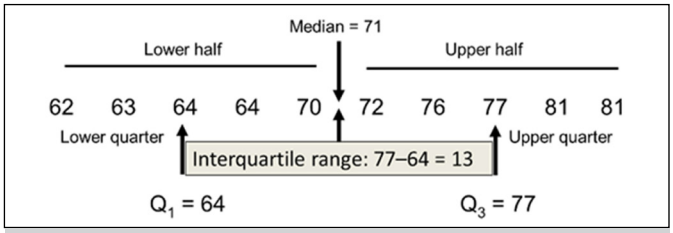

Figure 4. Median and Interquartile ranges.

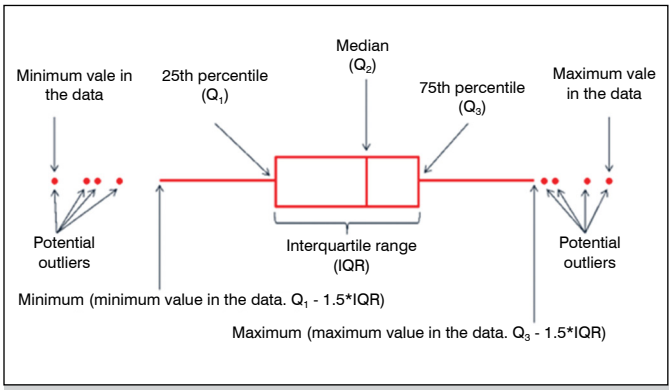

Figure 5. Anatomy of a Boxplot chart.

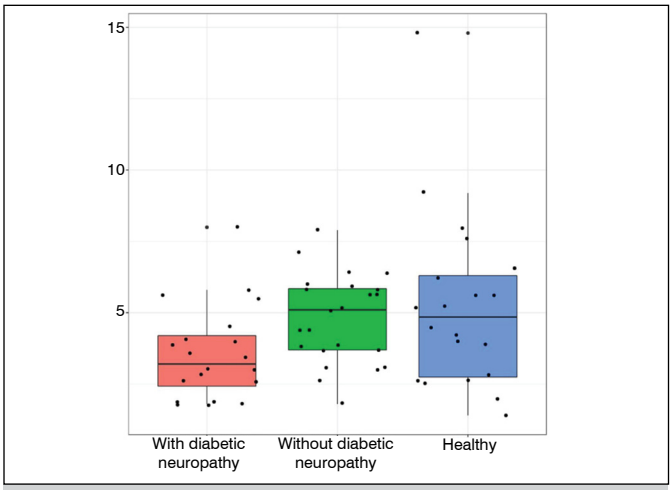

Figure 6. Results obtained for the Right Hallux (Neutral Position).

The evaluation of the intrinsic muscular strength of the foot is important, even in the presence of diabetic neuropathy, which initially manifests itself as a sensitive condition. Measurement of muscle strength can identify early changes in muscle function that are not detectable only by sensitive symptoms. The loss of muscle strength can precede and aggravate complications, offering an opportunity for preventive interventions before the deformities and ulcers develop. Therefore, intrinsic muscle strength can provide valuable insights about the progression of neuropathy and the risk of related complications.

Table 1. Data obtained after clinical evaluation and physical examination of patients.

Variable/Group	Total (n = 63)	With Diabetic Neuropathy (n = 20)	Non-Neuropathic Diabetic Patients (n = 23)	Healthy Controls (n = 20)	P value
AGE	66.0(62.5-68.0)	66.0(64.8-69.0)	66.0(65.0-68.0)	63.0(61.0-67.2)	0.226
Hallux (Right) – Neutral Position	4.5±2.3	3.6 ± 1.7	4.8 ± 1.6	5.2 ± 3.1	0.054
Right Toes – Neutral Position	4.0±1.5	3.6 ± 1.2	4.6 ± 1.7	3.8 ± 1.5	0.066
All Toes (Right) – Neutral Position	5.5(3.5-6.8)	3.7(3.2-5.4)	6.1(3.8-6.9)	5.9(4.8-6.9)	0.024
Right Hallux – Plantar Flexion	2.9(2.0-3.8)	2.8(1.8-3.5)	3.0(2.2-3.9)	3.0(2.2-4.1)	0.468
Right Toes – Plantar Flexion	2.9(2.0-3.8)	2.4(1.8-3.1)	2.9(2.1-4.0)	3.3(2.2-4.2)	0.072
All Toes (Right) – Plantar Flexion	3.8±1.8	3.0 ± 0.9	4.0 ± 1.7	4.3 ± 2.2	0.045
Left Hallux – Neutral Position	3.2(2.5-4.8)	3.0(2.5-4.7)	3.2(2.5-4.8)	3.7(2.6-4.7)	0.790
Left Toes – Neutral Position	4.5±1.7	4.6 ± 1.8	4.6 ± 1.9	4.4 ± 1.6	0.901
All Toes (Left) – Neutral Position	5.2±2.1	4.9 ± 2.0	5.4 ± 2.3	5.2 ± 2.2	0.746
Left Hallux – Plantar Flexion	2.4(1.8-3.2)	2.4(2.0-2.8)	2.1(1.8-2.6)	2.8(2.0-4.1)	0.244
Left Toes – Plantar Flexion	2.9(2.1-4.2)	2.5(2.1-3.4)	2.9(2.0-4.4)	3.4(2.7-4.8)	0.054
All Toes (Left) – Plantar Flexion	3.0(2.3-4.4)	2.8(2.2-3.8)	3.0(2.3-4.2)	4.0(2.8-5.1)	0.082

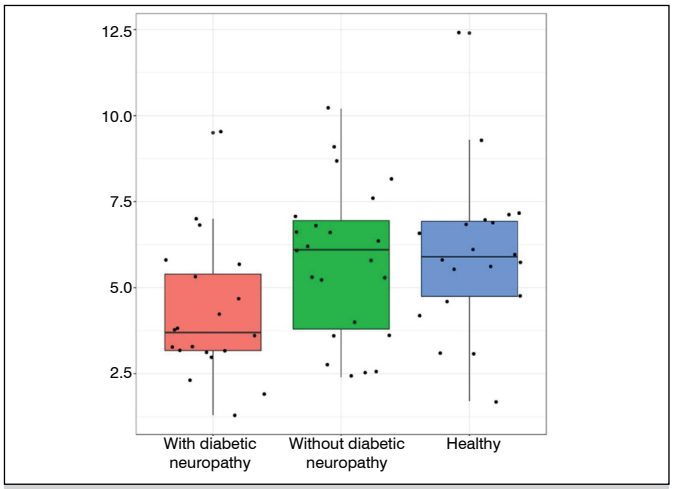

Figure 7. Results obtained for the variable All Toes (Right) – Neutral Position.

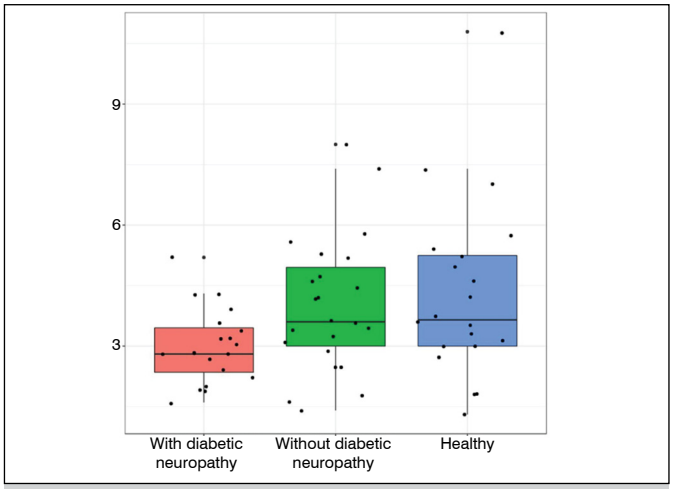

Figure 8. Results obtained for the variable All Toes (Right) – Plantar Flexion.

Evaluation Method and Results

To evaluate muscle strength in the lower limbs accurately and reliably, objective techniques such as dynamometry are preferable to manual muscle testing, which has limitations in sensitivity and reliability.[20] Dynamometry provides quantitative and reproducible measurements, although different types of dynamometers may present variations in muscle strength results.[21] It is important to note that the dynamometers do not activate the muscle; activation is performed by the patient during the test. Studies suggest that by keeping the ankle at maximum plantar flexion, the influence of the extrinsic flexors of the fingers is minimized, allowing for a more accurate evaluation of the intrinsic muscles.[22,23]

Data analysis (Table 1) showed that most variables did not present statistically significant differences between the groups. However, the

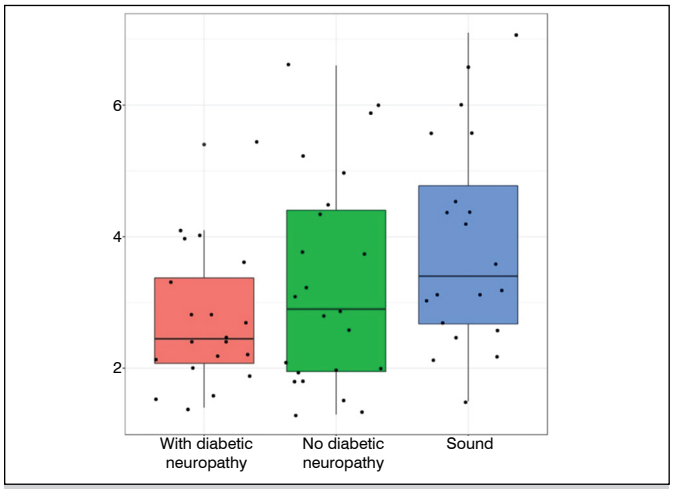

Figure 9. Results obtained for the variable Left Toes – Plantar Flexion.

variables Right Hallux – Neutral Position, All Toes (Right) – Neutral Position, All Toes (Right) – Plantar Flexion, and Left Toes – Plantar Flexion showed statistically significant differences. The comparative graphs (Figures 1–4) corroborate these findings, showing lower median values and narrower interquartile ranges in the groups with diabetic neuropathy.

The data from our study showed that patients with diabetes showed a 14% reduction in the strength of the flexors and 17% in the ankle extensors, plus a decrease in the strength and volume of the intrinsic muscle of the foot.[24] Although these results are indicative, they are not conclusive as to the increased risk of reduction in the strength of the intrinsic muscle of the foot in patients with diabetic neuropathy. Limitations of the study, such as the size of the sample and the lack of data on the time of disease progression and markers of clinical severity, restrict the more robust interpretation of the results.

CONCLUSION

This study sought to explore the strength of the intrinsic musculature of the foot in diabetic patients, using dynamometry as an evaluation tool. Although no conclusive evidence has been found to support the initial hypothesis, the data suggest that dynamometry can play a valuable role in the early detection of muscle weakness in patients with diabetic neuropathy. However, the limited size of the sample and the lack of detailed information on the time of disease progression and markers of clinical severity are limited to the generalization of the results.

It is concluded that this work establishes a basis for future research in the area, proposing a methodology that, with improvements, can contribute to the monitoring of muscle deterioration and the effectiveness of specific treatments in diabetic patients. It is considered essential to enlarge the study, with a larger sample and additional variables, such as disease progression time and progression markers, to validate and refine the preliminary conclusions, with potential to provide an early diagnostic marker of peripheral complications related to diabetes.

CONTRIBUTIONS OF THE AUTHORS

Each author personally and significantly contributed to the development of this article: DMO: Conceptualization, visualization, research, literature review, analysis and manuscript preparation; AAS, TMNTF and LMF: Conceptualization, conception, drafting of the original draft, design, methodology, analysis, revision and editing; FC: Conceptualization, conception, drafting of the original draft, design, methodology, visualization, research, literature review, analysis and preparation of the manuscript; TBF: design, methodology, analysis and preparation of the manuscript, revision and editing, translation review, recommendation of magazines for publication.

DATA AVAILABILITY DECLARATION

The contents underlying the research are available in the manuscript.

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BIOMECHANICAL EVALUATION OF FIXATION METHODS IN PATELLAR FRACTURES (AO 34-C3): AN IN VITRO ANALYSIS

AVALIAÇÃO BIOMECÂNICA DE MÉTODOS DE FIXAÇÃO EM FRATURAS DA PATELA (AO 34-C3): UMA ANÁLISE IN VITRO

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ABSTRACT

Objective: To describe the biomechanical outcomes of five different fixation models, namely: tension band wiring (TB), anterior star-shaped locking plate (PS), lateral orthogonal locking plates (LP), and two additional models using BT and LP combined with a circumferential cerclage wire around the patella, referred to as BTO and PSO, respectively, in a comminuted patellar fracture (AO34C3). Methods: This study analyzed, using the finite element method (FEM), the following variables: total displacement, fragment displacement, maximum and minimum principal stresses, and total and localized von Mises stresses, under loading conditions of 1,500 N (R1) and 3,000 N (R2). Results: Results were presented as absolute values and percentiles, as well as through observational analysis of individual stress distributions. From a biomechanical perspective, critical results in both absolute and percentile values were observed for most variables in the TB group compared with the PS and LP groups. While maximum and minimum principal stresses were similar among groups, differences in intensity and location were found for total and localized von Mises stresses. In addition, the inferior pole of the patella exhibited critical stress conditions across all groups. Conclusions: Tension band wiring demonstrated inferior outcomes compared with locking plates. Biomechanical benefits were observed with the use of two orthogonal locking plates compared with an anterior locking plate; however, both constructs still showed deficiencies in stabilizing the distal pole of the patella. ***Level of Evidence II; Prospective Study.***

Keywords: Finite Element Analysis; Fracturs Fixation; Patella; Bone Plates.

RESUMO

Objetivo: Descrever os resultados biomecânicos de cinco diferentes modelos de fixação: banda de tensão (BT), placa bloqueada anterior em forma de estrela (PE), placas bloqueadas ortogonais laterais (PL) e dois modelos adicionais combinando BT e PE com fio de cerclagem circunferencial ao redor da patela, denominados BTO e PEO, respectivamente, em uma fratura cominutiva da patela (AO 34-C3). Métodos: Este estudo analisou, por meio do método dos elementos finitos (MEF), as seguintes variáveis: deslocamento total, deslocamento dos fragmentos, tensões principais máxima e mínima e tensões de von Mises total e localizada, sob condições de carregamento de 1.500 N (R1) e 3.000 N (R2). Resultados: Os resultados foram apresentados em valores absolutos e percentis, bem como por meio de análise observacional das distribuições individuais de tensão. Do ponto de vista biomecânico, observaram-se resultados críticos, em valores absolutos e percentis, para a maioria das variáveis no grupo BT em comparação aos grupos PE e PL. As tensões principais máxima e mínima foram semelhantes entre os grupos; entretanto, observaram-se diferenças na intensidade e na localização das tensões de von Mises total e localizada. Ademais, o polo inferior da patela apresentou condições críticas de tensão em todos os grupos. Conclusões: A banda de tensão apresentou desempenho biomecânico inferior quando comparada às placas bloqueadas. Benefícios biomecânicos foram observados com o uso de duas placas bloqueadas ortogonais em relação à placa bloqueada anterior; contudo, ambas as construções apresentaram limitações na estabilização do polo distal da patela. ***Nível de Evidência: II; Estudo Prospectivo.***

Descritores: *Análise de Elementos Finitos; Fixação de Fratura; Patela; Placas Ósseas.*

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INTRODUCTION

The patella fracture represents 1% of the skeletal fractures, and can cause significant functional disabilities due to its relationship with the knee extension mechanism.[1-4] The patella supports forces of 3.3 to 7 times the body weight,[5,6] affecting the patellofemoral joint and resulting in comminuted fractures.[7,8]

It is a challenge for the surgeon to treat the biological damage and restore joint congruence, as well as maintain mechanical stability with stable synthesis.[9] The forces in the patella affect the synthesis, which must withstand various biomechanical conditions of the knee (flexion, patellar tilt, tibial and femoral rotation).[10]

Some methods, such as the tension band, have complications (22-30%) due to release, wire migration and local irritation, requiring reinterventions in up to 65% of cases.[7,8,11-14] The previous plates show more promising results, since in the comminuted fractures of the patella allow the individualized fixation of fragments with angular stability.[15,16]

Thus, the biomechanical benefits inherent in the use of plates, when compared to the use of Kirchner wires and voltage bands still need experimental studies making it possible to evaluate the most beneficial hypothesis from a biomechanical point of view.

The authors describe a biomechanical test, using the finite elements method, on a five-part patellar fracture (AO 34C3), fixed with five synthesis models, to evaluate total displacement, fragment displacement and tension distribution in the patella and in the syntheses, using Von Mises evaluation and presenting results in absolute values, percentiles and observational analysis of voltage distribution.

METHODS

Tomographic images were obtained of a left patella with 45 mm wide, 43 mm long and 20 mm thick, extracted from the synthetic model 1145-70 (large size) of the brand SawboneTM, composed of cortical and polyurethane sponge. The images were archived in the DICOM protocol using an Emotion tomograph (16 channels, Siemens, Germany) with a resolution of 512×512 and a cut-off distance of 1.0 mm. The file was imported into the InVesalius™ program for 3D reconstruction and archived in STL format. The study did not use data from humans or any living beings in the research, only synthetic and virtual models of a left patella as described above, thus being dispensed from terms of free and informed consent or approval by ethics committee.

The virtual 3D models of each system (bone, tendon and muscle) were made in the Rhinoceros™ 6 program (Robert McNeel & Associates, USA) and the finite element method (FEM) test was performed in the SimLab™ (HyperWorks, USA) using the solver Optistruct.

Cuts were made in the patella on the axial and coronal axes, reproducing a fracture AO34C3, dividing the patella into five fragments (P): two in the upper pole, lateral (P1) medial (P4), two in the lower pole, lateral (P2) medial (P3) and a central (P5). The fragments had corresponding muscular and tendon inserts, being the higher to the quadricipital tendon (TQ) and the lower to the patellar tendon (TP), with representations of soft parts and a solution of discontinuity near the edge of fractures. (Figure 1A)

The synthesis models for fracture fixation were virtually drawn: Kirchner wire of 2.0 mm (K), circling wire of 1.25 mm (O), locked LCP plates of 2.7 mm with 4 holes (LP), star-shaped locked plate (PS) and screws of 2.7 mm. The dimensions corresponded to a physical model in 316L stainless steel from the brand DePuy Synthes (Switzerland).

The configured groups (n=1) were named according to the type of fixation: Group (TB) with tension band, using two parallel K wires and a circling wire in eight; (Figure 1B) group (PS) with a 2.7 mm star-shaped blocked plate on the front of the patella; (Fig. 1D) group (LP) with two 2.7 mm orthogonal blocked plates, one on the lateral side and one on the medial side of the patella (Figure 1F). Two other groups, TBO and PSO, added O wire to the TB and PS models, circumventing the patella and transferring TP and TQ. (Figure 1C,E) In the TB and TBO groups, the K wires were positioned parallelly in the axial plane for better bone contact area in the peripheral fragments (1, 2, 3, 4). The figure-of-eight tension band wire contacted the posterior surface of the K-wires at their ends, transmitting TQ and TP. In the LP, PS and PSO groups, the plates were positioned with at least two screws in each fragment (P). Angles and lengths of the screws respected the joint surface and sought the best area of bone contact. In the LP and LPO group, the screws reached the opposite side of the patella, respecting the cortical.

After composing the groups, all models were imported to Simlab™ for trials. Each part of the models (cortical bone, sponge, tendon, muscle and steel) was identified and controlled for the mesh, keeping the size of the elements to avoid interference. A tetrahedral element was adopted, with a defined number of nodes and elements. For the simulations, the properties of the materials were defined: cortical bone (17,000 and 0.26), trabecular bone (1,700 and 0.26), patellar ligament and muscles (1,200 and 0.45) and steel alloy (200,000 and 0.29), referring to the elasticity module (Ma) and Poisson coefficient (v).

The trials applied a traction force in the TQ, in the cranial direction in the coronal axis, distributing the total load into three fractions: 50% for the intermediate, 30% for the lateral and 20% for the medial, fixed at the distal end of the TP. Between the charging point and the insertion, there was a bending tilt of 20 degrees relative to the long axis of the femur and 30 degrees to the tibia, supported later by the femoral condyle, drawn according to the patellar joint surface to optimize joint contact in the predetermined angles. (Figure 2A,B) Two loads on the Z-axis were applied to all groups: 1,500 N (R1) and 3,000 N (R2) for physiological and supraphysiologic evaluation. On axes X and Y, there was no charge. Movement restriction regions were determined in the test body and femoral condyle in all directions of the X, Y and Z axes to avoid unwanted displacement and rotations. (Figure 2)

From these conditions, the values of total patella displacement (Desl.T) in mm and localized displacement (Desl.L) in mm were obtained, evaluated individually between the adjacent fragments (Pl to P2, P4 and P5; P2 to P3 and P5; P3 to P4 and P5; P4 to P5). The main maximum voltage (Max.P) and the main minimum voltage

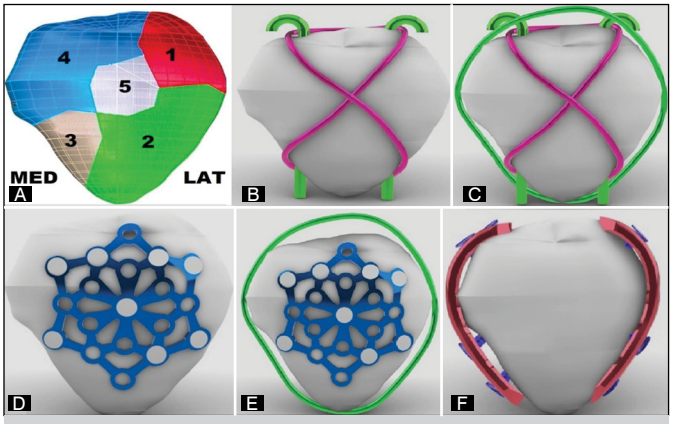

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Figure 1. A) illustration demonstrating the 3D patella model and its respective fragments and faces. B) 3D model of the TB group. C) 3D model of the TBO group. D) 3D model of the PS group. E) 3D model of the PSO group. F) 3D model of the LP group.

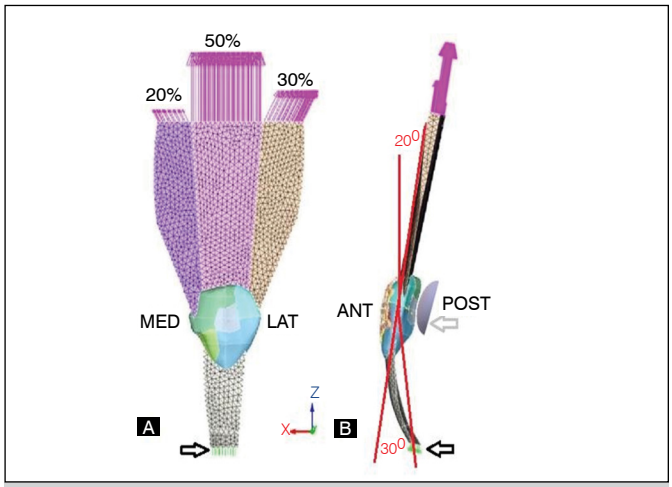

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Figure 2. Conditions and contours of the tests. A) Front view (bottom to top) - Black Arrow - fixation point on the patellar tendon. Pink arrows - locations, axis, direction - fractions of charges applied to the portions of the quadriceps. B) Side view - (lower to top)- Black arrow - fixation point on the patellar tendon. red lines- Representation of the knee flexion angle in 20 degrees with long axis of the femur and 30 degrees with long axis of the tibia.- Grey arrow - femoral condyle.

(Min.P) were also measured, both in MPa, in addition to the total Von Mises voltage (VT) and localized (VL) in the syntheses. The results were presented in absolute values, percentiles and observational analysis of the distribution of tensions.

RESULTS

When we observe Desl. T in R1, the values obtained were: 93.45; 80.47; 87.98; 80.12; 24.65 and in R2:156.14; 135.45; 146.63; 135.99; 43.19 (mm) for TB, TBO, PS, PSO and LP, respectively. The LP group had the lowest value and TB the highest values in both loads. TBO and PSO groups showed reduction relative to their individual pairs, and PS group had higher values compared to TBO group (Table 1). In relation to the percentile of LP and PS, 356% values were observed in R1 and 339% in R2 higher for PS compared to LP. In Desl. L, the fragments of the upper pole showed greater displacements compared to those of the lower pole (P1-P2 and P3-P4). The LP group presented the lowest absolute values. There was a reduction of this variable in the TBO and PSO groups relative to their individual pairs. The PS group had lower values, but close to those of the TBO group in R1, and lower values in R2. (Table 1) THE Max. P, which represents the traction force and its deformities, showed that the values in the LP group were superior to the PS and TB groups in R1 and R2. The TBO group had a lower value than the PSO, indicating lower distribution performance of the tensions in the TB and TBO groups. VT values were: 419.68; 359.97; 1805.53 (MPa) in R1 and 557.2; 720.19; 2171.06 (MPa) in R2, for LP, PS and TB, respectively. This indicates that the tension distribution in the syntheses was more effective in the LP group in R1 and PS in R2 (Table 1), with higher percentiles of LP versus PS at 85.77% in R1 and lower at 129.25% in R2.

In the descriptive analysis of VT, especially in R2, the tension was located in the synthesis in all groups at the inferior-medial pole of the patella, P4. In the TB and TBO groups, the voltage was at the distal end of the medial K wire, in the PS and PSO groups, in the peripheral connection hole at the northwestern tip of the star, and in the LP group, at the base of the screws in contact with the medial plate. The voltage was distributed to the wire "O" in the TBO and PSO groups, reducing the absolute value without changing the location in R1 and R2. (Fig. 3 and 4)

VL values consolidate the results of the variables described, linked to the critical point of tension (break). In R1, the values were: 20.55; 235.12; 148.54 (MPa) and in R2:31.25; 498.12; 304.69 (MPa), for LP, PS and TB, respectively. This indicates effectiveness in the voltage distribution in the LP group, with lower values compared to the PS group, similar in R1 and R2 (Table 1), with percentiles of LP versus PS of 1,144% in R1 and 1,593% in R2.

In the descriptive analysis of VL in R2, there was similarity in the location of this variable with the VT in the PS and PSO group, concentrated in the peripheral connection hole of the northwestern tip of the star, P4. In the TB, TBO and LP groups, there was migration to the median third of the medial K thread in TB and TBO, and to the median third of the medial plate in LP. The distribution of voltage to the wire "O" in the TBO and PSO groups resulted in a reduction compared to the pair without "O". (Fig. 3 and 4)

For Min.P, represented in negative values to indicate direction and direction opposite to Max.P, we observed that the values in the LP group were superior to the PS and TB groups in R1 and R2. The TBO group had a lower value than the PSO, indicating lower distribution performance of the tensions in the TB and TBO groups. Min.T values were:570,67;-298,69;-306,5(MPa) in R1 and -688-599,57;-613.01 (MPa) in R2, for LP, PS and TB, respectively. Compression voltage distribution was most effective in the LP group, similar in R1 and R2. The distribution to the wire "O" was also observed in the TBO and PSO groups, reducing the voltage compared to the pair without "O". (Table 1) (Figures 3 and 4)

DISCUSSION

This study evaluated five types of biomechanical fixation in the patella fracture 34C3 in five parts using FEM. Obtaining as main results less deviation from the fractured fragments, better stability,

Table 1. Variable values by group and load.

Loading	1500 N					3000 N				
Groups	BT	BTO	PS	PSO	PL	BT	BTO	PS	PSO	PL
Desl T (mm)	93.45	80.47	87.98	80.12	24.65	156.14	135.45	146.63	135.99	43.19
Desl 1-2 (mm)	2.45	2.21	2.37	2.24	1.61	4.66	4.48	5.12	4.56	3.84
Desl 1-5 (mm)	0.56	0.41	0.31	0.3	0.12	1.28	1.14	715	0.74	0.45
Desl 1-4 (mm)	0.63	0.54	0.42	0.33	0.19	1.45	1.23	0.96	0.84	0.45
Desl 2-3 (mm)	0.51	0.36	0.3	0.29	0.1	1.18	0.83	0.74	0.71	0.24
Desl 2-5 (mm)	1.51	1.32	1.46	1.26	1.11	3.47	3.05	3.45	2.94	2.68
Desl 3-4 (mm)	2.21	2.17	2.12	2.06	1.51	5.03	4.99	4.81	4.84	3.47
Desl 3-5 (mm)	0.53	0.42	0.41	0.35	0.28	1.23	0.96	0.98	0.81	0.65
Desl 4-5 (mm)	0.61	0.45	0.34	0.33	0.13	1.41	1.25	0.78	0.81	0.49
Max.P (MPa)	180.51	171.05	213.65	205.45	499.88	361.03	342.1	428.25	410.91	701.95
VT (MPa)	1085.53	1353.43	359.97	1308.09	419.68	2171.06	2712.86	720.19	2616.19	557.2
VL (MPa)	148.54	84.62	235.12	158.98	20.55	304.69	121.65	498.12	212.98	31.25
Min.P (MPa)	-306.5	-281.98	-298.69	-327.71	-570.67	-613.01	-563.97	-599.57	-655.42	-688

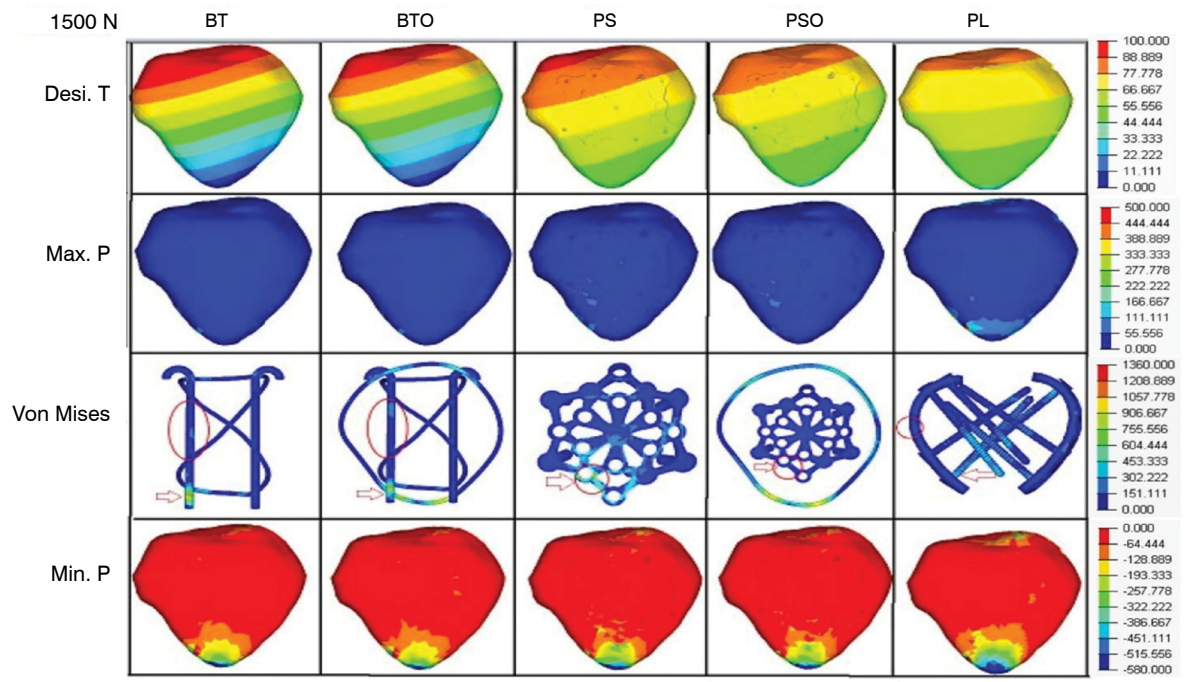

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Figure 3. Loading 1500N- Images of the groups and variables with their respective grading scales. (red arrow) - Von Mises total. (red circle) - Von Mises located.

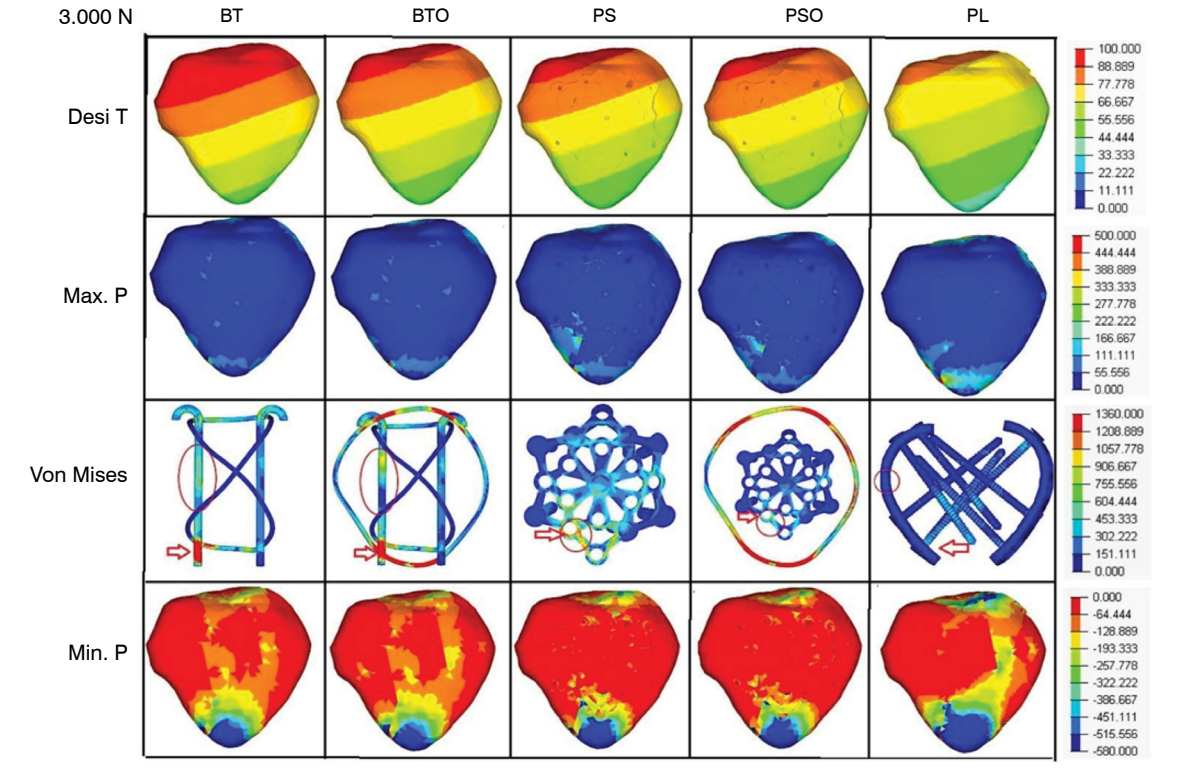

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Figure 4. Loading 3000N- Images of the groups and variables with their respective grading scales. (red arrow) - Von Mises total. (red circle) - Von Mises located.

with the uses of orthogonal or previous plates when compared with the tension band, these results are similar to those obtained by Kfuri et al.[15]

The method allowed to compare quantitative and observational variables, focusing on the distribution of tensions, this visual approach helps to understand the behavior of the variables in the fixation groups of these complex fractures, bringing to light observational possibilities only possible with the use of FEM.

Biomechanical studies of comminuted patella fractures are rare in the literature, [15-17] possibly due to the reproduction difficulties of these tests, these difficulties minimized by the use of FEM. This method can reproduce complex conditions and is widely validated in traumatology.[18,19]

The biomechanics and anatomy of the knee are complex, involving multiple intrinsic variables, musculature, ligaments and adjacent joints. Even with the FEM, the reproduction of these conditions is a search for accurate information for research, considering various descriptions of conditions, contours, positioning, values and axes of loads, influential anatomical structures, among others. In addition to the FEM, counter tests are also important, although they may present vulnerabilities and biases.

The authors applied anatomical and biomechanical details, considering the patella and the patellofemoral joint and their possible deformations. They included the fractional application of the load on the femoral quadriceps and its axes described by Mesfar et.al.[20] total load between three times the body weight of an adult from 50 to 100 kg, described by Zderic et.al. 2017,[6] angles of the load and the degree of flexion of the knee with the FEM, described by Ling et.al.[21] These criteria were selected by associating significant load to the patella and patellofemoral joint at their moment of highest tension.[22,23]

It is possible to present some comparative observations to them, which are: the presence of excessive Desl.T in the group TB in relation to the other groups, linked mainly to the fragments of the upper pole(1-4) in relation to the lower(2-3) and the medial fragments (3-4) in relation to the laterals(1-2), similar to the one described by Kfuri et al.[15,12,24] with smaller displacements to the groups with blocked plates.

The lower pole of the patella was close to some form of fixation in the PAO and TBO groups, showing tension in the wire “O” in the transfixation area, resulting in tensional changes and small displacement reductions. Stoffel et al.[17,24]

This study presents limitations, as it did not consider the chondral component of the patellofemoral surface, the surface of the femoral condyle, the mechanical properties of the synovial liquid, the actions of ligaments and meniscal structure. It also considered only one angle of bending of the knee, restricting the trial on the three axes and preventing the rotational mobility of the patella. It is also worth mentioning as a limitation the absence of other fixation methods used in the treatment of this type of fracture (canular screw + tension band).

As strengths, attention is highlighted to the details of conditions and contours, considering the most important factors for this type of study, and the observational similarities with research using fresh cadaveric models. Future studies should contemplate new designs of previous blocked plates, which can optimize stability in areas of demonstrated fragility.

CONCLUSION

In the fractures 34C3 of the patella evaluated by the FEM, the tension band has lower results when compared to the blocked plates, and biomechanic benefits were perceived in fixing this fracture model when we used two orthogonal plates blocked on the sides (medial and lateral) of the patella and when used the previous blocked plate, but both still with deficiencies in the stabilization of the distal pole.

CONTRIBUTIONS OF THE AUTHORS

Each author personally and significantly contributed to the development of this article: HSSGM and AF: manuscript writing; LRB: data curation and article method. SLMN: editing, reviewing data and results, validating the process; MAF and BNO: reviewing and approving the final version of the manuscript; HSSGM: Writing the work, data analysis, editing; AF: conceptualization, writing the work, approval of manuscript, revision.

DATA AVAILABILITY DECLARATION

The data will be made available when requested.

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ORIGINAL ARTICLE
ORTHOPEDIC TRAUMA

HUMERAL SHAFT NONUNION. RETROSPECTIVE STUDY OF SEVERAL SURGICAL TECHNIQUES

PSEUDOARTROSE DIAFISÁRIA DO ÚMERO. ESTUDO RETROSPECTIVO DE DIVERSAS TÉCNICAS CIRÚRGICAS

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ABSTRACT

Objective: To evaluate demographic aspects, risk factors, trauma mechanisms, and therapeutic outcomes in humeral shaft nonunion. Methods: Retrospective analyzing of 14 patients treated between 2011 and 2024 at a quaternary hospital. Evaluating Demographic data, fracture characteristics, treatments, and outcomes were descriptively assessed. Results: The average patient's age was 51 years. Half of the patients were involved in domestic accidents, and half in traffic accidents. 58% of fractures occurred in the middle third of the shaft, and 93% were classified as atrophic non-unions. Healing failure was linked to lack of bone contact, implant loosening, and infection. After definitive treatment, 71% achieved union, 21% remained in nonunion. Bone grafting combined with compression was associated with superior outcomes. Conclusion: Trauma mechanism, AO classification, and comorbidities were not significant predictors of healing. Grafting associated with compression was crucial for union achievement. ***Level of Evidence IV; Case series.***

Keywords: Pseudoarthrosis; Humeral Fracture; Fracture Healing.

RESUMO

Objetivo: Avaliar aspectos demográficos, fatores de risco, mecanismos de trauma e desfechos terapêuticos na pseudoartrose da diáfise do úmero (PDU). Métodos: Análise retrospectiva de 14 pacientes tratados entre 2011 e 2024 em hospital quaternário, com análise descritiva de dados demográficos, características das fraturas, tratamentos e desfechos. Resultados: A média de idade dos pacientes foi de 51 anos. Metade dos casos sofreu acidentes domésticos, e a outra metade, acidentes automobilísticos. 58% das fraturas ocorreram no terço médio da diáfise, com predominância de pseudoartrose atrófica (93%). Falhas de consolidação foram associadas à infecção, ausência de contato ósseo e soltura de material. Após o tratamento definitivo, 71% consolidaram, 21% permaneceram em pseudoartrose. A utilização de enxerto ósseo e compressão demonstrou melhores resultados. Conclusão: Mecanismo de trauma, classificação AO e comorbidades não foram fatores preditivos significativos. A combinação de enxertia óssea e compressão demonstrou bons resultados na consolidação. ***Nível de Evidência IV; Série de casos.***

Descritores: *Pseudoartrose; Fratura do Úmero; Consolidação da Fratura.*

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INTRODUCTION

Humer diaphysis fractures comprise 1% of all fractures, and the incidence of humeral shaft nonunion (HSN) remains low. According to the French Agency of Information on Hospital Care (ATIH), 8% of diaphysis fractures evolved to non-union.[1]

Sarmiento reported consolidation rates above 95% for humeral shaft fractures treated non-operatively; however, more recent authors have not achieved the same effectiveness with non-operative treatment.[2] Toivanen et al., in 2005, reported a nonunion rate of 22.5% in non-surgical management, mainly associated with fractures in the proximal segment.[3] Rutgers and Ring, in 2006, had failure in 10% of their patients treated non-operatively, also more related—similarly to Toivanen et al.—to fractures in the proximal segment of the humeral shaft.[4] The indications for surgical treatment have been expanding, being absolute (open fracture associated with nerve injury, polytrauma, association with articular fracture, fractures in pathological bone, floating elbow, and failure of non-operative treatment) and relative (associated obesity, muscular atrophy, bilateral fracture, and association with brachial plexus injury).[5] However, between operative and non-operative approaches for humeral shaft fractures, nonunion has remained a complication.

All authors declare no potential conflict of interest related to this article.

The study was conducted at Santa Casa de Misericordia de Sao Paulo, Trauma Group, Department of Orthopedics and Traumatology, R. Dr. Cesario Mota Junior, 112, Vila Buarque, Sao Paulo, SP, Brazil. 01221-010.
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According to Gonçalves et al., pseudoarthrosis reached 10.3% of patients treated with open reduction and internal fixation, the method still considered today the most efficient, said gold standard.[6] Thus, it becomes necessary to study the complications arising from the various therapies used in cases of diaphyseal fractures of the humerus. Another complication associated with poor prognosis of humeral shaft nonunion (HSN) is infected pseudoarthrosis, which in turn has risk factors such as high-energy trauma with exposure, advanced age, extensive soft tissue loss, immunodeficiency, diabetes, smoking, vascular insufficiency, and prolonged hospital stay, among others.[7]

OBJECTIVE

Analyze possible demographic factors, risk factors, trauma mechanisms that led to HSN and how the procedures carried out led to various outcomes in the treatment of the disease.

METHOD

The data used were from medical records, radiographs and tomographs from the hospital's files. Demographic characteristics of patients were collected: age, gender, location of fracture trace, dominance, comorbidities, classification of trauma mechanism, laterality, initial treatment and definitive treatment. In addition, the date of the fracture, the initial approach, the date of the final approach, the use or not of the graft, methods, principles of stability used and time between the approaches were analyzed. The data was analyzed descriptively. We used Weber and Cech's radiographic classification developed in 1976 that divides pseudoarthrosis into hypertrophic, oligotrophic and atrophic.[8]

The terms of consent were signed by the patients we gained access to; however, the same process was not carried out for those whose contact was not effective. The waiver request was requested and approved by the Ethics Committee (CAAE: 83200924.0.0000.5479). From January 2011 to July 2024, we analyzed 17 patients who had HSN in a quarterly health care institution.

The inclusion criteria are pseudoarthrosis of membrane fracture of the homer (defined in this study as lack of evidence of union after a minimum period of six months) and maintenance of outpatient follow-up. We use the AO classification that defines the diaphysis as the part of the bone that is located between the distal and proximal extremities that is limited by the Heim square. Therefore, cases that did not fall within these were excluded.[9]

Patients with incomplete data, poor quality X-rays, patients with consolidation subsequently classified as retarded and not as pseudoarthrosis and those who did not agree to participate in the study were excluded. There were 14 patients who met the exclusion and inclusion criteria.

The patients analyzed had an average age of 51 years (26 - 77), of the 14 patients analyzed seven are men. Comorbidities were found in six (43%) patients. Among the comorbidities, three patients are smokers, three are hypertensive, and one has heart disease. All patients were right-handed, ten (72%) suffered fractures in the non-dominant limb.

RESULTS

The trauma mechanisms observed in the treated patients seven (50%) suffered car accidents and seven (50%) domestic accidents (fall from stairs, fall to the ground). Of the car accidents, six evolved to consolidation after the treatment of pseudoarthrosis and one remained in pseudoarthrosis. In the domestic accidents four consolidated, one was performed endoprosthesis and two continued in pseudoarthrosis.

Regarding the fracture location, eight (58%) were in the middle third, four (28%) in the proximal third, and two (14%) in the distal third of the diaphysis. The fracture types were as follows: five oblique, one spiral, seven transverse, and two comminuted.

In the AO classification of fractures, among the patients analyzed, three 12 A1, five 12 A2, two 12 A3, two 12 C2, two 12 C3. (Table 1)

In the initial approach, inadequate treatment with a bone clamp was performed in six (43%) of the patients, two (14%) patients performed bridge plate (one by the technique of Belangeiro and Livani, a blocked plate), two (14%) patients used compression plate and traction screw, three (22%) performed compression plate, one (7%) prosthesis and double orthogonal plate.

Of the 14 patients analyzed, 13 were considered with pseudoarthrosis atrophic and one with hypertrophic. In nine (64%) there was a need to use a graft in the final treatment.

Among the treatments for pseudoarthrosis, one opted for compression board and shortening more traction screw in two (14%), compression board in eight (58%), infection treatment in two (14%) and endoprosthesis one (7%), wave plate in one (7%).

The radiographic factors observed in cases of humeral diaphysis fractures that progressed to pseudoarthrosis were lack of fragment contact in seven (50%) patients, loosening of fixation material in four (28%), and fracture-related infection in three (22%).

Of the patients analyzed after follow-up time, ten (71%) showed consolidation, three (21%) remained in pseudoarthrosis and in one patient (7%) there was a need to perform endoprosthesis. After the surgical procedure two 12 A2, a 12 C2 maintain the outcome of pseudoarthrosis. A 12 C3 endoprosthesis was performed.

Table 1. Results.

	AO Classification	Type of pseudoarthrosis	Final treatment	Use of Graft	Consolidation at the end of treatment
1	12 A2	Atrophic	Neutralization plate and traction screw	Y	Y
2	12 A2	Atrophic	Treatment of infection	N	N
3	12 A2	Atrophic	Compression plate	Y	N
4	12 C2	Atrophic	Compression plate	Y	N
5	12 A2	Hypertrophic	Compression plate	N	Y
6	12 C3	Atrophic	Neutralization plate and traction screw	Y	Y
7	12 C2	Atrophic	Compression plate	Y	Y
8	12 A3	Atrophic	Compression plate	Y	Y
9	12 A3	Atrophic	Compression plate	Y	Y
10	12 A2	Atrophic	Compression plate	Y	Y
11	12 A1	Atrophic	Treatment of infection	N	Y
12	12 C3	Atrophic	Endoprosthesis	--	--
13	12 A1	Atrophic	Plate in wave	Y	Y
14	12 A1	Atrophic	Compression plate	N	Y

Legend: Y-Yes; N-No. **Source:** Database of patients of orthopedics and traumatology of the hospital Santa Casa de Misericórdia de São Paulo.

DISCUSSION

Despite the therapeutic possibilities, there is no consensus or precise protocols on the best strategy to be adopted in the context of pseudoarthrosis of the humerus.[10]

In disagreement with Raven et al. we were unable to correlate high energy as a risk factor for HSN and neither the mechanism of trauma nor the severity of the fracture taking as reference the AO classification proved relevant in our patients.[7]

Peters et al. in a systematic review with metanalysis concluded that the open reduction and fixation with plates and screws associated with the autologous graft achieved higher consolidation rates and a small rate of complications; similarly, the external fixation also presented good bond rates, in addition to presenting an advantage when the patient presented infection according to the authors.[11] In eight (57%) of the 14 patients, we achieved increased stability with plates and screws as a definitive approach and 71% (ten) of these treatments with focus compression either by compression by the plate or by neutralization plate associated with traction infusion showed consolidation.

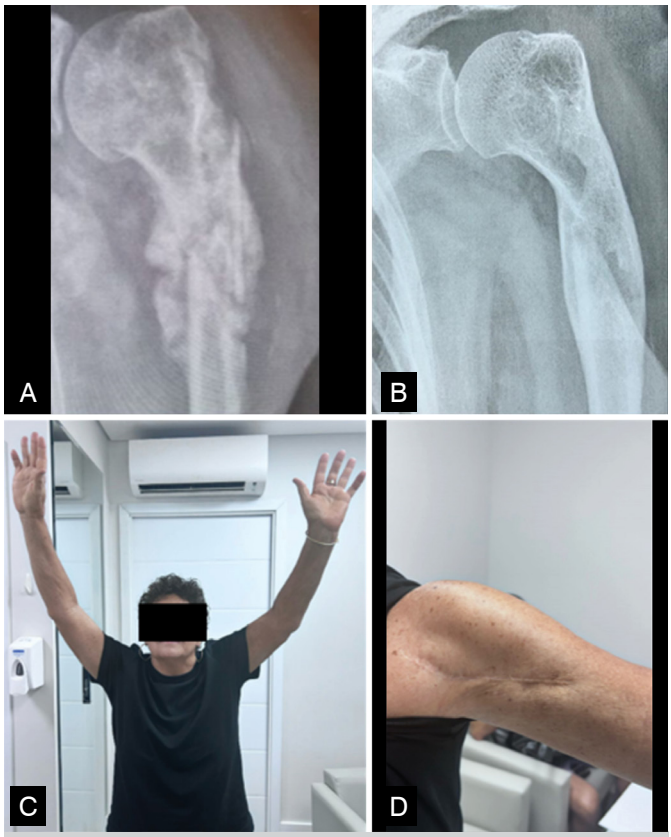
Fozzato et al. concluded that, in addition to the open reduction and internal fixation with plates and screws in a stable way associated with corticospongiform graft, the complete removal of non-viable tissue from the pseudoarthrosis focus led to higher ratio of union.[12] In this context, we performed this revitalization with insertion in seven patients, of which five showed consolidation. Healy et al. recommend the ingestion as a routine, and should not be reserved only for cases of atrophic pseudoarthrosis.[13] Despite little mention, we found that the acute shortening with compression could also help revitalize, exposing and coupling viable edges with better quality bone. After resection of the devitalized tissues, this shortening was performed in nine patients.

We observed that the stable fixation with plates and screws presented good results, however Patel et al. demonstrated that another method – Ilizarov's circular external fixator – also presented high consolidation rates, having achieved union in 15 of 16 patients.[14] In our case, in a patient with infection already under control, treatment with linear fixator was attempted, unsuccessfully and with reabsorption of the graft. We only obtained consolidation when we exchanged the fixer for revitalization, shortening, grafting and compression with plate and screws. A second patient was also treated with a monoplanar external fixator for infection control. Although the bacterial infection was resolved with cleaning, debridement, and placement of antibiotic-loaded cement, bone consolidation was still not achieved. Despite the pseudoarthrosis and functional limitations, the patient declined further surgery. Our experience suggests that the use of a monoplanar external fixator is effective for infection control when combined with cleaning, debridement, and placement of antibiotic cement, but bone consolidation was not achieved in either of the two patients.

Although most studies indicate that the main modifier of the natural history of pseudoarthrosis is the gain of stability and osteogenic stimulation, we observed that, in the context of infected pseudoarthrosis, one of our patients presented consolidation of the fracture while treating the infection, even after removal of synthesis material. This patient did not use the fixator because the fracture was in the proximal diaphysis region. Opted for the time by removing the plate and screws, cleaning, revitalizing and placing cement with local antibiotic. The proximal region was preserved considering the next fixation procedure. However, after four weeks, the cement with antibiotic was removed and the fracture was consolidated, not requiring further fixation. (Figure 1)

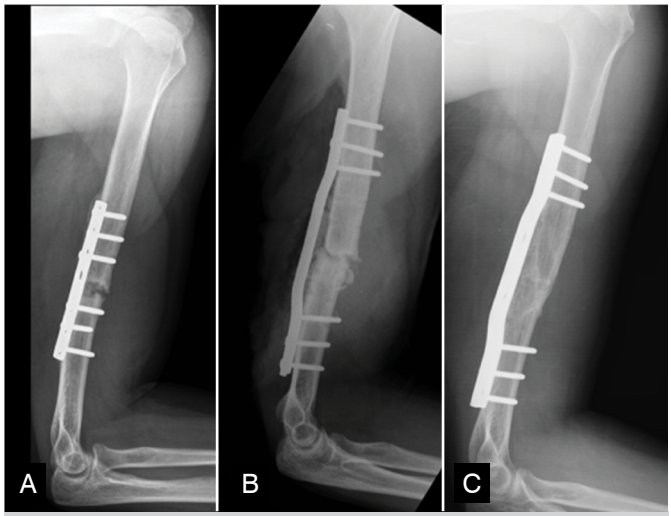
In one of the patients who presented with atrophic HSN, we used RAFI with a long plate, wave-shaped with graft. The reason was that there was great bone loss in the screw holes due to the release of the implant associated with non-consolidation, being enlarged the working area with a long plate protecting these holes and being fixed in viable bone, which was wave-shaped for placement of tricortical graft under the implant and spongy in the cortical *trans*, leading to the consolidation of pseudoarthrosis. (Figure 2)

Of our 14 patients, 13 presented atrophic HSN; of the patients with atrophic pseudoarthrosis, three initially presented characteristics



Source: Personal file of the authors, with informed consent and anonymity guaranteed.

Figure 1. X-rays of patient 11 showing infected hypertrophic pseudoarthrosis (A) and consolidation (B) after treatment of infection; and clinical images of functionality (C) and scar (D).



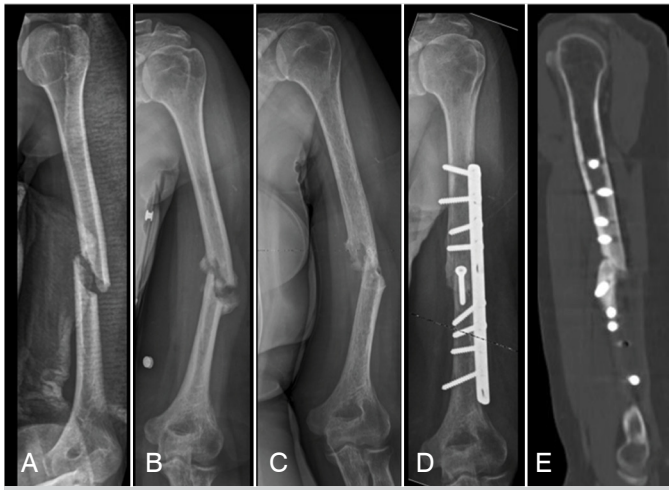
Source: Xero Viewer Data Base from the Santa Casa de Misericórdia de São Paulo.

Figure 2. X-rays of the patient 13 showing atrophic pseudoarthrosis with reabsorption of the fracture focus (A), after graft placement and fixation with wave-molded plaque (B) and after bone consolidation (C).

interpreted by us as an attempt to form bone calo, initially classified as hypertrophic pseudoarthrosis, however, in the follow-up, this tissue was reabsorbed, becoming considered pseudoarthrosis avascular. These three were under 60 years old and had fragmented fractures. Knowing that these patients initially classified as hypertrophic pseudoarthrosis evolved into atrophic pseudoarthrosis, we decided to recommend ingestion for all cases of pseudoarthrosis. (Figure 3)

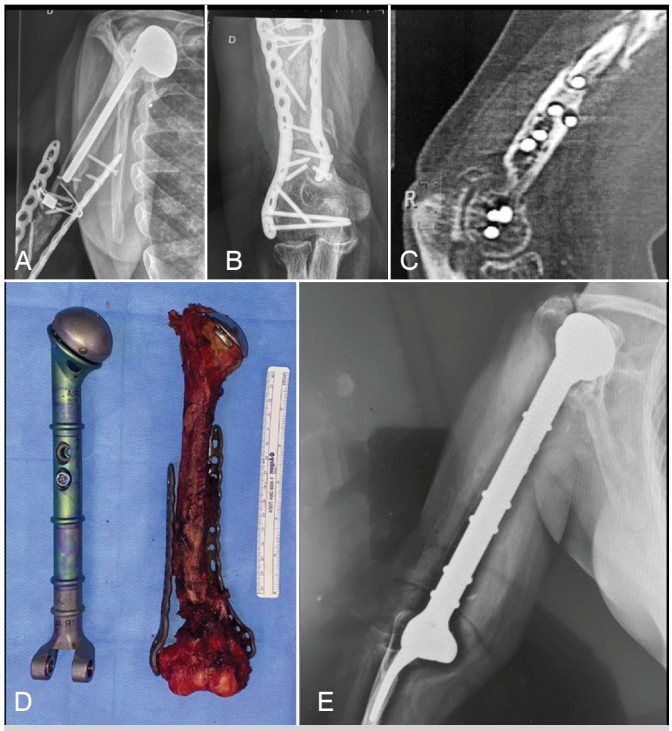
The patient who needed to undergo bone replacement by a total endoprosthesis of the humerus had two focuses of pseudoarthrosis in the diaphysis despite several surgeries. There was rupture and release of the implants. As an antecedent, she had rheumatoid arthritis and had done a complete prosthesis of the ipsilateral shoulder more than ten years ago.

One of the limiting factors was that patients had irregular follow-up and a temporary standardization was not possible. (Figure 4)



Source: Xero Viewer Data Base from the Santa Casa de Misericórdia de São Paulo.

Figure 3. Images of the patient 1. X-rays showing the initial aspect of the fracture, evolution of 17 months, 29 months, 41 months (A, B, C and D); and tomography after 41 months of the fracture (E).



Source: Personal file of the authors, with informed consent and anonymity guaranteed.

Figure 4. Images of the patient 12. X-rays and tomography showing the two focuses of pseudoarthrosis, fracture and release of the implant and the complete prosthesis of the shoulder (A, B and C). Image of the resected humerus and evidence of the endoprosthesis model used (D). Final X-ray with total replacement of humerus (E).

CONCLUSION

Mechanism of trauma, initial AO classification of the fracture and comorbidities were not significant for consolidation of patients in this series of cases. The best results were obtained from the use of the graft and compression of the pseudoarthrosis focus with or without shortening.

CONTRIBUTIONS OF THE AUTHORS

Each author personally and significantly contributed to the development of this article: RELTP, VNQB and EBS: data collection; RELTP and VNQB: data maintenance and insertion on the collection platform; JOSH, RWC and CZ: Article review, methodology and supervision; JOSH, RWC and CZ: formal analysis and data curation; CZ: Validation and writing – review and editing; JOSH, RWC and CZ: performed surgeries used as data in the study.

DATA AVAILABILITY DECLARATION

The contents underlying the research are available in the manuscript.

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ANEURYSMAL BONE CYST: A CASE SERIES OF AN AGGRESSIVE BENIGN TUMOR

CISTO ÓSSEO ANEURISMÁTICO: SÉRIE DE CASOS DE UM TUMOR BENIGNO AGRESSIVO

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ABSTRACT

Objective: The Aneurysmal Bone Cyst (ABC) is a benign yet aggressive bone tumor. This study aimed to evaluate sex, age, tumor location, tumor size, type of treatment (surgical, infiltration, embolization), and recurrence. Methods: Descriptive and quantitative statistical analyses were applied. The prevalence ratio and 95% confidence interval were calculated for the association between recurrence and sex, age, tumor size, Capanna's classification, and treatment type. Results: Twenty-three cases of ABCs were included, eleven (47.8%) females and twelve (52.2%) males. The mean age of treated patients was 11.2±1.8 years. Most cysts were located in the lower limbs (56.5%). The mean follow-up time was 42.8±14.01 months. The mean cyst diameter at the beginning of treatment was 5.58± 1.04 cm; of these, 17.4% were up to 3 cm, 43.5% from 3.1 to 6 cm, and 39.1% over 6 cm. Regarding initial treatment, 6 (26%) patients received infiltration, and in total 20 (86.9%) underwent surgery with bone grafting. The overall recurrence rate was 30.4%. No association was identified between recurrence and the variables studied ($p \geq 0.05$). The epidemiological data obtained are consistent with pediatric cohorts reported in the literature. Conclusion: All evaluated methods are suitable for treating aneurysmal bone cysts. ***Level of Evidence IV; Case Series.***

Keywords: Calcitonin; Child; Adolescent; Neoplasms; Benign Neoplasm.

RESUMO

Objetivo: O Cisto Ósseo Aneurismático é um tumor benigno, porém agressivo. O objetivo deste estudo foi avaliar sexo, idade, localização e tamanho do tumor, classificação, tratamento realizado (cirúrgico, infiltração, embolização) e recidiva. Métodos: Foi aplicada análise estatística descritiva e quantitativa para calcular a razão de prevalência e o intervalo de confiança de 95% entre presença de recidiva e sexo, idade, tamanho, classificação e tipo de tratamento. Resultados: Foram incluídos 23 casos de COA, sendo onze do sexo feminino e doze do sexo masculino. A média de idade foi de 11,2±1,8 anos. A maioria dos cistos localizava-se nos membros inferiores (56,5%). O tempo médio de seguimento foi 42,8±14,01 meses. O diâmetro médio dos cistos no início do tratamento foi 5,58±1,04 cm; destes, 17,4% tinham até 3 cm, 43,5% entre 3,1 e 6 cm, e 39,1% mais de 6 cm. Quanto ao tratamento inicial, 6 (26%) pacientes receberam infiltração e, ao todo, vinte (86,9%) foram submetidos à cirurgia com colocação de enxerto. A taxa global de recidiva foi 30,4%. Não identificamos associação entre recidiva e as variáveis estudadas ($p \geq 0,05$). A epidemiologia observada corresponde às coortes pediátricas encontradas na literatura. Conclusão: Todos os métodos avaliados são adequados para o tratamento do cisto ósseo aneurismático. ***Nível de Evidência IV; Série de Casos.***

Descritores: *Calcitonina; Criança; Adolescente; Neoplasia; Neoplasia Benigna.*

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INTRODUCTION

The aneurysmal bone cyst (ABC) was first described by Jaffe and Lichtenstein, then known as Jaffe-Lichtenstein Disease.[1] Previously classified as a pseudotumoral lesion, it was reclassified in 2020 in the World Health Organization's tumor classification compendium as a benign tumor lesion and grouped with giant cell tumor and non-ossifying fibroma as osteoclastic lesions rich in giant cells.[2] This occurred after the finding that the pathogenesis of ABC originates from the translocation of the USP6 gene, evidenced by in situ hybridization *in situ* by fluorescence (FISH).

This tumor most frequently affects children and young adults, mainly in the second decade of life, with a slight predominance in

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females.[3] The lesions preferentially occur in the metaphysis of long bones, predominantly in the femur, tibia, and humerus.[4] Although rare, they can occur in small tubular bones such as metacarpals, metatarsals, and phalanges.[5-7] In lesions of the spine and pelvis, they represent greater difficulty for surgical treatment.[8-10]

The radiological characteristic is a lytic lesion, typically eccentric and expansive, with a preference for the metaphyseal region of long bones. Lytic lesions may present cortical thinning and widening of the affected segment.[2,3] The appearance of the lesions shows marked thinning of the cortex over the site, with minimal bone formation; changes are sometimes better visualized on computed tomography or magnetic resonance imaging.[11]

Although ABC is benign, there may be clinical and imaging characteristics that denote aggressiveness.[10] Surgical treatment options include simple resection in non-displaceable bones or curettage of the lesion, associated with local adjuvant therapy and filling the tumor bed with bone graft or polymethylmethacrylate.[10,11] The prognosis after treatment is considered good, although about 20% of cases present recurrence.[10] A treatment program based on the evaluation of the morphological type and aggressiveness of these tumors is recommended and recognized as the Capanna Classification.[12] The aim of our study was to evaluate a series of cases of children and adolescents with aneurysmal bone cysts and compare the treatment outcomes.

METHODS

A retrospective case series study on the aneurysmal bone cyst was conducted. Data were compiled from patients' electronic clinical and imaging records, and a specific database was created for the study with full protection of patient identity. The study was approved by the institutional Ethics Committee and is registered in the Brazil Platform under the number CAE 25729119.0.0000.5505.

The records of 23 patients who underwent surgical and/or clinical treatment for cystic lesions suggestive of ABC with the first consultation in the service between January 1, 2005, and December 31, 2023, were analyzed.

The inclusion criteria for the study were patients with an anatomopathological diagnosis of aneurysmal bone cyst, aged between 1 and 30 years at diagnosis, treated at the institution, and who agreed to participate in the study, with signed informed consent and assent form. The exclusion criterion was noncompliance by the patient or their legal representative with participating in the study at any time.

We used Microsoft Excel (Microsoft Office®) for creating the database and tables, and SPSS® V26 (2019) and Minitab 21.2 (2022) for statistical analysis.

All patients were evaluated according to the epidemiological variables: (1) sex; (2) age; (3) tumor location; (4) tumor size (largest diameter); (5) Capanna radiological classification for ABCs;[12] (6) type of treatment performed (surgical, calcitonin injection, embolization, or a combination of modalities), (7) follow-up time in months, and (8) recurrence of the lesion. Epidemiological analyses of the studied variables were performed, with description of categorical variables (frequency and percentage) and continuous variables (mean and standard deviation). Fisher's exact test was used in analyses where the smallest frequency studied was less than 5. The prevalence ratio was calculated with a 95% confidence interval.

RESULTS

Table 1 presents complete data on sex, age, location, size, classification, type of treatment (infiltration, embolization, surgery), and recurrence status.

Twenty-three patients were included in the study, with eleven (47.8%) being female and twelve (52.2%) being male. As this is a pediatric hospital, the average age of the treated patients was 11.2±1.8 (4 to 18 years). The graphs in Figures 1 and 2 compare recurrence with the factors "Sex" and "Age", respectively. The results were not statistically significant.

Table 1. Descriptive analysis of the cases.

Cases	Sex	Age	Location	Cyst size (largest measurement)	Capanna classification	Type of treatment	Recurrence
1	F	16	Humerus	4.5 cm	I	C	Yes
2	M	14	Tibia	8.6 cm	III	C	No
3	F	12	Tibia	6.0 cm	II	C	Yes
4	F	12	Tibia	4.6 cm	III	C	No
5	F	7	Fibula	5.0 cm	II	C	No
6	M	6	Column	3.2 cm	II	I + E	No
7	M	17	Fibula	10.6 cm	II	C	No
8	M	8	Metatarsus	2.2 cm	I	C	No
9	M	18	Pelve	6.9 cm	II	I	No
10	F	4	Tibia	4.2 cm	III	C	Yes
11	M	6	Tibia	7.0 cm	II	I	Yes
12	F	8	Femur	7.0 cm	III	I + C	No
13	M	15	Metacarpus	4.5 cm	II	C	No
14	M	15	Column	6.2 cm	II	E + C	No
15	M	6	Metacarpus	2.4 cm	II	C	No
16	F	6	Metacarpus	2.4 cm	II	C	Yes
17	F	8	Talus	4.2 cm	II	I + C	Yes
18	M	13	Tibia	8.5 cm	III	C	No
19	F	15	Femur	11.2 cm	II	I + C	No
20	F	17	Radius	2.8 cm	I	C	No
21	M	14	Clavicle	3.4 cm	II	C	No
22	F	9	Femur	7.5 cm	II	C	No
23	M	12	Humerus	5.5 cm	II	C	Yes

F: Female; M: Male; I: Infiltration; C: Surgery; E: Embolization; I + E: Infiltration plus embolization; I + C: infiltration plus surgery; E + C: Embolization plus surgery

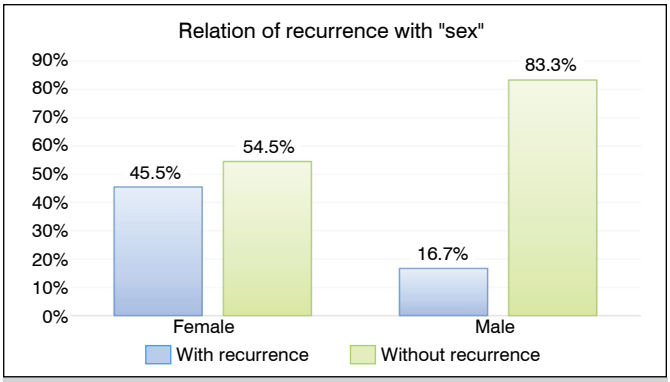

Figure 1. Bar graph of the relationship between recurrence and the qualitative factor "sex".

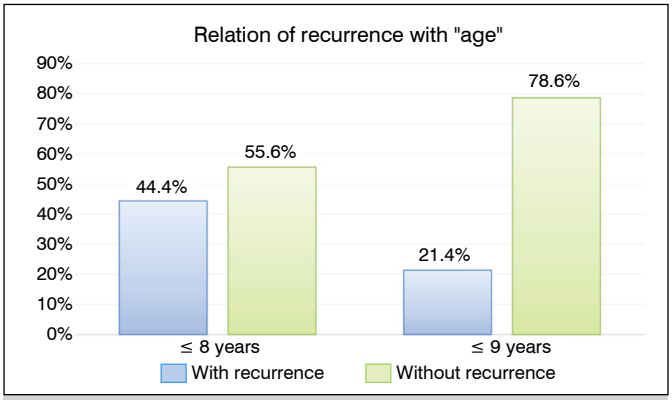

Figure 2. Bar graph of the relationship between recurrence and the qualitative factor "age".

Regarding the location of the ABC, we observed that the vast majority were located in the lower limbs (56.5%). Figure 3 shows the distribution of the topographic location of the ABCs.

The tumor sizes were evaluated as the average of the largest diameter on the initial treatment radiograph, which was 5.58±1.04 cm, with the smallest 2.2 cm and the largest 11.2 cm. Of these, 17.4% corresponded to cysts up to 3 cm, 43.5% from 3.1 to 6 cm, and 39.1% to cysts above 6 cm. The comparison of these factors with recurrence is illustrated in the graph in Figure 4, but the result was not statistically significant.

The radiographic classification of Capanna for the ABC was proposed in 1985, based on the radiographic appearance and morphology of these tumors. The classification is divided into five subgroups, as shown in Figure 2. Type I represents lesions centered in the metaphysis, without causing cortical thinning or expansion. Type II involves expansive tumors with cortical thinning that completely involve the affected metaphysis. Type III is the most common type reported by Capanna et al., characterized by an eccentric metaphyseal lesion that typically affects only one cortex. Type IV corresponds to subperiosteal lesions, which grow away from the bone, and type V involves periosteal lesions that expand around the bone and eventually penetrate the cortex below.[12] Figure 5 presents a comparative graph of recurrence rates for the types of Capanna found in the study patients, with equivalent recurrence rates for types I and II and slightly lower for type III.

Regarding treatment, 6 (26%) patients received infiltration: 5 (83.3%) received calcitonin infiltration, and 1 (16.7%) received dexamethasone and calcitonin infiltration. The dose of each calcitonin infiltration was three ampoules (3 ml) at a concentration of 100 UI/ml. In the case involving the association of calcitonin with dexamethasone,

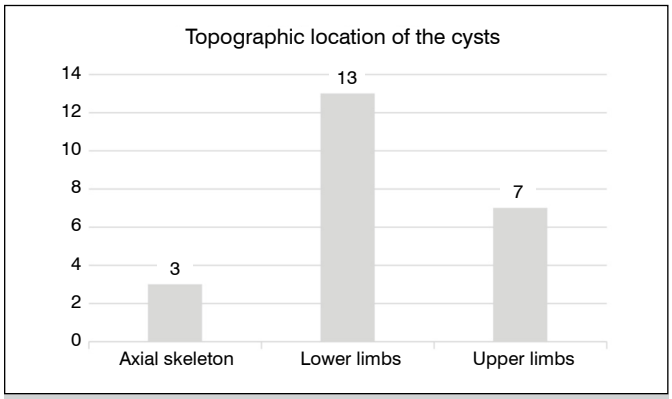

Figure 3. Graph of the distribution of the topographic location of Aneurysmal Bone Cysts: Axial skeleton (pelvis and spine); Lower limbs; Upper limbs.

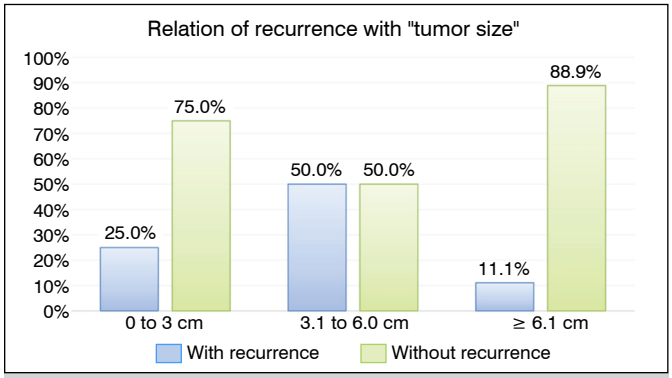

Figure 4. Bar graph of the relationship between recurrence and the qualitative factor "tumor size."

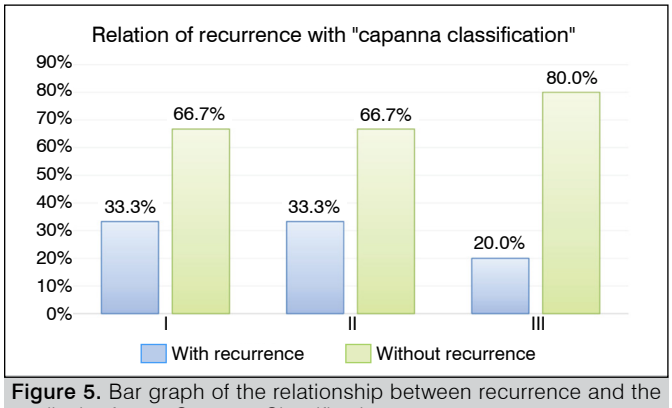

Figure 5. Bar graph of the relationship between recurrence and the qualitative factor "Capanna Classification."

the dose was 2.5 ml at a concentration of 2 mg/ml. Of the patients treated with infiltration, two (33.4%) underwent surgical treatment after recurrence.

Two patients in total (8.6%) underwent embolization for local control. Both cases were of cysts located in the spinal column, and of them, only one underwent surgery for curettage and grafting at a later time. In total, twenty (86.9%) patients underwent surgery with bone autograft. The surgical treatment of all cases consisted of intralesional resection with extensive curettage, associated with local thermal (electrocautery or Argon scalpel) or chemical (absolute alcohol) adjuvant therapy, followed by grafting in the tumor bed using autograft from the iliac crest. Figure 6 presents a comparative graph of patients who underwent surgery and those who did not, and their respective recurrence rates. Although it shows a slight

reduction in the recurrence rate among cases submitted to surgical intervention, the difference is not statistically significant.

The follow-up time was defined as the period from the first consultation in the service to the last consultation included in the study, yielding an average of 42.8±14.1 months (3 to 127 months).

The overall recurrence rate was 30.4% (n=7). We considered the cases that recurred in which there was progression of the size of the radiolucent image in the treated area in two consecutive follow-up X-rays or in which the attending physician reported, in the medical record, the diagnosis of recurrence. In the groups treated with a single method (surgery, infiltration, or embolization), the recurrence rate was 33.4%. In the group that combined the two methods, the recurrence rate among those who started treatment with surgery was 20% (1 in 5 cases). There was no recurrence in the only case initially treated with embolization and then surgery, nor in the only case treated with embolization associated with infiltration, both located in the spine.

Table 2 presents the prevalence and the bivariate analysis of the association between recurrence and the other variables mentioned. We did not identify any association between the investigated variables.

Only one patient developed a pathological fracture, 90 months after curettage and grafting. Initially, conservative treatment was attempted with a cast to perform a new curettage and grafting at a later time. The case was re-evaluated, and a new curettage and grafting, followed by internal fixation with plate and screws, was chosen after 17 days of conservative treatment. By the end of this study, the aforementioned patient is three years post-operative with satisfactory consolidation and no signs of recurrence.

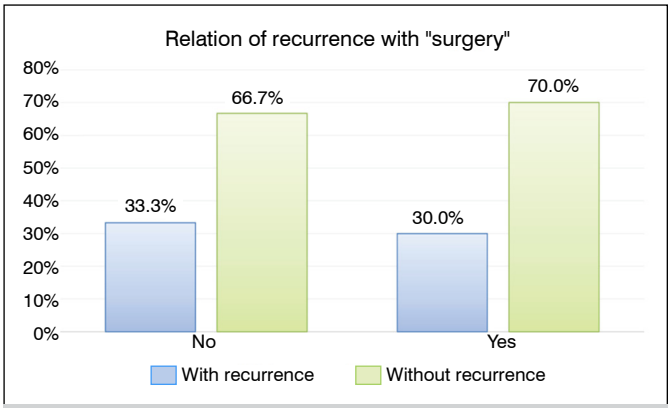

Figure 6. Bar graph of the relationship between recurrence and the qualitative factor "Surgery."

DISCUSSION

The aneurysmal bone cyst (ABC) is a benign neoplasm rich in giant cells, cystic in appearance, multiloculated, and containing blood.[2] Although current medical literature presents several works with case series on ABC,[3-7] there is a lack of studies in Brazil conducted with children and adolescents treated in specialized orthopedic oncology institutions.

In the present study, aneurysmal bone cyst was the predominant lesion in male children and adolescents. This finding differs from the data found in national[13] and international literature.[14,15] In fact, most patients diagnosed with ABC are children and adolescents.[13] ABC can occur in childhood and early adulthood, and there is a slightly increased incidence rate in women (1 to 1.3).[13,15]

In our case series, the cysts were predominantly located in the lower limbs, including the tibia, femur, talus, and metatarsus. Although ABC can occur in any bone, it usually affects the metaphysis of long bones, such as the femur and tibia.[13,15] It can also affect the vertebrae and pelvis.[13,15] The diagnosis should be confirmed by biopsy and histopathological evaluation.[15] ABC is an aggressive benign tumor that often presents with pain, pathological fracture, and may show local recurrence after treatment.[10,13,16]

In this study, most cases of ABC presented radiological classification of Capanna type II, that is, a central expansive lesion affecting the entire bone diameter, promoting cortical thinning. Another study also conducted in Brazil[14] and international studies also found in their sample a higher prevalence of Capanna type II lesions,[17,18] although in its original description, Capanna states a higher prevalence of type II.[12] We did not find in our study, among the included patients, lesions of classification IV or V.

The treatments included infiltration (calcitonin or dexamethasone + calcitonin), embolization for cases involving the spine, and surgery with graft placement. Once ABC is suspected, the patient should be referred to an orthopedic oncologist.[13] Since it is a bone lesion that may present as active with aggressive and destructive characteristics of the bone, the aneurysmal bone cyst is indicated for intervention. The most frequently performed treatment, according to the literature, is intralesional resection, with curettage of the lesion.[2,11,12]

The use of local adjuvants (phenol, absolute alcohol, electrocautery, argon scalpel) is recommended to optimize tumor cell destruction after curettage, and the affected area should subsequently be filled with bone graft (autologous or homologous) or polymethylmethacrylate (PMMA).[11] Extensive lesions and/or those involving joints may be treated with marginal or even wide resection, requiring reconstruction of the joint (endoprosthesis) or the affected bone segment.[11] Endovascular embolization of the arteries supplying the

Table 2. Relation of recurrence with the distribution of qualitative factors: sex, age, tumor size, surgery performed, and Capanna classification (n = 23).

		With Recurrence		Without Recurrence		PR (CI 95%)	P-value
		N	%	N	%		
Sex	Female	5	45.5%	6	54.5%	Ref.	0.124
	Male	2	16.7%	10	83.3%	0.37 (0.10 to 1.36)	
Age	≤ 8 years	4	44.4%	5	55.6%	Ref.	0.187
	≥ 9 years	3	21.4%	11	78.6%	0.48 (0.14 to 1.64)	
Tumor size	0 to 3 cm	1	25.0%	3	75.0%	0.50 (0.10 to 2.45)	0.336
	3.1 to 6.0 cm	5	50.0%	5	50.0%	Ref.	- x -
	≥ 6.1 cm	1	11.1%	8	88.9%	0.22 (0.04 to 1.12)	0.084
Surgery	No	1	33.3%	2	66.7%	Ref.	0.474
	Yes	6	30.0%	14	70.0%	0.90 (0.15 to 5.26)	
Capanna Classification	I	1	33.3%	2	66.7%	Ref.	- x -
	II	5	33.3%	10	66.7%	1.00 (no CI)	0.485
	III	1	20.0%	4	80.0%	0.60 (0.06 to 6.45)	0.536

N: number of individuals; PR: Prevalence Ratio; CI: confidence interval; Ref.: value set as reference for the PR study; "- x -": value for which the statistic is not applicable.

lesion can be performed, aiming to achieve local control in large or surgically difficult-to-access lesions.

In recent years, due to its predominantly osteolytic characteristics and its abundance of multinucleated giant cells resembling osteoclasts, Denosumab has been studied for the treatment of ABC. This medication works by blocking the RANKL receptor, thereby reducing osteoclast activation.[19] However, studies still do not present a consensus regarding the appropriate dose and duration of treatment.

In the present study, the recurrence rate of ABC was 30.4%. By treatment modality, we observed a recurrence rate of 33.4% in patients who received a single treatment (either surgery or infiltration only). In the combined therapy, we observed a 20% recurrence. Moreover, we found no association between recurrence rates and sex, age, or size of the cyst. The recurrence rate after ABC treatment is quite high, in accordance with its aggressive benign nature.

In the literature, recurrence rates after surgical treatment varied from 10 to 59%.[20] Vergel de Dios et al.[20] reported that 90% of recurrences occurred in patients under 20 years of age, and the authors associated recurrence with younger age. Still, the statistical analysis did not confirm this association.[20]

CONCLUSION

The observed epidemiology is consistent with pediatric cohorts reported in the literature. Both surgery and calcitonin infiltration are appropriate treatment options for aneurysmal bone cysts, and embolization can be employed concomitantly or independently of these methods to achieve local control of lesions in surgically difficult-to-access locations. There was no statistically significant relationship between recurrence and the evaluated risk factors, as the value 1.0 was always within the Prevalence Ratio interval.

CONTRIBUTIONS OF THE AUTHORS

Each author significantly contributed to the development of the article. CFG: article composition, review, acquisition of epidemiological data, and statistical analysis; NSPO and JGG: review, collection of epidemiological data on the platform, article composition; MTP, Filho, and RJGF: review, collection of epidemiological data on the platform; DCMV: intellectual conception of the work, article review, collection of epidemiological data on the platform.

DATA AVAILABILITY DECLARATION

The underlying data related to the research are available with the corresponding author.

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DOI: http://dx.doi.org/10.1590/1413-785220263401e292923

NEGATIVE PRESSURE WOUND THERAPY AS AN ADJUNCT TO SKIN GRAFTING IN TRAUMATIC INJURIES

TERAPIA POR PRESSÃO NEGATIVA COMO ADJUVANTE AOS ENXERTOS DE PELE EM LESÕES TRAUMÁTICAS

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ABSTRACT

Introduction: Wounds are a challenge for health professionals and services, prolonging hospital stays and increasing morbidity and mortality rates. Wounds occur due to post-traumatic tissue loss, which is often difficult to resolve and is complicated by infections, local vascular impairment, and systemic diseases. To demonstrate the effectiveness of negative pressure therapy (NPT) using the vacuum gauge of the hospital gas pipeline and low-cost materials in the immediate postoperative period. To use resources available in daily hospital practice to create a low-cost vacuum dressing. To describe the equipment, preparation, and technique involved in low-cost NPT treatment as an adjuvant in the repair of lesions with indication for skin grafting. Methodology: Prospective, interventional, randomized clinical study with a non-inferiority design. Results: Fifteen patients were included at the end of the study, with complete follow-up until complete healing. The mean age of the selected patients was 38.27 years. Regarding gender, 93.3% were male and 6.7% were female. Conclusion: Thus, the results suggested that the use of low-cost NPT on skin grafts is a valuable, safe, easily reproducible and low-morbidity tool for surgeons technical arsenal. ***Level of Evidence: IV; Case Series.***

Keywords: Skin Transplantation; Patient Isolators; Low Cost Technology; Wounds and Injuries.

RESUMO

Introdução: As feridas constituem um desafio aos profissionais e serviços de saúde, prolongam o tempo de hospitalização e aumentam a taxa de morbimortalidade. As feridas ocorrem devido perdas teciduais pós trauma, muitas vezes de difícil resolução, complicadas por infecções, comprometimento vascular local, doenças sistêmicas. Objetivos: Demonstrar a efetividade do uso da terapia por pressão negativa (TPN) utilizando o vacuômetro da régua de gases hospitalares e materiais de baixo custo no pós-operatório imediato. Utilizar recursos disponíveis no cotidiano hospitalar para confeccionar um curativo a vácuo de baixo custo. Descrever os equipamentos, a preparação e a técnica envolvidos no tratamento de TPN de baixo custo como adjuvante na reparação de lesões com indicação de enxertia de pele. Metodologia: Estudo clínico prospectivo, intervencionista, randomizado com desenho de não inferioridade. Resultados: Foram incluídos 15 pacientes ao final do estudo, com acompanhamento completo até a cicatrização total. A média de idade dos pacientes selecionados foi de 38,27 anos. Quanto ao sexo tivemos 93,3% masculino e 6,7% feminino. Conclusão: Assim, os resultados sugerem que o uso da TPN de baixo custo sobre enxertos de pele constitui uma ferramenta de grande valia, segura, de fácil reprodutibilidade e associada a baixa morbidade no arsenal técnico dos cirurgiões. ***Nível de Evidência: IV; Série de Casos.***

Descritores: *Enxertia de Pele; Isoladores de Pacientes; Tecnologia de Baixo Custo; Ferimentos e Lesões.*

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INTRODUCTION

The wounds constitute a challenge for health professionals and services, prolonging hospitalization time and increasing morbidity and mortality rates. Wounds occur due to tissue loss post-trauma, often difficult to resolve, complicated by infections, local vascular compromise, systemic diseases.[1]

In this study, we emphasize the use of skin grafts associated with negative pressure therapy (NPT). Apelqvist et al.[2] describe that NPT has the ability to adapt to the contours of the wound bed, promoting the approximation of the edges and retraction of the lesion, mechanical stress on the wound edges, improving microcirculation and tissue perfusion, has an anti-edematous effect, leading to

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angiogenesis and granulation tissue formation and reduction of inflammatory exudate.

This method also provides the same benefits to surgical wounds, favoring proper and aseptic closure over an extended period, without the need for frequent interventions.[2]

The skin graft is the transfer of skin tissue between areas of the body, does not carry a vascularized pedicle and depends on an adequate recipient bed. After incorporation, skin grafts provide wounds with protection against the environment, pathogens, temperature, and excessive water loss like normal skin.[3]

Grafts may present some short-term complications: seroma, hematoma, infection, shearing or traction; and long-term: contracture, aesthetic problems, pigment and texture differences between the grafted skin and the donor site.[3]

According to the surveys by Lima et al.[1], the integration of NPT after grafting was significantly greater when compared to conventional dressings, reducing the risks of complications.

In addition to better graft integration, NPT reduces hematoma formation, promotes a shear-free environment, and optimizes integration. In these cases, NPT should always be administered in continuous mode.[1]

NPT has been approved since 1997 by the *Food and Drug Administration* (FDA) in the United States. In 2003, NPT was introduced in Brazil, and in 2008, the simplified vacuum dressing with national technology was registered by the University of São Paulo (USP).[4,5]

The study by Kamamoto,[4] shows us that the costs related to NPT can be quite high. Through the method developed by USP, in which a valve was designed that, when connected to the hospital gas network, can control the pressure exerted on the sealed dressing, using gauze on the wound bed and sealing film. The result of the work was able to counter the cost-effectiveness of the similar method compared to the commercial method regarding the stimulation of healing (effectiveness), in addition to having an average cost of $15.15 compared to $872.59 spent on the commercial method. According to the authors, the dressing model developed by USP, called "adapted low cost", did not show lower efficiency compared to the commercial "gold standard" system to which it was compared.[4]

This work is justified by the need for discussion and implementation of effective and safe systems that reduce the disturbances caused by traumatic injuries. Also, the promotion of new studies for reflection on such a relevant topic for public health. In this context, it is important that the health team develops wound repair techniques that optimize the healing of devitalized tissues.

OBJECTIVES

Demonstrate the effectiveness of using TPN with the vacuum meter of the hospital gas gauge and low-cost materials in the immediate postoperative period of definitive closure of skin grafting in patients due to trauma.

To use available resources in the hospital routine to create a low-cost vacuum dressing.

To describe the equipment, preparation, and technique involved in low-cost NPT treatment as an adjunct in the repair of injuries with indications for skin grafting.

METHODOLOGY

Prospective, interventional, randomized clinical study with a non-inferiority design. After approval from the Ethics Research Committee of the ABC School of Medicine under number: 7,187,630, the selected patients signed the Informed Consent Form.

This study was conducted in a high-complexity State Teaching Hospital in the Greater ABC region, in the state of São Paulo. The population consisted of patients with upper or lower limb injuries with tissue loss sufficient to prevent primary closure of the injuries, subjected to soft tissue injury coverage with skin grafts.

Inclusion criteria were patients over 18 years old, victims of high-energy trauma with wounds requiring skin grafting. Exclusion criteria were coagulopathies, uncontrolled active bleeding, and refusal to participate.

The interventional procedures performed were wound treatment using skin grafts and at the same surgical time the application of vacuum dressing with cellulose acetate (Rayon) over the wound bed, sterile surgical dressing, 16 French nasogastric tube, sealed with a sterile transparent polyurethane film dressing connected to the secretion collection system with a backflow valve linked to the hospital vacuum meter with continuous pressure regulated to 100 to 120 mmHg. (Figure 1)

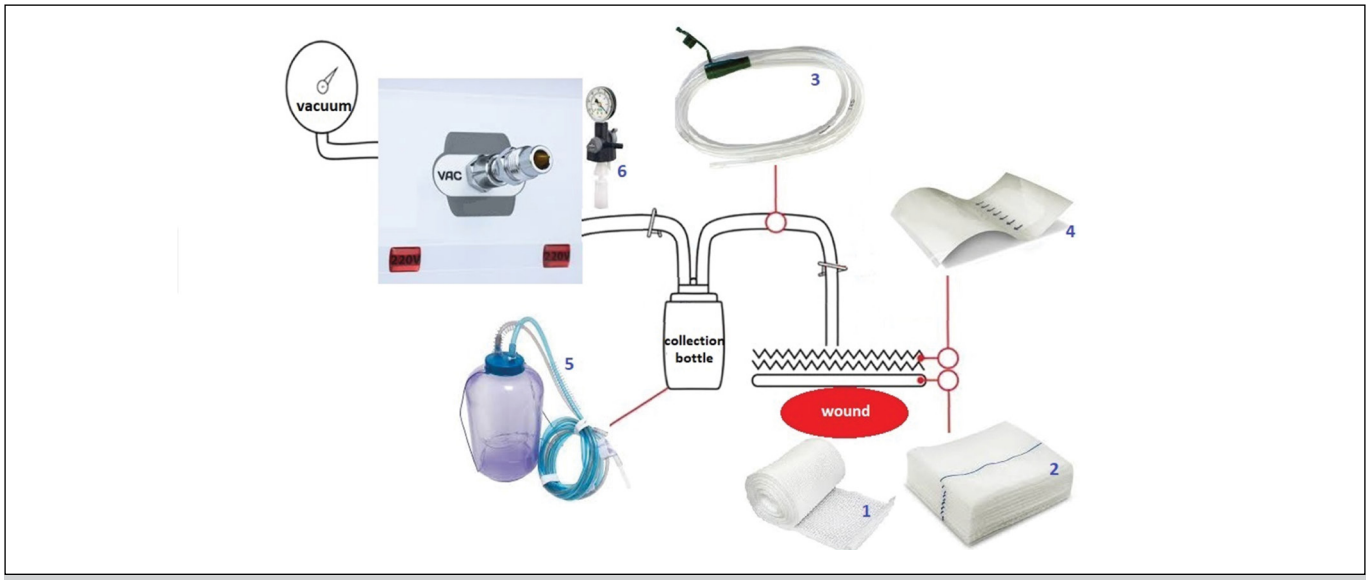

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Figure 1. 1: Gaze Rayon, 2: Dressing; 3: Gastric Tube Levine N 16, 4: Transparent adhesive film, 5: Suction bottle and filter 6: manometer (pneumatic), vacuum dressing model.

The dressing was monitored in the ward during hospitalization to assess the pressure on the vacuum meter, bleeding, or detachment of the dressing due to shear or friction, as well as hematoma of the covered area. The dressing was removed for evaluation between the 5th and 7th day, a period in which the phases of graft take, plasma imbibition, inosculation, and revascularization normally occur. Partial thickness skin grafts are typically adherent after 5 to 7 days with the advancement of the wound healing stages. (Figure 2)[3] After removal of the dressing with TPN, the graft was monitored weekly until complete healing. A questionnaire was developed for data collection with basic information on cause, type of trauma, age, as well as the assessment of the composition of the wound area post-graft, the percentage of tissue where shear, hematoma, or necrosis occurred at the time of vacuum removal. These factors were evaluated in a single stage by the same team. Since they are factors responsible for the failure of skin grafts and delay in the healing process. Data collection was performed during the postoperative follow-up.

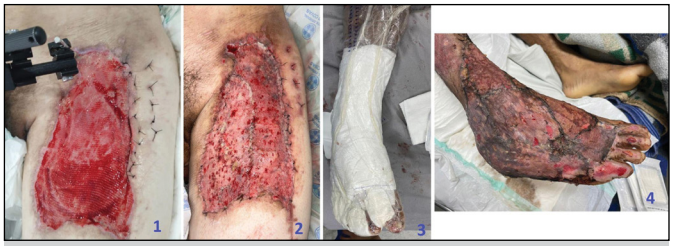

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Figure 2. 1: Morel-Lavallée injury post vacuum dressing 2: Morel-Lavallée injury post partial skin graft and vacuum. 3: Vacuum dressing mounted 4: Injury due to degloving (Partial skin graft + vacuum dressing after 5 days of procedure).

RESULTS

A total of 15 patients were included by the end of the study, with complete follow-up until total healing. The average age of the selected patients was 38.27 years. Regarding sex, we had 93.3% male and 6.7% female.

Only 26.7% of patients had pre-existing comorbidities. Of which only 1 (6.7%) had type 1 diabetes *mellitus*, which can hinder the healing process. The others presented hypertension, asthma, and schizophrenia.

Of the selected patients, 33.3% had active addictions at admission, with the most cited being the use of illicit drugs such as marijuana and cocaine, in addition to legal drugs like alcohol and tobacco.

In Table 1 we can observe that the main causes are traffic accidents with 60% of cases of major trauma with skin loss, requiring grafting. In addition to the location of these injuries, with 76.55% in the lower limbs.

Scope of the propositions and goals of the study demonstrating the applicability of low-cost TPN on skin grafting using the hospital gas meter vacuum, associated with routine hospital materials. Validation of the technique of using the vacuum meter and low-cost materials in the immediate postoperative of definitive closure of skin grafting in patients resulting from trauma.

Regarding the complication rate of grafts, we had a variance of 0 to 40%. The average hematoma was 15.73%, shear 8.07%, and necrosis 2.6%. It can be verified that in the use of TPN the lowest observed rate was necrosis. (Figure 3)

Regarding size, we had an average of 13.6 cm in length and 12.8 cm in width of the lesions, with the largest size being 38 cm and 33 cm respectively. At the moment we compared the size of the lesions with the percentage complication index, we noticed that

Table 1. Type of trauma and location of the injury.

Type of Trauma		
	N	%
Accidents Motorcycle/Car/Truck/Train	9	60
Tumor	2	13.3
Runover	3	20
Injury + Osteomyelitis	1	6.7
Total	5	100
Location of the injury		
	N	%
Thigh	1	6.7
Leg	3	20
Leg and foot	3	20
Forearm	1	6.7
Arm	1	6.7
Hand	1	6.7
Foot	2	13.3
Popliteal fossa	1	6.7
Thigh, Knee, and Popliteal fossa	1	6.7
Thigh + Forearm	1	6.7
Total	15	100

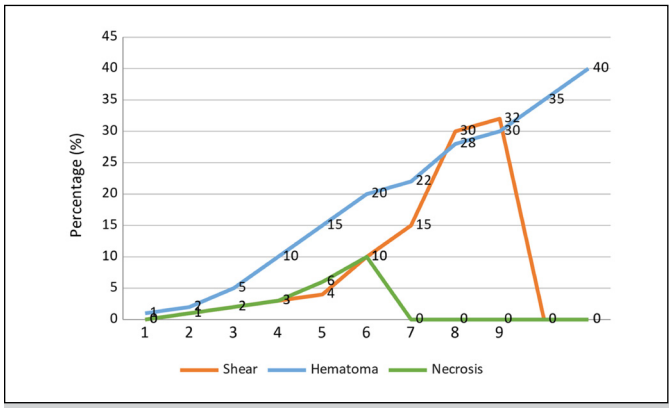

Figure 3. Size of the Lesion versus percentage of Shearing, Hematoma, and Necrosis.

the highest rates of hematoma, shearing, and necrosis occurred in patients with the largest area of skin loss and consequently with the most grafts performed.

DISCUSSION

The average age of the patients was 38.27 years. The most affected age group is from 20 to 59 years, representing about 70% of deaths from traffic accidents. Young people between 21 and 30 years are particularly vulnerable due to greater exposure and risk behaviors in traffic, such as high speed and driving under the influence of alcohol.[6]

Men are the main victims, representing about 81% of deaths, corroborating our data. This predominance is attributed to risk behaviors, such as a higher frequency of aggressive driving and alcohol consumption before driving.[6]

The study data revealed that the lower limbs were more affected. The predominant injuries include trauma to the limbs, such as fractures and contusions, which are common among survivors.[6]

The cost of hospitalizations and productivity losses due to traffic accidents is estimated in billions of reais annually, highlighting the economic and social relevance of the problem.[6]

In the study by Souza et al.[7] that evaluated the financial viability of TPN, participants were divided into two groups: wound treatment with the simplified vacuum dressing model (MCVS), which is the TPN model of our study, and the other group with hydrofiber treatment with silver (HFP). The participants in the study were mostly men with an average age in the 6th decade. Regarding age, the average in our study was 38.27 years.

In the study by Kamamoto,[4] which is the precursor of low-cost TPN in Brazil, participants were also divided into two groups: TPN USP group and VAC (*Vacuum Assisted Closure*). Regarding trauma, the TPN group (94%) and VAC group (84%) had traffic accidents as the main cause, representing 60% of cases in our study. Regarding treatment costs, the TPN group had an average cost of R$47.89 and the VAC R$2,757.40. The VAC system uses polyurethane foam while TPN, both from USP and ours, uses rayon gauze and compress, as it is sterile, low-cost, highly available in health services, and less painful when changing the dressing. Regarding wound healing, satisfaction with the results comes from faster regeneration, with lower risks of contamination and complete closure of the lesion in less time compared to the VAC group.

According to the reviews by Santos et al.[8] and Lima et al.[1], TPN helps create a conducive environment for healing by stimulating granulation tissue and perfusion, as well as reducing edema and removing exudate and infectious material. In the postoperative period, it promotes tissue formation after debridement and reinforces skin grafts, favoring moisture balance, advancing the epithelial edge, and the rate of graft non-integration is lower when TPN is used. In line with this, TPN becomes an ally in graft integration.

According to Scalise et al.[9], hematomas and seromas are complications resulting from the accumulation of blood and serum, respectively, in internal spaces. Even with excellent surgical technique, bleeding and inflammation can occur, and consequently, serum leakage increases the likelihood of infection, slower healing, additional clinical visits, and surgical interventions. In the study using low-cost vacuum dressings, even though the average hematoma rate was 15.73%, it did not contribute to loss or the need for new interventions on the graft.

Condé-Green et al.[10] in their study compared the treatment of open abdominal wounds with traditional dressings and negative pressure therapy, which showed rates of skin and fat necrosis of 9% with TPN and 18% with traditional dressing, respectively.

The percentage of necrosis of the partial skin graft was evaluated in our study and presented a percentage of 2.6%. The low index correlates with the aforementioned study.

Alves et al.[11] highlight that the most frequent causes of graft loss are the presence of hematoma, which mechanically separates the graft from its bed, and shear movements, which prevent graft adhesion, both hindering vascularization. Shear occurred in 8.07% of cases treated with graft plus negative pressure therapy.

For Webster et al.[12], graft loss rates may be lower when negative pressure therapy is used. Products designed and built in hospitals are as effective in this area as commercial applications. There are clear cost benefits when non-commercial systems are used to create the negative pressure needed for wound therapy, with no evidence of a negative effect on clinical outcomes.

It was observed that, with the increase in the size of the wound area, shear rates, hematoma, and necrosis also increased. This fact can be explained by the difficulty of maintaining a homogeneous and continuous pressure over large graft areas.

More than half of the research participants were victims of automobile accidents, which generate multiple injuries, both soft tissue and bone, with indications for external fixation to control damage.

During the application of TPN on grafts, one of the difficulties was creating a vacuum due to air entering the peri-fixator system. Thus, as described by Lima et al.[1], it is necessary to cut the adhesive film dressing into smaller fragments to achieve sealing and correct vacuum loss, which can be assessed by listening for escape points.

The article by Gao et al.[13] reinforces the data presented above. The authors demonstrated in their article that the healing time of the analyzed wounds was shorter compared to the control group, with a significantly lower healing rate.

CONCLUSION

This study highlighted the reduction of hematoma, necrosis, and shear. It denoted the versatility and effectiveness of vacuum dressings for various wound sizes. It enabled the implementation of an effective and safe system that reduced the complications caused by traumatic wounds. In this context, the development of a wound repair technique that optimizes the healing of devitalized tissues was important.

It described available resources in everyday hospital settings and low-cost options to create a vacuum dressing aimed at reducing costs associated with the treatment of soft tissue injuries, in addition to being an adjunct in the repair of injuries with indications for skin grafting.

Thus, the results suggested that the use of low-cost TPN on skin grafts is a highly valuable tool, safe, easy to reproduce, and with low morbidity to the surgeons' technique arsenal. It contributed to improving the adhesion of grafts to the recipient bed and subsequent healing of complex injuries that require skin coverage, often leading to prolonged hospitalization and difficult therapeutic management.

CONTRIBUTIONS OF THE AUTHORS

Each author contributed individually and significantly to the development of this article. CCR: writing, data analysis, data interpretation; RLF: data analysis and compilation; LS: critical review of the article; ABC: critical review of the article; CHVF: critical review of the article; LYK: critical review of the article; ERAS: support in placing vacuum dressings; CRS: support in placing vacuum dressings, review of collected data, initial writing.

DATA AVAILABILITY DECLARATION

The underlying contents of the research are available in the manuscript.

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PATIENTS WITH SOFT TISSUE SARCOMA AFTER TREATMENT BY NON ORTHOPEDIC ONCOLOGIC SURGEONS: EPIDEMIOLOGICAL PROFILE, STAGING, AND THERAPEUTIC CHALLENGES

PACIENTES PORTADORES DE SARCOMA DE PARTES MOLES, PÓS-TRATAMENTO COM MÉDICOS NÃO ONCOLOGISTAS ORTOPÉDICOS: PERFIL EPIDEMIOLÓGICO, ESTADIAMENTO E DESAFIO TERAPÊUTICO

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ABSTRACT

Objective: this study analyzed patients with soft tissue sarcoma treated by non-orthopedic oncologists, evaluating their epidemiological profile, staging, and therapeutic challenges. Method: an analytical study in the form of a retrospective cohort, conducted through a review of medical records of patients treated at the hospital from January 1, 2011, to December 31, 2021. Results: a total of 61 patients were included, mostly male (55.7%), with a mean age of 42.8 years. The most frequent histological subtypes were synovial sarcoma (29.5%) and undifferentiated pleomorphic sarcoma (21.3%), with a predominance of high-grade tumors (75.4%). The majority of cases (77%) underwent resection, but without proper planning, leading to high recurrence rates (77%) and metastases (49.2%), with the lungs being the primary metastatic site. The mortality rate was 47.5%, with an average time to death of 3.1 years. Conclusion: the findings highlight the need for early diagnosis, specialized treatment, and multidisciplinary management to optimize clinical outcomes for patients. The adoption of standardized protocols in referral centers may reduce inadequate interventions and improve survival and quality of life for patients with soft tissue sarcoma. ***Level of Evidence III; Retrospective[f] comparative study[e].***

Keywords: Epidemiology; Neoplasm Staging; Bone Neoplasms; Sarcoma.

RESUMO

Objetivo: este estudo analisou pacientes com sarcoma de partes moles tratados por médicos não ortopedistas oncologistas, avaliando perfil epidemiológico, estadiamento e desafios terapêuticos. Método: estudo analítico em forma de coorte retrospectiva, realizado através de revisão de prontuários dos pacientes atendidos no hospital no período de 1 de janeiro de 2011 até 31 de dezembro de 2021. Resultado: foram incluídos 61 pacientes, majoritariamente homens (55,7%), com média de idade de 42,8 anos. Os subtipos histológicos mais frequentes foram sarcoma sinovial (29,5%) e sarcoma pleomórfico indiferenciado (21,3%), com predomínio de tumores de alto grau (75,4%). A maioria dos casos (77%) foi submetida à ressecção, mas sem planejamento adequado, resultando em elevadas taxas de recorrência (77%) e metástases (49,2%), sendo o pulmão o principal sítio metastático. O índice de mortalidade foi de 47,5%, com média de 3,1 anos para o óbito. Conclusão: os achados destacam a necessidade de diagnóstico precoce, tratamento especializado e manejo multidisciplinar para otimizar os desfechos clínicos dos pacientes. A adoção de protocolos padronizados em centros de referência pode minimizar intervenções inadequadas e melhorar a sobrevida e qualidade de vida dos pacientes com sarcoma de partes moles. ***Nível de Evidência III; Estudo retrospectivo[f] comparativo[e].***

Descritores: *Epidemiologia; Estadiamento de Neoplasias; Neoplasias Ósseas; Sarcoma.*

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<< SUMÁRIO

INTRODUCTION

Sarcomas are defined as a heterogeneous group of malignancies of mesenchymal origin, which can be divided into osteosarcomas, originating from bone tissue, and soft parts sarcoma (SPS), which arise from connective tissues. SPSs present more than 80 histological subtypes, with varying clinical presentations, leading to a difficult diagnosis.[1]

From an epidemiological point of view, SPSs have an incidence considered rare, representing about 1% of cancers in adult patients and 15% of pediatric cancers. However, it is believed that these data are underestimated, due to the pathogenesis not yet fully clarified and its challenging anatomopathological findings.[2]

Although there is no clearly defined pathogenesis, some risk factors have already been identified, such as genetic predisposition, immunodeficiency, lymphedema, and history of exposure to oncogenic viruses, radiation, chemotherapy and/or chemical carcinogens. In addition, certain genetic syndromes have a well-characterized association with SPS, such as Li-Fraumeni syndrome, Bloom syndrome, Beckwith-Wiedemann syndrome, hereditary retinoblastoma and neurofibromatosis type 1.[3]

The anatomical location of the disease is a significant variable, with great influence on the patient's treatment and prognosis. The upper and lower extremities account for 43% of cases, corresponding to the most common primary site. However, tumors can develop anywhere in the body, such as the viscera (19%), retroperitoneum (15%), trunk (10%), and head and neck (9%).[4]

SPSs can occur at any time in the patient's life, with histological variations according to the age group in which their evolution occurs. When all ages are considered, the most common forms include: liposarcoma; indifferent pleomorphic sarcoma, sinovial sarcoma and leiomiosarcoma. In the elderly, mixfibrosarcoma also gains importance. In childhood, rabdomiosarcoma is the main representative, causing 50% of cases.[5]

Despite the presence of small variations according to each histological subtype, the main clinical form associated with SPS is the emergence of a painless nodular lesion, which can last for weeks or months. Less commonly, they present pain, pathological fractures, or are discovered incidentally. This non-specific picture contributes to the difficulty of making the proper diagnosis.[6]

Some characteristics associated with the tumor may indicate to the responsible doctor the need for a more urgent investigation. The existence of a painless mass with progressive increase in size is the main alarm sign for an increased risk of malignancy. In addition, lesions with a length greater than 5 centimeters, or of more hardened consistency than the surrounding tissue, or of deep location relative to the fascia, require more cautious investigation.[7]

Histopathological diagnosis remains the gold standard for identification of the subtype and proper conduct of the case. However, it is worth noting that differentiating between malignant and benign conditions can be extremely complicated, requiring experienced pathologists for a reliable judgment.[8]

The complete staging includes imaging of the primary tumor and verification of the presence of metastatic disease. Magnetic resonance imaging is the modality of choice to define the size of the tumor, as well as its muscular involvement and the relationship with neurovascular structures. Considering that the most common places of metastasis appearance are the lungs, thoracic CT gains importance for confirmation or discard.[9]

The main method of treatment for SPS is surgical, with extensive resection, removing enough tissue to reach macroscopically negative margins, with the objectives of preventing local recurrence and preserving limb function. In the case of highly infiltrative tumors, radiation therapy, neoadjuvant or adjuvant, may be necessary for better control of the margins. Adjuvant chemotherapy gains importance for therapeutic aid in metastatic diseases.[10]

The main risk of death related to SPSs of extremities is the occurrence of metastases. The most relevant prognostic factors for overall survival are: age of the patient; tumor dimensions; degree of malignancy; histological subtype; and depth. In addition, local recurrence is a constant danger associated with SPS, contributing to a worse patient outcome.[11]

Therefore, considering the complexity involving the different pathological forms of SPS, it is crucial to recognize the importance of managing such cases in specialized units. Driving by experienced teams with the help of appropriate technologies is linked to greater overall survival of patients, as well as higher rates of surgeries with negative margins and ease of receiving adjuvant therapy.[12]

METHODOLOGY

This study is descriptive, retrospective and observational. The research was conducted in the oncological orthopedic sector of Ophir Loyola Hospital, linked to the Government of the State of Pará, located in the Municipality of Belém, State of Pará.

The target population of this study consists of patients diagnosed with soft part sarcoma and treated by non-orthopedic oncologists, who are subsequently referred to Ophir Loyola Hospital for specialized treatment in the sector of orthopedic oncology.

The sample of this study was carefully selected to adequately represent the target population. Patients registered in the Ophir Loyola Hospital system were selected between 2011 and 2021. The sample size was determined considering the availability of eligible patients and the available resources for data collection and analysis, totalling 61 patients selected.

It was considered as inclusion criteria to have a confirmed histopathological diagnosis of soft-part sarcoma, with prior treatment by non-orthopedic oncologists, aged over 12 years and availability of medical records.

The exclusion criteria consisted of: patients who received treatment exclusively by oncological orthopedists, without a confirmed histopathological diagnosis of soft-part sarcoma, who were younger than 12 years of age and/or unavailable or insufficient medical records.

The data collection for this study was conducted in a systematic and comprehensive manner, in accordance with ethical protocols and data privacy regulations, ensuring the confidentiality and anonymity of patients.

The data was obtained by reviewing the medical records of the patients included in the study to collect detailed information about demographic profile, medical history, imaging examinations, tumor characteristics, received treatments and clinical outcomes.

The data analysis for this study was conducted in a thorough and rigorous manner, using statistical and epidemiological methods suitable for each specific purpose of the research.

Initially, the evaluation of the studied population was carried out, with identification of the epidemiological profile, staging and corresponding therapeutic challenges. These data were tabulated in a spreadsheet elaborated in the Microsoft® Office Excel® 2016 software, going through descriptive statistical analysis of the sample characterization, with frequency, percentages, average, standard deviation, median, interquartile interval (p25%-p75%), exposed in tables and/or graphs. The continuous quantitative variables, such as age (years) and time (years), were first submitted to the Shapiro-Wilk test to analyse their normality distribution. All statistical analysis was performed in SPSS 20.0 software.

This research was approved by the Ethics Committee of the Institution, through the Brazil Platform, with identification CAEE 81512924.5.0000.5550 (amendment no. 7.059.611). The collection of records in a retrospective manner was preceded by a

Data Usage Commitment Terms, to ensure the reliability of the collected information, and the Request for waiver of the Free and Informed Consent Terms.

RESULTS

The study included 61 subjects, mostly male (55.7%), with an average age of 42.8 (±19.4) years, brown (62.3%), 34.2% with some comorbidity, among the most frequent hypertension (29.5%) and diabetes (23.0%), as can be seen in Table 1.

Table 2 presents the clinical profile of 61 patients with soft-part sarcoma treated by non-orthopedic oncologists. In terms of anatomical location, the thigh was the most affected region, representing 52.5% of the cases (IC95%: 41.0 – 65.6). Other locations, such as leg (11.5%; IC95%): 4.9 - 19.7), hand and forearm (9.8% each; 95% CI: 3.3 – 18.0), arm (8.2%; IC95%: 1.6 - 14.8), foot (6.6%; 95% CI: 1.6 - 13.1) and handle (1.6%; IC95%: 0.0 – 4.9), were less common. The most common histological subtypes were sinovial sarcoma, with 29.5% of cases (95% CI: 18.0 – 42.6), and indifferent pleomorphic sarcoma, with 21.3% (CI95%: 11.5 – 31.1). Other subtypes, such as liposarcoma and epithelial sarcoma (9.8% each), rabdomiosarcoma and leiomiosarcoma (8.2% each), as well as less prevalent types, such as angiosarcoma, dermatofibrossarcoma protuberans, fibrosarcoma, mixfibrosarcoma, neurofibrosarcoma and alveolar sarcoma (1.6% to 3.3%), presented a lower incidence.

As for the degree of sarcoma, 75.4% of cases were of high degree (CI95%: 64.0 – 85.2), while low and moderate degrees were observed in 14.8% and 9.8% of patients, respectively. Regarding the procedures performed, the majority of patients (77%; 95% CI): 65.6 – 86.9) was subjected to resection, while biopsy was performed in 23% of cases (CI95%: 13.1 – 34.4).

When analysing the treatments performed by Ophir Loyola Hospital for patients including, it was observed that these patients began follow-up in the reference unit about 1.1 (±2.2) years after the diagnosis of the disease, and the vast majority underwent chemotherapy (77.0%) and radiotherapy (60.7%), as evidenced in Table 3.

Table 4 presents a detailed analysis of recurrence, metastases and deaths among 61 patients with soft-part sarcoma previously treated by non-orthopedic oncologists. Regarding recurrence, 77% of patients had recurrence (CI95%: 67.2 - 86.9), with an average recurrence time of 1.7 years (±1.2; 95% CI: 1.4 – 2.0). Only 23% of patients do not had recurrence (CI95%: 13.1 – 32.8).

Table 1. Demographic and epidemiological profile of patients with soft-part sarcoma treated by non-orthopedic oncologists.

Variables	Frequency (n. 61)	Percentage (%)	CI95%
Sex			
Male	34	55.7	44.3 - 67.2
Female	27	44.3	32.8 - 55.7
Age (years)			
Average (SD)	42.8 (19.4)		38.1 - 48.0
Median (p25%-75%)	36.0 (28.0 - 60.0)		33.0 - 46.0
Ethnicity			
Black	15	24.6	13.1 - 36.0
Pardo	38	62.3	49.2 - 75.4
White	8	13.1	4.9 - 23.0
Comorbidities			
Hypertension	18	29.5	19.7 - 41.0
Diabetes	14	23.0	13.1 - 34.4
Colelithiasis	2	3.3	0.0 - 8.2
Benign prostatic hyperplasia	2	3.3	0.0 - 8.2
Encephalic Vascular Accident	1	1.6	0.0 - 4.9

CI. Confidence interval. SD. standard deviation. p. percentile.

Table 2. Clinical profile of patients with soft part sarcoma treated by non-orthopedic oncologists.

Variables	Frequency (n. 61)	Percentage (%)	CI95%
Anatomic Localization			
Arm	5	8.2	1.6 - 14.8
Frontarm	6	9.8	3.3 - 18.0
Hand	1	1.6	0.0 - 4.9
Hand	6	9.8	3.3 - 18.0
Butt	32	52.5	41.0 - 65.6
Leg	7	11.5	4.9 - 19.7
Foot	4	6.6	1.6 - 13.1
Histological subtype			
Angiossarcoma	1	1.6	0.0 - 4.9
Dermatofibrosarcoma protuberans	2	3.3	0.0 - 8.2
Fibrossarcoma	2	3.3	0.0 - 8.2
Leiomiossarcoma	5	8.2	1.6 - 16.4
Liposarcoma	6	9.8	3.3 - 18.0
Mixfibrossarcoma	1	1.6	0.0 - 4.9
Neurofibrossarcoma	1	1.6	0.0 - 4.9
Rabdomiossarcoma	5	8.2	1.6 - 14.8
Sarcoma alveolar	1	1.6	0.0 - 4.9
Epithelial sarcoma	6	9.8	3.3 - 18.0
Undifferentiated pleomorphic sarcoma	13	21.3	11.5 - 31.1
Sinovial sarcoma	18	29.5	18.0 - 42.6
Grade			
Low	9	14.8	64.0 - 85.2
Moderate	6	9.8	3.3 - 18.0
High	46	75.4	64.0 - 85.2
Procedures			
Resection	47	77.0	65.6 - 86.9
Biopsia	14	23.0	13.1 - 34.4

CI. Confidence interval.

Table 3. Treatment carried out by Ophir Loyola Hospital of patients with soft-part sarcoma treated by previous non-orthopedic oncologists.

Variables	Frequency (n. 61)	Percentage (%)	CI95%
Time to start the treatment			
Average (SD)	1.1 (2.2)		0.5 - 1.6
Treatment performed			
Resection	29	47.5	34.4 - 59.0
Amputation	23	37.7	26.2 - 50.8
Chemotherapy	47	77.0	67.2 - 86.9
Radioterapia	37	60.7	49.2 - 72.1

CI. Confidence interval. dp. standard deviation.

As for metastases, 49.2% of patients developed metastases (95% CI: 36.1 – 62.3), with an average time to appearance of 2.4 years (±1.6; 95% CI: 1.9 – 3.0). The lung was the most common site of metastases, affecting 41% of cases (IC95%: 27.9 – 54.1), followed by the spine (13.1%; 95% CI: 4.9 – 22.9), skull (8.2%; IC95%: 1.6 – 16.4), inguinal lymph nodes and liver (6.6% each; 95% CI: 1.5 – 13.1). Other localizations, such as axillary, coastal and adrenal lymph nodes, were less common, ranging from 1.6% to 3.3%.

On deaths, 47.5% of patients died (IC95%: 34.4 – 60.7), while 52.5% survived (IC95%: 39.3 – 65.6). Among deaths, 9.8% occurred prior to treatment (IC95%: 3.3 – 18.0). The mean time to death was 3.1 years (±1.8; 95% CI: 2.4 – 3.7).

Table 4. Analysis of recurrence, metastases and deaths of patients with soft part sarcoma treated by previously non-orthopedic oncologists.

Variables	Frequency (n. 61)	Percentage (%)	CI95%
Recurrence			
Yes	47	77.0	67.2 - 86.9
No.	14	23.0	13.1 - 32.8
Time for Recurrence (years)	1.7 (1.2)		1.4 - 2.0
Metastasis			
Yes	30	49.2	36.1 - 62.3
No.	31	50.8	37.7 - 63.9
Time for Metastasis (years)	2.4 (1.6)		1.9 - 3.0
Sites of metastases			
Lung	25	41.0	27.9 - 54.1
Vertebral Spine	8	13.1	4.9 - 22.9
Axilar lymph nodes	2	3.3	0.0 - 8.2
English lymph nodes	4	6.6	1.5 - 13.1
Cranium	5	8.2	1.6 - 16.4
Liver	4	6.6	1.5 - 13.1
Costa Arch	1	1.6	0.0 - 4.9
Adrenal	1	1.6	0.0 - 4.9
Death			
Yes	29	47.5	34.4 - 60.7
No.	32	52.5	39.3 - 65.6
Pre-Treatment Death	6	9.8	3.3 - 18.0
Time for Death (years)	3.1 (1.8)		2.4 - 3.7

CI. Confidence interval. SD. standard deviation.

In general, the results reveal that patients with soft-part sarcoma treated by non-orthopedic oncologists, the majority were male (55.7%), with an average age of 42.8 years, predominating brown (62.3%). The most common comorbidities were hypertension (29.5%) and diabetes (23.0%). The thigh was the most frequent anatomical location (52.5%), followed by leg (11.5%) and forearm/hand (9.8% each). The main histological subtypes were sinovial sarcoma (29.5%) and indifferent pleomorphic sarcoma (21.3%). Most tumors were of high degree (75.4%) and were treated with resection (77%). Metastases occurred in 49.2% of cases, with the lungs being the main site (41%). Recurrence was observed in 77%, with an average time span of 1.7 years. Deaths occurred in 47.5%, with an average of 3.1 years to the outcome.

DISCUSSION

The epidemiological profile of soft-part sarcoma indicates that this is a rare condition, corresponding to approximately 1% of all cancers. The prevalence is slightly higher among men than women, according to the results found. Most cases are observed among the black population, while cases among whites are less frequent. The significant predominance of cases among pardos, identified in the research, can be attributed to the ethnic differences present in the Amazon region.[13]

Regarding the age group, the results found a comprehensive range, from 14 to 99 years, with the predominance of cases in older patients, in accordance with the standard epidemiological profile of soft-part sarcomas. However, there was a difference between the average age identified by the work, 42.8 years, and the main literature, 60 years. This finding may be a reflection of inadequate interventions during early stages of the disease, accelerating the development of lesions.[14]

Soft-part sarcomas can develop in various areas of the body, but there is a preference for specific locations. The extremities, especially the lower limbs, are the regions most frequently affected, which is in accordance with the results of the study, which showed a significant predominance of cases with the primary site in the thigh and leg.[15] The large histological variety identified in this study is consistent with what is described in the literature, which has already documented more than 80 distinct subtypes of soft-part sarcomas. In addition, the most common histological types include liposarcoma, indifferent pleomorphic sarcoma, sinovial sarcoma and leiomiosarcoma, corroborating the observed results. However, the high frequency of high-grade subtypes may be the result of inadequate initial management.[5]

Regarding the initial treatment performed by non-specialists, the study revealed inadequate interventions in most cases, such as incomplete resections and the absence of adequate adjuvant therapies. Since it is a heterogeneous group of rare neoplasms, soft-part sarcomas should be accompanied by an experienced multidisciplinary team, ideally composed of a radiologist, pathologist, radiotherapist, oncologist and oncological orthopedist, in order to obtain the best standard of care.[16]

In addition, only 23% of patients treated by non-orthopedic oncologists underwent biopsy, which is fundamental not only for histopathological diagnosis, but also for the planning of appropriate therapy. The absence of this procedure increases the risk of lesions with positive margins and subtreatment, reinforcing the relevance of standardized diagnostic protocols in specialized centers.[4]

The main objectives in the treatment of soft-part sarcoma are to ensure patient survival, prevent recurrence, preserve function and reduce morbidity, with emphasis on surgical treatment as the main therapeutic approach. However, for the treatment to be effective, it is essential to perform a wide resection with free margins, which may not have been achieved in the initial treatment carried out by professionals who are not specialized in orthopedic oncology.[17]

In addition, 37% of cases have evolved to amputation, which is considered an extreme measure for patients with bulky tumors, involvement of the neurovascular beam and dysfunctional limbs. Although amputation should not be considered a therapeutic failure, it is worth considering the possibility of a positive outcome with the application of appropriate therapies.[3]

Recurrence of soft-part sarcoma is strongly associated with high histological degree, which corroborates the findings of this study. However, the recurrence rate of 77% over an average period of 1.7 years differs considerably from the average range of 17-26% after 5 years. This discrepancy may suggest the relevance of early diagnosis and proper treatment to reduce recurrence rates.[9]

During optimal treatment, up to 40% of patients with soft-part sarcoma develop metastatic disease. However, the study confirmed the occurrence of 49.2% of metastases in cases treated by non-orthopedic oncologists. This reinforces the importance of specialized follow-up for achieving the best possible outcome. Regarding the most common locations, the prevalence of lung and vertebral metastases is consistent with the epidemiology.[18]

The deaths observed among the analysed patients demonstrate the severity associated with the management of soft-part sarcomas. The death rate of 47.5%, with an average of 3.1 years to the outcome, represents an unfavorable finding compared to the global, of 35% in 5 years. The high prevalence of high-grade histological subtypes, the underuse of adjuvant therapies and the inappropriate conduct of non-specialists may have contributed to a worse result in this study.[19]

In view of the above, soft-part sarcomas represent a significant challenge in diagnosis and treatment due to their rarity, histological diversity and the complexity involved in their management. The findings of this study highlight the importance of early diagnosis, specialist treatment and multidisciplinary approach to improve outcomes. The high rate of mortality and recurrence

observed, as well as the evolution to metastases in many cases, highlights the urgent need for more effective diagnostic protocols, as well as the implementation of appropriate therapies from the start of treatment.

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CONTRIBUTIONS OF THE AUTHORS

Each author contributed individually and significantly to the development of this article. CFFB, YED and BLE: critical review of your intellectual content and final approval; MFG: contribution in the design of the work, critical review of your intellectual content and final approval; ANVM and FSAB: contribution in the design, analysis and interpretation of the data for the work, writing of the work and critical review of your intellectual content.

DATA AVAILABILITY DECLARATION

The data set for this article is available in the Figshare digital repository: https://doi.org/10.6084/m9.figshare.30698471

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ORIGINAL ARTICLE

OSTEOPOROSIS AND OSTEOMETABOLIC DISEASES

CAN TELEMEDICINE SUPPORT THE MANAGEMENT OF OSTEOGENESIS IMPERFECTA?

A TELEMEDICINA PODE APOIAR O MANEJO DA OSTEOGÊNESE IMPERFEITA?

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ABSTRACT

Objective: Considering the need for specialized care in Osteogenesis Imperfecta (OI), we investigated whether the use of a digital platform could optimize treatment and improve patients' quality of life. Methods: A guidance and data collection app was developed, complemented by a YouTube channel with physiotherapy instructions. The interventional clinical trial without a control group was carried out for 12 months with patients with OI. Patient functionality was assessed using the Functional Independence Measure (FIM) scale. Patients were monitored via telemedicine, receiving remote multidisciplinary support. Results: Thirteen participants completed the study. Significant improvements were observed in self-care, transfers, and mobility subitems ($p<0.05$). Program adherence was high, and no patient sustained injuries during the intervention. Additionally, a total transportation cost savings of R$ 113,852.00 was recorded, reducing direct expenses for patients and their families. Conclusion: Telemedicine proved effective in the rehabilitation of OI patients, providing both clinical and financial benefits. The digital platform facilitated treatment adherence, optimizing medical and physiotherapy support. Future studies should explore its large-scale application and integration into public health policies. ***Level of Evidence II; Therapeutic Studies— Investigating the Results of Treatment.***

Keywords: Telemedicine; Osteogenesis Imperfecta; Rehabilitation; Physical Therapy; Health Technology.

RESUMO

Objetivo: Considerando a necessidade de assistência especializada para Osteogênese Imperfeita (OI), investigamos se o uso de uma plataforma digital pode contribuir para otimizar o tratamento e melhorar a qualidade de vida dos pacientes. Métodos: Foi desenvolvido um aplicativo de orientação e coleta de dados, complementado por um canal no YouTube com instruções fisioterapêuticas. O ensaio clínico intervencional sem grupo controle foi realizado por 12 meses com pacientes portadores de OI. A funcionalidade dos pacientes foi avaliada utilizando a escala Medida de Independência Funcional (MIF). Os pacientes foram acompanhados por telemedicina, recebendo suporte multidisciplinar remoto. Resultados: Treze participantes concluíram o estudo. Houve melhoria significativa nos subitens de autocuidados, transferências e locomoção ($p<0,05$). A aderência ao programa foi elevada e nenhum paciente sofreu lesão durante a intervenção. Além disso, foi observada uma economia total com transporte de R$ 113.852,00, reduzindo os custos diretos para os pacientes e suas famílias. Conclusão: A telemedicina foi eficaz na reabilitação de pacientes com OI, proporcionando benefícios clínicos e financeiros. A plataforma digital facilitou a adesão ao tratamento, otimizando o suporte médico e fisioterapêutico. Estudos futuros devem explorar sua aplicação em maior escala e sua integração com políticas públicas de saúde. ***Nível de Evidência II; Estudo terapêutico - Investigação dos resultados do tratamento.***

Descritores: *Telemedicina; Osteogênese Imperfeita; Reabilitação; Fisioterapia; Tecnologia em Saúde.*

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INTRODUCTION

Telemedicine is defined as the use of digital communication technologies for the provision of remote health care services, allowing remote access to medical care and optimizing the treatment of various clinical conditions.[1] With the technological evolution and the increasing digitization of health care, this model has been widely adopted to broaden the reach of medical care.[1]

The advancement of telemedicine was especially driven by the COVID-19 pandemic, making it an essential tool to ensure continuity of care in various specialties. However, its potential has been being

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exploited for decades. One of the first records of its use occurred in 1966, when the Nebraska Psychiatric Institute, in partnership with the Norfolk State Hospital, implemented a telemedicine system for communication, education and psychiatric research using closed circuit televisions.[2,3]

Recent studies demonstrate the effectiveness of telemedicine in different clinical contexts. Shan and collaborators[4] conducted a systematic review showing that remotely monitored diabetic patients showed better glucose control compared to those treated in person. In addition, 66% of the world's population already owned a smartphone, which expands the possibilities of adherence to this modality of care.[4]

Given the success of telemedicine in the management of chronic diseases, there is a possibility of its application in the field of rare diseases, such as Imperfect Osteogenesis (IO). OI is a genetic disorder characterized by bone fragility, resulting from mutations in the genes COL1A1 and COL1A2, responsible for the synthesis of collagen type I. The prevalence of the disease is estimated at approximately 1 in every 20,000 births in the United States, although specific data on the Brazilian population are not yet available.[5]

IO management requires ongoing multidisciplinary follow-up and specialized care, often unavailable in conventional health centers. In addition, the daily care burden imposed on patients and caregivers is significant. Pinto and colleagues have demonstrated that individuals with rare diseases, including IO, demand about 13 hours a day for essential care only, and moving to health care is one of the main factors that impacts time and family costs.[6]

In view of this scenario, national and international guidelines emphasize the need for innovative strategies for the care of patients with IO.[7] Telemedicine, in this context, can be an effective alternative to reduce geographical and financial barriers, as well as broaden access to specialist medical support. To evaluate this possibility, we developed an application for guidance and data collection, associated with telemedicine care, with teleconsultations, teleorientation and tele-education. The main objective of this study was to evaluate the functionality of this platform in the follow-up of patients with IO and analyze its impact on treatment costs.

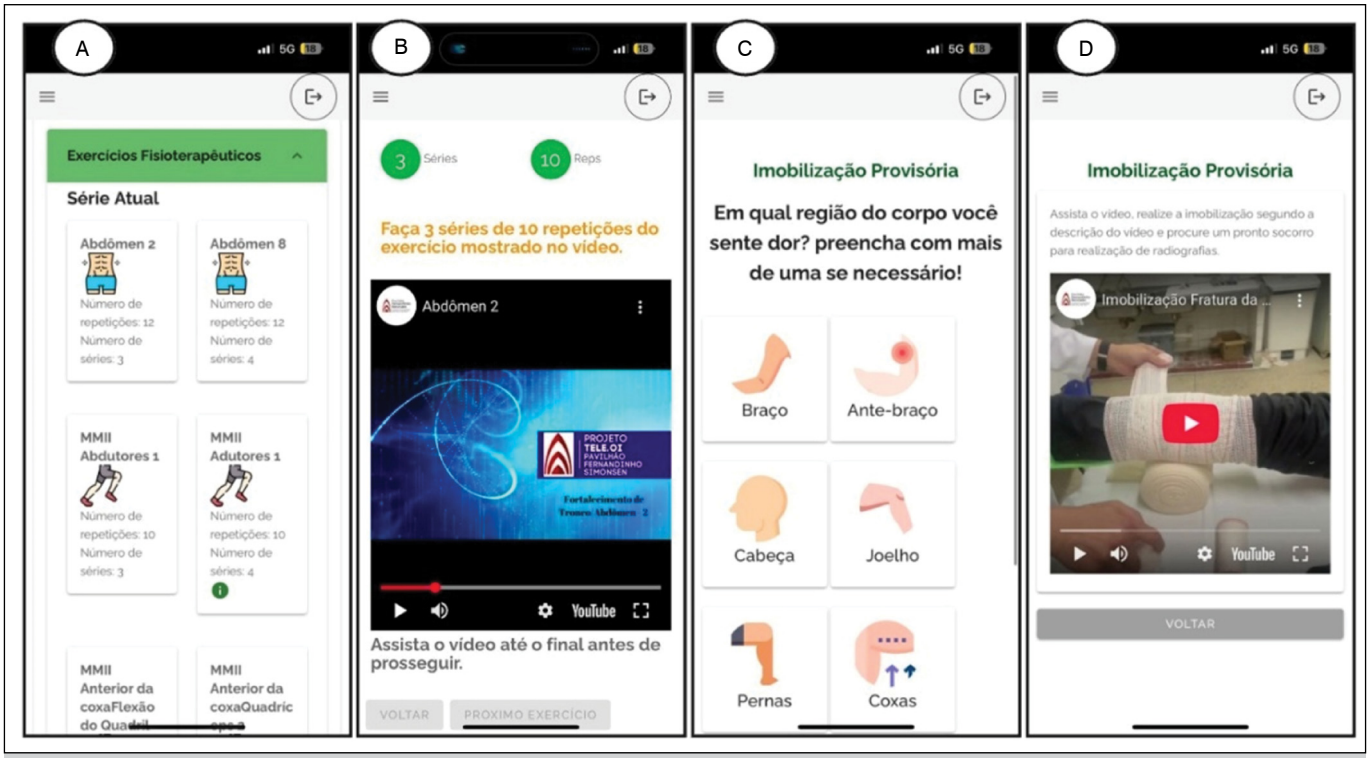
METHODS

Development of educational material and application for guidance and data collection

To enable the conduct of the study, a data collection and telemedicine tool was developed, consisting of a separate application and a YouTube channel with visual guidance on strengthening exercises and immobilization techniques for fractures (Figure 1). The platform was developed using the JavaScript programming language and works both as an environment for data collection and as a virtual medium to offer physiotherapeutic guidance and pre-hospital care, in addition to allowing the reporting of emergency situations. The YouTube channel – *Ortopedia Pediátrica Santa Casa*[8] – was created to assist patients at home, providing instructive videos on daily strength exercises (Fig. 1A and 1B) and pre-hospital immobilization techniques in cases of fractures (Fig. 1C and 1D). These videos are also integrated into the app through an automated system based on "Decision Trees", allowing direct access without the need to connect to YouTube. Access to the platform takes place through the domain www.orthotech.app, restricted to users authorized by the controller.

All patients underwent a first face-to-face consultation and subsequent telemedicine treatments were carried out within the outpatients of the Department of Orthopedics of the Santa Casa de São Paulo, using the electronic medical records.

In addition, an algorithm was developed to estimate the resource savings per physical therapy session, based on the *Treatment Outside the Home* (TOH)[9] and the *Atende* Program of the Prefecture of São Paulo.[10] The calculation took into account the distance



Source: Own app developed by the group. (www.orthotech.app).

Figure 1. Images that represent the instructions found in the app. A and B: practical examples and videos of recommended physiotherapeutic exercises. C and D: practical examples and videos of provisional immobilization.

between the patient's residence and the Santa Casa de São Paulo, classifying patients according to the applicable transport program. For residents in the capital, the cost was estimated based on the *Atende* Program, while for those outside, the TOH was used. Considering the average price of gasoline at R$ 5.00 during the intervention period, the app automatically calculated the cost of transportation per physical therapy session according to the record of the exercises on the platform.

CLINIC TESTING

Following the development of the digital tools, an interventional clinical trial, without a control group (Level of Evidence III), was conducted in the period from October 2022 to October 2023, with patients with Imperfect Osteogenesis (IO) treated in the Reference Center in Imperfect Osteogenesis (CROI) of the Orthopedics and Traumatology Service of Santa Casa de São Paulo. The inclusion criteria covered patients over three years of age, who had IO, who had a smartphone with Internet access and the ability to support the Google Meet program, used in teleconsultations. Patients under three years old, those without any kind of internet access, individuals on pamidronate treatment and elderly patients who, due to lack of familiarity with mobile devices, could not use the app or follow the therapy remotely were excluded. The Sillence classification[11] was not considered an inclusion or exclusion criterion.

During the study, patients received monthly multidisciplinary follow-up, conducted digitally and synchronously (online), with an orthopedist and a physiotherapist. The frequency of follow-up was changed only in cases of fracture. During the intervention period of one-year, digital physiotherapy was prescribed, recommending the activities to be performed at least three times a week, asynchronously (offline). The multidisciplinary telemedicine consultations were held every three months, when the physiotherapeutic training was adjusted to optimize muscle stimulation. For younger patients, it was recommended to use light bullets or toys as substitutes for the elastics. In addition, plastic pools were provided for walking training in a water environment in the cases of deformities of the lower limbs, as described in the literature.[12] In the initial consultation, the degree of functional independence of patients was evaluated through the Functional Independence Measurement (FIM), an instrument previously translated and validated into Brazilian Portuguese.[13]

Statistical analysis

For each FIM subitem, the data was submitted to Student's t test for paired samples. The adopted level of significance was of $p<0.05$.

Ethical aspects

The project was approved by the Research Ethics Committee of the Brotherhood of Santa Casa de Misericórdia de São Paulo under the number CAAE 42783021.3.0000.5479 and the clinical trial was registered in the *Clinical Trials* (NCT06555536).

RESULTS

The study included 15 participants, 7 women and 8 men, aged between 3 and 47 years, followed for a period of 12 months. Two participants quit the study, resulting in a total of 13 patients who completed the intervention. According to Sillence classification,[12] 1 patient (7.7%) had Imperfect Osteogenesis Type I, 6 (46.1%) were classified as Type III and 6 (46.1%) as Type IV. Among the patients who discontinued participation, a 15-year-old female teenager chose to discontinue the intervention, while a 47-year-old male patient reported difficulties in using the app.

The functional evaluation was carried out through the Functional Independence Measure (FIM), applied at the start of the intervention, after three months and at the end of the follow-up. To optimize the analysis, an automated algorithm was developed based on the document "Functional Guidance for the Use of FIM",[14] aimed at standardizing the application of the scale and minimizing intra and inter observers variations. The FIM is subdivided into 18 subpoints, including food, personal hygiene, bathing, clothing, toilet use, sphincter control, transfers, mobility, communication, social interaction, problem solving and memory. The intervention demonstrated statistical significance ($p<0.05$) in subpoints related to self-care, transfers and locomotion (Figure 2). The largest individual gains in FIM were observed in transfers to toilet (up to 550%), shower (up to 500%) and from bed to wheelchair (up to 450%). (Figure 3)

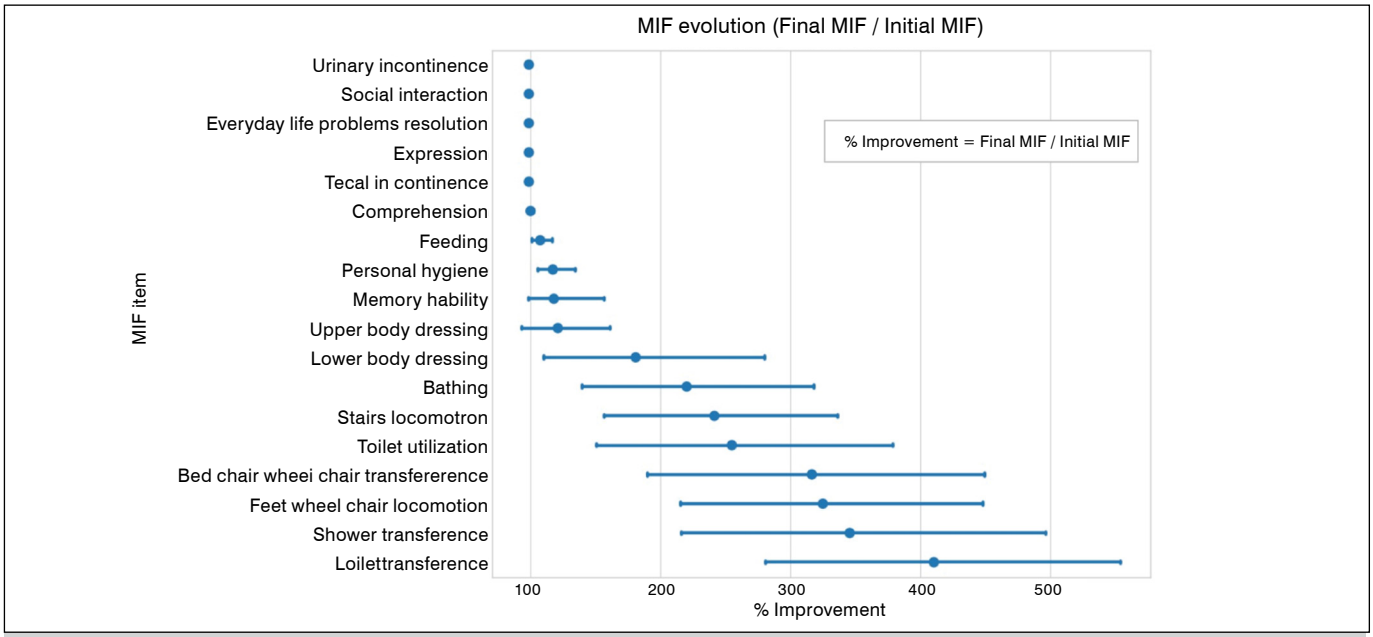


Figure 2. Hypothesis test for each FIM item (Ho: FIM before = FIM after). The circles represent the values of p and the color indicates the statistical significance ($p<0.05$). Values of p less than 0.05, the null hypothesis at the 95% confidence level is rejected, assuming there was an improvement in the FIM.

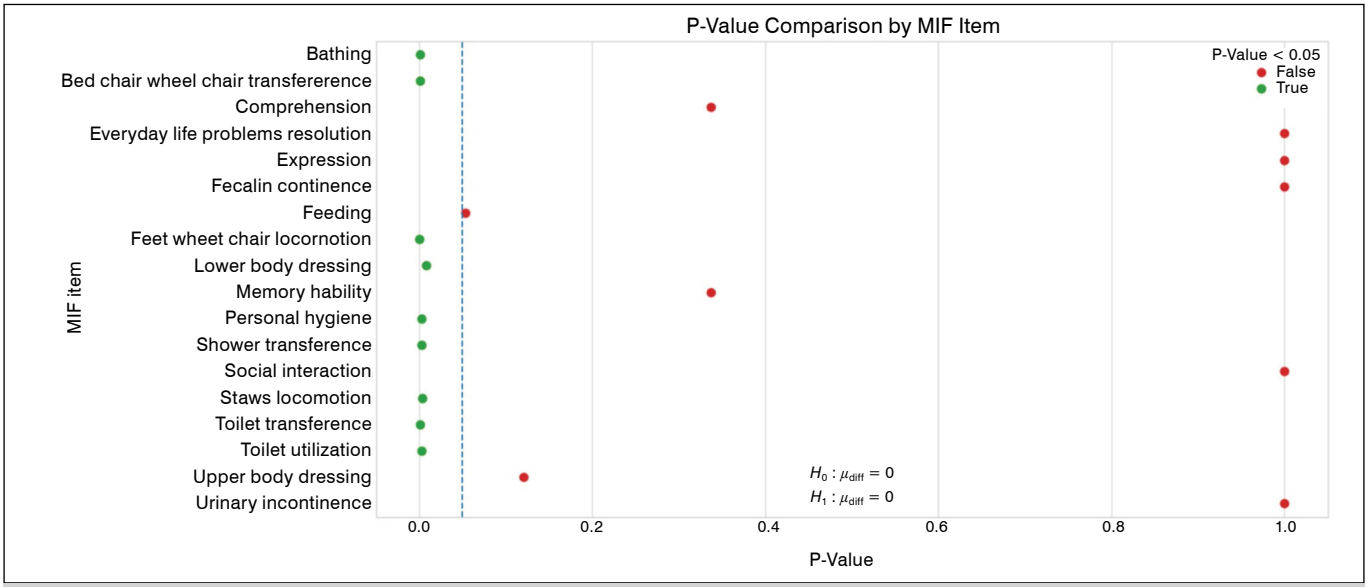


Figure 3. Percentage variation of FIM items, calculated by the ratio (final FIM/initial FIM) x 100. Values of 100% indicate no improvement, while 300% triple the initial value.

No patient suffered injuries related to the practice of distance-oriented physiotherapeutic exercises. Adherence to the program was high, with 13 of the 15 participants using the app at least three times a week throughout the year of intervention.

At the end of the study, the total cost saved with transportation was R$ 113,852.00, corresponding to a monthly average of R$ 8,758.00 per patient over the 12 months of follow-up. (Figure 4)

DISCUSSION

Our results showed that the use of telemedicine in the treatment of Imperfect Osteogenesis (IOS) resulted in significant functional gains, especially in the items of the Functional Independence Measure (FIM) related to transfers (toilet, shower and from the wheelchair bed). In addition, we observed substantial savings with transport over the intervention period.

One of the challenges frequently mentioned in the literature on IO and other rare diseases is the impact of continuous care on caregivers, both in terms of dedicated time and financial impact on family income. Studies indicate that time spent in care can generate indirect costs due to the reduction in working hours or even the caregiver's exit from the professional market.[6] In our intervention, caregivers report not only functional improvements in patients (such as greater independence to dress and make transfers), but also an increase in time and quality of life for themselves.

Another relevant aspect was the possibility of following patients inside their homes through telemedicine. This approach has identified specific challenges of the household environment, including precarious housing conditions that can contribute to the increased incidence of fractures. From these observations, it was possible to suggest customized adaptive strategies for each case. In addition, telemedicine enabled the implementation of a teleorientation service for caregivers and health professionals in municipalities in the interior of São Paulo and Minas Gerais. This initiative included from instructions for pre-hospital care to guidelines for proper immobilization of fractures. In one case, an orthopedic immobilization technician was remotely guided on the best technique for stabilization, allowing pain control until the patient was transferred to a specialized service (Figure 5). Similarly, we were able to assist a generalist orthopedist in the therapeutic decision, contraindicating the use of plates and

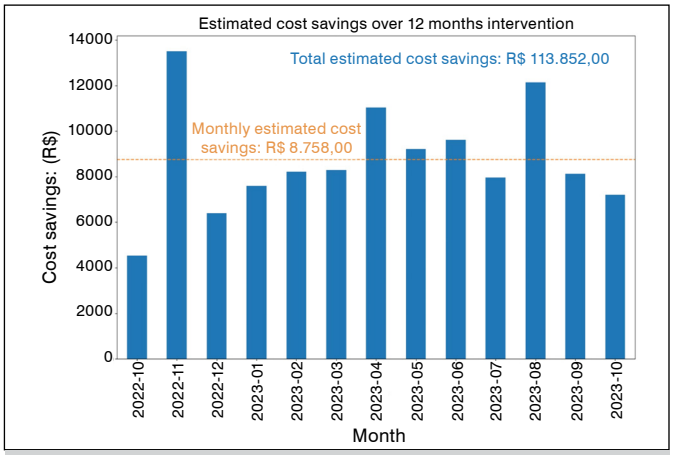
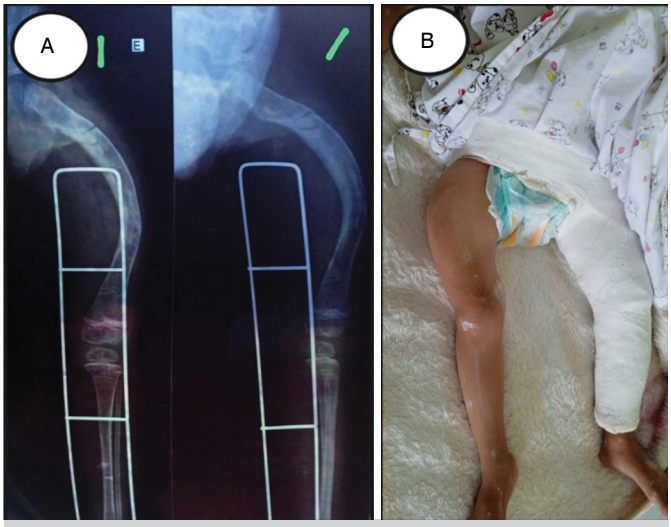


Figure 4. Economy (in real) generated by intervention activities over twelve months.



Source: Author.

Figure 5. X-ray image of left femur fracture (A) and immobilized left lower limb photograph of patient with fracture (B).

screws – an inappropriate approach for patients with IO, as it can result in unfavorable outcomes.[15]

From a physiotherapeutic point of view, the use of the FIM Scale allowed not only to quantify the individual limitations of patients, but also to adapt the training in a personalized way. At the end of the intervention period, the re-evaluation with the FIM demonstrated the effectiveness of the applied approach. Adherence to the program was high, and no patient suffered injuries due to remote exercises. We believe that implementation of engagement strategies, such as application gamification, can further improve participant adherence. One limitation of the study was the absence of a control group. Due to the rarity of the disease and the refusal of the patients to participate if there was a possibility of allocation in the control group, we chose to offer the intervention to all 15 participants.

In general, rare diseases require innovative solutions to ensure effective therapies, as the diagnosis and treatment of these patients still presents significant challenges. It is estimated that about 50% of individuals with suspected rare disease remain unconfirmed.[16] In Europe, approximately 25% of these patients take between 5 and 30 years to get a definitive diagnosis, and 40% undergo incorrect diagnoses and inadequate treatments.[17] In Brazil, these data are still not well documented.

As far as medication is concerned, the situation is even more critical: less than 3% of rare diseases have a specific pharmacological therapy, and the costs of these medications can be up to 13.8 times higher than those of conventional drugs.[18,19] In response to this problem, the *International Rare Diseases Research Consortium* (IRDiRC) was created in 2011 with the aim of fostering international collaboration for the development of new diagnostic and treatment methodologies. In 2017, this organization established a 10-year strategic plan, with one pillar focused on assessing the impact of treatments for rare diseases.[20] We hope our findings can contribute to these guidelines and encourage new approaches to the management of patients with IO. Finally, the developed digital tool proved to be effective both in expanding the access to specialist telemedicine care and in improving the function of patients, in addition to providing significant savings with transportation for face-to-face consultations.

CONCLUSION

Our results indicate that the approach applied in this study may be an important ally in the treatment of Imperfect Osteogenesis, contributing to the improvement of the quality of life of patients and reducing the costs associated with treatment. Although the number of participants has been limited, this restriction is justified by the rarity of the disease. Similar digital tools should be considered for public health policies, expanding access to specialized therapies for patients with rare diseases. Future research with larger samples will be fundamental to more comprehensively assess the clinical outcomes and the impact of this approach in the long run.

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CONTRIBUTIONS OF THE AUTHORS

Each author personally and significantly contributed to the development of this article: PHMMVC: design of the project, development of the clinical trial, collection and interpretation of the data and writing of the article; FHVN: analysis and interpretation of the data; VYAN: development of the clinical trial; EOG and MA: critical review of relevant intellectual content; CS: final version to be published and conducted critical review of relevant intellectual content.

DATA AVAILABILITY DECLARATION

The data will be made available when requested.

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ANALYSIS OF SHOULDER ROTATOR TRAINING BY ARM WRESTLERS

ANÁLISE DA FORÇA DE ROTADORES DE OMBRO POR PRATICANTES DE LUTA-DE-BRAÇO

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ABSTRACT

Introduction: Armwrestling is officially recognized in 160 countries affiliated with the World Armwrestling Federation. The movements involve the upper limbs, with emphasis on shoulder rotators, elbow flexors and extensors, and wrist. Objective: To evaluate the balance of agonist/antagonist forces in shoulder rotation and to verify differences in relation to the normative value of 64% for strength and 66% for power in the ratio between internal and external rotators. Methods: Seventeen international-level athletes were selected. A Biodex isokinetic dynamometer was used to evaluate the balance in peak torque of agonists and antagonists in shoulder rotation with strength and power tests. Results: Significant differences were found in the agonist/antagonist ratio in the power tests, with an average of 57% (P-value = 0.001) versus 66% of the normative value, and in the strength test, 58% (P-value = 0.064) versus 64% of the normative value. Discussion: The muscular strength and power of the subjects were analyzed in relation to normative parameters for internal and external shoulder rotation. The results were significant in relation to the normative values, indicating muscular imbalance in this group of athletes. Conclusion: The results indicate that the training performed by armwrestlers promotes a greater gain in strength of the internal rotators of the shoulder than their antagonists. ***Level of Evidence I; Diagnostic criteria previously tested on consecutive patients (with a universally applied "gold standard").***

Keywords: Physical Functional Performance; Muscle Strength Dynamometer; Shoulder; Resistance Training; Sports Medicine.

RESUMO

Introdução: A Luta-de-Braço é reconhecida em 160 países filiados à Federação Mundial. Os movimentos envolvem membros superiores, com destaque para rotadores de ombro, flexores e extensores de cotovelo e punho. Objetivo: Avaliar o equilíbrio das forças agonistas/antagonistas na rotação do ombro e verificar diferenças em relação ao valor normativo de 64% para força e 66% para potência na razão entre rotadores internos e externos. Métodos: Foram selecionados 17 atletas de nível internacional. Utilizou-se um dinamômetro isocinético Biodex para avaliar o equilíbrio no pico de torque dos agonistas e antagonistas na rotação de ombro com testes de força e potência. Resultados: Foram encontradas diferenças significativas na razão agonista/antagonista nos testes de potência com média 57% (valor-P= 0.001) contra 66% do valor normativo e no de força 58% (valor-P= 0.064) contra 64% do valor normativo. Discussão: A força e potência muscular dos avaliados foram analisados em relação a parâmetros normativos para rotação interna e externa de ombro. Os resultados se revelaram significativos em relação aos valores normativos, indicando desequilíbrio muscular neste grupo de atleta. Conclusão: Os resultados indicam que o treinamento realizado por praticantes de luta-de-braço promove um ganho de força dos rotadores internos de ombro maior que seus antagonistas. ***Nível de Evidência I; Teste de critérios diagnósticos desenvolvidos anteriormente em pacientes consecutivos (com padrão de referência "ouro" aplicado).***

Descritores: *Desempenho Físico Funcional; Dinamômetro de Força Muscular; Ombro; Treinamento Resistido, Medicina Esportiva.*

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INTRODUCTION

Arm-fighting is a force-fighting sport, a form of fighting between two opponents where there is no draw. According to historians of the World Arms Federation, papyrus was found in Egypt, more than 2,000 years before Christ, showing activities similar to arms struggle. Other works demonstrate the practice of this sport by the Vikings in Scandinavia and various artistic reports in other parts of the world. Currently it is a popular sport, but few people know it is officially recognized by the International Olympic Council and that it has a world federation (*World Armwrestling Federation*) with 160 affiliated countries. In Brazil, wrestling began to be practiced as a sport in 1949 at an event promoted by the school Ateneu Paulista

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in Campinas. In the 1960s there were tournaments promoted by the Paulistani newspaper Gazeta Esportiva where the campers Hugues Jorge, Nivaldo Félix, Washington Lázaro, the twins Celso and Célio Capelli among others also from Campinas were promoted.[1] At the 2022 World Cup held in Antalya, Turkey, about 1500 athletes from 51 countries participated.[2]

In this sport are required mainly, much of the musculature of the chest and upper limbs especially the shoulder rotating groups, flexors and extenders of the elbow and wrist. The most experienced athletes outperform their opponents because they can maintain a greater amount of muscles contracted simultaneously, this process is called intermuscular coordination. The goal is to unite all the forces in a single vector. Among these movements is the internal shoulder rotation that is carried out by the muscles: chest,[3] deltoid (front portion), large dorsal, larger round and subescapular; The antagonist movement, the external rotation of shoulder, is carried out by the muscles: intra-spinal, minor round and deltoid (back portion).[4]

The main wrestling techniques called "cross" and "up" consist of stabilizing the inner rotation of the shoulder while the athlete performs the elbow flexion beyond all strength in the wrist, which is required to master the "top" of the opponent's lever, at the same time a lateral bend of the trunk is performed to "open the set" and lower the opponent's hand to win the fight.[5]

The aim of this study is to evaluate the proportion of the agonist (internal rotation) and antagonist (external rotation) forces in the shoulder rotation movement of wrestling practitioners.

MATERIALS AND METHODS

A cross-sectional study was conducted to test the dominant limbs of athletes with an isokinetic dynamometer [5] (Biodex System 4 Pro from Biodex Medical System, Inc. 20 Ramsey Road Shirley, New York) in a concentric/concentric assessment of the balance of shoulder rotation agonists/antagonists.

All have completed the Terms of Free and Informed Consent – TCLE. The study was approved by the Ethics and Research Committee Number of the CAAE: 76328317.9.0000.5404

Inclusion Criteria

- Male adults, over 18 years of age and at least 5 years of wrestling practice;
- International competitive level, only Brazilian champions and vice champions.

Exclusion criteria

Present functional score DASH[6] greater than 39, where less than 20 is excellent, 20 to 39 is good, 40 to 60 regular and above 60 bad (functional disability), that is, were excluded those who had regular and bad score, being accepted only those rated as excellent and good.

Show pain or discomfort before or during tests.

Isocinetic Assessments

Athletes were allowed to perform body warming according to each individual's habit. We provided varied and elastic weights, such as those used by athletes in the heating of training and championships, before being positioned on the equipment.

As a protocol, the evaluation on the isocinetic dynamometer promotes familiarity with the same speeds that will be tested, but with fewer repetitions, and is initiated by the force test with movements performed at 60o/s and for the shoulder power test at 180o/s.[7,8] Between familiarity with the equipment and the main test as well as between the variations in the force to power test, a 90-second interval was observed for each series of movements. The tests were performed sitting to stabilize the athlete's body and isolate the shoulder movement without influence from other muscle groups that were not being tested.

The dominant arm was positioned with the shoulder abducted 45 degrees, the elbow and forearm supported on support fixed on the lever and hand holding manople that kept the handle in neutral position without flexo-extension or prono-supination. The machine axis was positioned 20 degrees of vertical rotation and 50 degrees of horizontal rotation and the height of the chair was adjusted with the elbow supported without lifting or depressing the scapula.[9]

The evaluations were performed with concentric strength tests, which is the strength that shorten the muscle generating movement, in all variations to extract the difference between the agonist (internal rotators) and antagonist (external rotators) muscles involved in the joint movement tested.

Statistical Methodology

To describe the profile of the sample for the variables in study were made frequency tables of the categorial variables (age, height, weight, BMI, forearm length and practice time), with absolute frequency values (n) and percentage (%), and descriptive statistics of the numerical variables of the peak torque, found in the evaluation of external/internal shoulder rotation movements with strength and potency test, mean values, standard deviation and confidence interval.[10]

The data that best demonstrate the differences between the agonist and the antagonist and their variations in the joint movements involved were selected to evaluate results: the peak torque that has its result expressed in Nm (newton-meter) and the agonist/antagonist ratio of the forces in these movements found by dividing the peak torque of the agonist by the peak torque antagonist and multiplied by 100 expressed in % (percentage).[5]

To compare the results of the athletes with the normative values stipulated by the equipment, the hypothesis test for average was used and the confidence level of the sample (n=17) was validated by the confidence interval of 95%.[11] The level of significance adopted for the statistical tests was 5%.[8]

RESULTS

17 athletes were selected for the tests. Table 1 shows the characteristics of the total sample (n=17). The average age of the participants was 36 years. Table 2 presents the performance of athletes in terms of peak strength in external and internal rotations with values expressed in Newton per meter (Nm), with these values being calculated the ratio between agonist and antagonist, which present significant values or not.

Table 3 shows separately the comparisons of the values found in the evaluation of the athletes with the normative parameters in the difference in peak torque, the agonist/antagonist ratio both in the strength test, which was not significant with value-P = 0.064, and the power, which was significant with value-P = 0.001, with their differences expressed in percentage (%).

Table 1. Descriptive analysis of the evaluated group.

Variable	N	Media	SD	IC 95%
Age (years)	17	36	8.42	(31.6; 40.3)
Height (cm)	17	180	0.08	(176; 184)
Weight (kg)	17	95.7	19.6	(85.7; 105.8)
BMI	17	29.5	4.89	(26.9; 32.0)
Frontarm (cm)	17	50.2	2.15	(49.1; 51.3)
Practice Time (years)	17	19.3	8.28	(15.1; 23.6)

95% CI: 95% confidence interval of the average.

<< SUMÁRIO

Table 2. Athletes' performance on the peak torque of the external and internal shoulder rotations.

Variable	N	Media	SD	IC 95%
External Rotation Force	17	58.9Nm	13.6	(52; 66)
Internal Rotation Force	17	102.7Nm	22.8	(91; 114)
External Rotation Power	17	55.2Nm	12.2	(49; 61)
Internal Rotation Power	17	96.8Nm	18.6	(87; 106)

95% CI: 95% confidence interval of the average.

Table 3. Comparison of athlete parameters with the normative values of the agonist/antagonist ratio.

Variable	N	Media	SD	Normative value	IC95%	P-value
test strength	17	58.6%	11.2	64 %	(53; 64)	P=0.064
Power test	17	57.4%	8.8	66 %	(53; 62)	P=0.001

* P-value referring to the hypothesis test for average for comparison with the normative values. 95% CI: 95% confidence interval of the average.[10-12]

DISCUSSION

This study analyzed the strength and muscle potency of athletes in relation to normative parameters for internal and external shoulder joint rotation of international-class wrestling practitioners. The internal and external rotations were evaluated as fundamental for the execution of a good technique in this sport.

The muscles are contracted in different functions, here they have been analyzed as agonists and antagonists. The agonists are the muscles responsible for the desired action while the antagonists simultaneously contract in opposition and relax to allow a smooth movement.[13]

In high-performance athletes and especially in this sport of strength and power, the peak torque is crucial in the result of the fight. For the parameters (spike torque and agonist/antagonist torque ratio) the evaluation on the isokinetic dynamometer is very accurate and these were the data analyzed in this study, also considering that this force difference is used to demonstrate the balance of the muscles that act in the joints.[5]

"*Isocinetic dynamometers are measuring instruments that provide clinicians with information about the dynamics, i.e., movement, mechanical performance of muscle groups*" Dvir.[14]

The shoulder rotators were tested with the sole aim of observing a possible change in the agonist/antagonist ratio because in this sport the internal rotation is much more demanding.[14]

According to the results, the difference between the sample parameters and the normative values for agonist/antagonist ratio in the assessment of shoulder rotation was significant in the power test at 180 degrees per second, according to the data P=0.001 and no significant difference with P=0.064 in the strength test at 60 degrees per second as demonstrated in Table 3.

The normative values used in this work, from 64% to 66% agonist/antagonist ratio in strength and potency tests respectively, are the reference values of the Biodex System isokinetic dynamometer that coincide with an approximate average of the values found by Brown et al. (1988) 61% a 72% in male athletes, Connelly et al. (1989) 62% in the test with male average audience, Ellenbecker (1988) 65% to 72% for male athletes, McMaster et al. (1991) 55% to 78% for the general public and male athletes and Reid et al. (1989) 53% to 66% for the general public and male athletes respectively.[15]

The ratio between the internal and external rotor forces found in wrestling athletes is different from normative values, but even greater differences were found in judo player as demonstrated by Marcondes et al. (2019) where the percentage difference in the shoulder rotators tests of the dominant arms was 49.4% for strength and 42.9% for power, according to the isokinetic assessment for the shoulder rotators.[16]

Re-evaluating training is fundamental in the pursuit of a better balance of forces and with that, a possible improvement in performance in this sport.

CONCLUSION

The results of the evaluations of shoulder rotators in wrestling practitioners show, by analysis of performance at peak torque of the agonists and antagonists of this movement, a lower power of the external rotators relative to the internal ones.

CONTRIBUTIONS OF THE AUTHORS

Each author personally and significantly contributed to the development of this article: LLSG: drawing of the work, interpretation of the data and writing; MAP: analysis of the data and review of the content and help in writing; PRM: drawing of the work and interpretation of the data of isokinetic analysis; ALLA: drawing of the work and intellectual concept of the article; ME: intellectual concept of the article, interpretation of the data, review of the writing and final approval of the version of the manuscript to be published.

DATA AVAILABILITY DECLARATION

The data set of this article is available in Mauro CIPED - It is necessary to publish in REDU UNICAMP, via the login of the guide: https://www.sbu.unicamp.br/sbu/repositorio-de-dados-de-requisa-da-unicamp/. Source: NORMATIVA_PRPG_CCPG_1_2024.

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PREVALENCE OF PALMARIS LONGUS TENDON AGENESIS IN A POPULATION OF MEDICAL STUDENTS

PREVALÊNCIA DA AGENESIA DO TENDÃO DO MÚSCULO PALMAR LONGO EM UMA POPULAÇÃO DE ESTUDANTES DE MEDICINA

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ABSTRACT

Objective: To assess the prevalence of palmaris longus (PL) tendon agenesis in a population of medical students. Methods: The presence of the palmaris longus tendon was evaluated using Schaeffer's test, applied bilaterally in 100 medical students, totaling 200 evaluated antimeres. The data were analyzed for unilateral or bilateral presence of the tendon, in relation to gender, laterality (right or left), and the occurrence of agenesis. Results: Agenesis of the palmaris longus tendon was observed in 32 out of 200 evaluated antimeres (16%). Among female participants, agenesis was identified in 21 antimeres (21%), whereas in males it was observed in 11 antimeres (11%). Tendon absence was more frequent in the left upper limb, regardless of gender. Conclusion: The prevalence of palmaris longus tendon agenesis in the studied sample was 16%, with a higher frequency among females. Tendon absence was more common on the left side in both genders. ***Level of Evidence IV; Case series.***

Keywords: Tendons; Anatomic Variation; Wrist; Anatomic Landmarks.

RESUMO

Objetivo: Avaliar a prevalência da agenesia do tendão do músculo palmar longo (PL) em uma população de estudantes de medicina. Métodos: A presença do tendão do músculo palmar longo foi investigada por meio da manobra de Schaeffer, aplicada bilateralmente em 100 estudantes de medicina, totalizando 200 antímeros avaliados. Os dados obtidos foram analisados quanto à presença unilateral ou bilateral do tendão, em relação ao sexo, à lateralidade (direita ou esquerda) e à ocorrência de agenesia. Resultados: A agenesia do tendão do músculo palmar longo foi observada em 32 dos 200 antímeros avaliados (16%). Entre as mulheres, a agenesia foi identificada em 21 antímeros (21%), enquanto entre os homens, foi observada em 11 antímeros (11%). A ausência do tendão foi mais frequente no membro superior esquerdo em ambos os sexos. Conclusão: A prevalência da agenesia do tendão do músculo palmar longo na amostra estudada foi de 16%, sendo mais comum em mulheres. A ausência do tendão apresentou predomínio no lado esquerdo, independentemente do sexo. ***Nível de Evidência IV; Série de Casos****.*

Descritores: *Tendões; Variação Anatômica; Punho; Pontos de Referência Anatômicos.*

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INTRODUCTION

The *palmaris longus* (PL) is a superficial muscle covered by the anterior fascia of the forearm,[1-3] being responsible for the flexion of the wrist and the tension of the palm aponeurosis.[2-5] It originates from the medial epicondyle of the humerus, extending medially to the carpal's radial flexor muscle and laterally to the carpal's ulnar flexor, entering the palm aponeurosis.[2,3,6] It is innervated predominantly by the median nerve and can, in some cases, receive innervation from the ulnar nerve, and is vascularized by recurrent ulnar arteries.[1,7]
This muscle is considered to be the most variable of the human body, and may present variations in duplication, agenesis, position and insertion.[8,9] According to Yammine et al.[10], the variations in the PL are associated with a dominant pattern of autosomal inheritance with incomplete penetration. There is evidence of its evolutionary role: in arboreal primates, the muscle is consistently present, assisting in climbing and grasping objects; whereas in bipedal or predominantly terrestrial primates, its presence becomes more variable, PL has become less necessary, which justifies its vestigial characteristic.[5,8,11,12]
The prevalence of PL tendon agenesis is approximately 15%.[2,5] Epidemiological studies suggest that the regression of this muscle may be related to the migration of the *Homo sapiens* from the African

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<< SUMÁRIO

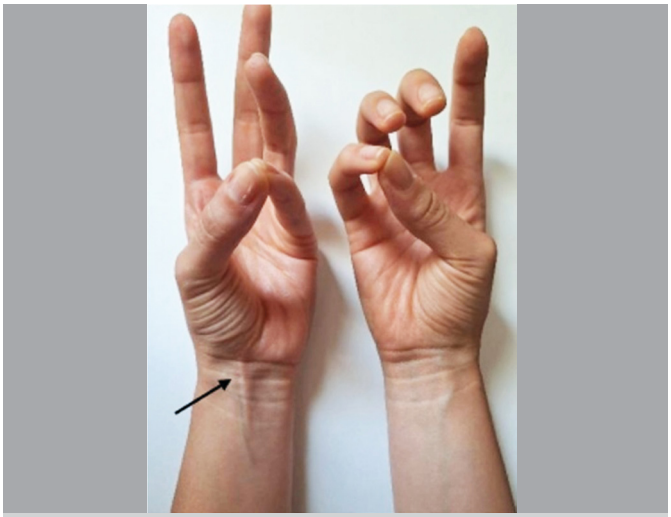
continent, as Caucasian populations present higher rates of PL agenesis (26%) compared to descendants of Asian peoples (12%) and African peoples (6%).[7,13] Some articles also indicate that the absence of PL occurs more frequently unilaterally, predominantly on the left side, and is more common in female sex.[2,12,14,15]

Although it is considered a vestigial muscle, the absence of which does not compromise the movements of the hand or wrist, the PL has clinical and surgical relevance.[9,16] It is used in reconstructive procedures, such as replacement and repair of other tendons, repair of facial paralysis and reconstruction of labia defects, in addition to serving as an anatomical reference for the location of the median nerve.[3,15,17,18] Changes in its anatomical insertion may also be associated with clinical manifestations of carpal tunnel syndrome.[11,12,19-23]

Knowledge of the prevalence of PL agenesis is essential to provide anatomical information to surgeons, allowing them to know its variations, which are useful in planning surgeries involving the use of palmar muscle as a graft or as an anatomical reference in surgical procedures. We found few studies addressing the prevalence of this tendon in the Brazilian population. Thus, the present study aimed to identify the frequency of PL muscle tendon agenesis in a population of 100 medical students.

MATERIALS AND METHODS

This research was approved by the Ethics Committee of our institution under the number 57590422.0.0000.5373. The sample was composed of 100 medical students, from the first to the sixth year of the course, aged between 18 and 30 years, of whom 50 were female and 50 were male. Only individuals without a history of trauma in the ventral area of the forearms or any history of diseases affecting the soft parts of this area were included. The selection of participants was random, depending on the availability of volunteers. Verification of the PL tendon was performed using Schaeffer's maneuver on both forearms. This test consists of asking the volunteer to keep the elbow folded at 90 degrees and perform the wrist flexion associated with the opposition of the thumb to the smallest finger (Figure 1). The presence of the tendon was considered positive when it became visible in the distal third of the wrist; if it was not possible to visualize it, its absence was recorded.



Source: St. Archive of the Faculty of Medical and Health Sciences of the Pontifical Catholic University of São Paulo.

Figure 1. Anterior photographic view depicting bilateral performance of Schaeffer's maneuver. The PL tendon is present on the left side (arrow) and there is agenesis of this tendon on the right side.

The tests for each antimere were conducted independently by two authors (PDT and CC), and the results were later compared to verify inter-observer agreement. The gender of the student and the side (or bilaterality) of the long PL tendon agenesis were recorded.

RESULTS

Based on the data obtained, the prevalence of absence of the long PL tendon in the evaluated population was 16%. When stratified by sex, absent tendon was observed in 21% of women and 11% of men. In the female sample (n = 100 upper antimeres), the absence of the tendon was identified in 21 antimeres, with eight in the upper right limb and 13 in the left. Three volunteers presented bilateral agenesis. In the male sample (n = 100 upper antimeres), the absence was observed in 11 members, five to the right and six to the left, with the occurrence of bilateral agenesis in three individuals.

Considering the general prevalence of the absence of the PS tendon by antimeres, regardless of gender, a rate of 13% was observed in the right upper limb and 19% in the left upper limb. Stratified by sex, the data indicated that the absence on the right side was 16% in women and 10% in men; on the left side, the agenesis was 26% in women and 12% in men. The overall prevalence of bilateral PL tendon agenesis, considering both sexes, was 6% in both sexes. The results are grouped in Table 1.

Table 1. Prevalence of long PL tendon agenesis in the population of 100 students of the medical course, according to sex and side.

Sex	Female	Male
Unilateral and bilateral agency	21%	11%
Unilateral agenesis to the right	16%	10%
Unilateral agenesis to the left	26%	12%
Bilateral agency	6%	6%
Total of individuals	50	50

DISCUSSION

The main finding of this study was the verification of the prevalence of PL tendon agenesis (PL) in 16% of the sample analyzed. This condition was more common among female individuals (21%), compared to male individuals (11%). Regarding laterality, the absence of tendon was more prevalent in the left upper limb (19%) than in the right (13%). Bilateral agenesis was observed in 6% of participants, with no difference between the sexes.

National studies conducted by Garcia et al.[1] Bonsi,[24] Pierucci et al.[25] and Morais et al.[26] report frequencies of agenesis of PL of 13.9%, 16.5%, 9.2% and 26.5%, in this order. The findings of this study approximate the data reported by Bonsi,[24] whose prevalence was 16.5%, and are also comparable to the results of Garcia et al.[1] (13.9%). On the other hand, the percentages observed by Pierucci et al.[25] and Morais et al.[26] showed more pronounced discrepancies. A methodological analysis of the studies of Garcia et al.[1] and Bonsi[24] revealed that both used the same procedure adopted in this research, i.e. Schaeffer's maneuver, and it is therefore not possible to attribute the observed divergence to the technique employed. However, Morais et al.[26] applied four different maneuvers to confirm the absence of the PL tendon, which may justify, at least partially, the observed difference.

In comparing our results with international literature data, we found that Cohen et al.[13] reported the absence of PL tendon in 28% of the Israeli population, while Al Risi et al.[19] found the prevalence of agenesia in 7.5% of the Oman population. In the Pakistani population, Javaid27 observed a 12% prevalence of this tendon agenesis. On the other hand, Kapoor et al.[15] observed the absence of tendon in 17.2% of the Indian population and Sadacharan et al.[12] identified

this absence in 12.8% of the African population. Cohen et al.[13] suggested that PL prevalence is more strongly associated with geographical than ethnic factors, observing that individuals of different ethnicities, but residing in the same geographic region, presented similar rates of agenesis. This hypothesis may contribute to the understanding of the observed variations in relation to the data obtained in this study.

The results presented here corroborate the findings of Morais et al.[26] Javaid,[27] Ioannis et al.[2] Raouf et al.[14] and Kapoor et al.[1] which also report greater prevalence of PL agenesis in female subjects and left antimere. These findings contrast with the data from Al Risi et al.[19] that identified a higher prevalence of the absence of PL in males. Pierucci et al.[25] in turn described a higher prevalence of unilateral agenesis in the left antimeric among men (15.2%) and in the right antimeric among women (18%).

Additionally, the data from this study indicate that bilateral PL agenesis occurs less frequently than unilateral agenesis, a pattern also reported by Garcia et al.[1] Pierucci et al.[25] Sadacharan et al.[12] and Morais et al.[26] No records were identified in the literature indicating a higher prevalence of bilateral agenesis of the PL tendon compared to the unilateral form.

As a limitation of our study we can cite that we did not investigate whether there was a correlation between the side of the agenesis of the PL tendon and the dominant limb and, as a clinical relevance, we can highlight that the agenesis of this tendon is present in a small portion of the population and that the bilateral absence is even smaller, which makes the PL tendon a viable option for use as a tendon graft in lesions of the finger flexors, for example, since the removal of this anatomical structure practically does not interfere with the flexion of the wrist, since other tendons, such as the ulnar flex of the carp and the radial flexor of the carp perform this function more efficiently.

CONCLUSION

In the analyzed population, PL tendon agenesis was observed in 16% of subjects, being more common in females and left antimeres. Bilateral agenesis was identified in 6% of individuals of both sexes.

CONTRIBUTIONS OF THE AUTHORS

Each author contributed individually and significantly to the development of this article. PDT and CC: data acquisition and critical review of their intellectual content; JCG: data interpretation and final drafting of the article; WRD and LGM: bibliographic research; EBC: general guidance and review of the manuscript.

DATA AVAILABILITY DECLARATION

The contents underlying the research are available in the manuscript.

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RETROSPECTIVE ANALYSIS OF MALLET FINGER TREATED WITH EXTENSION BLOCK FIXATION USING THE ISHIGURO TECHNIQUE

ANÁLISE RETROSPECTIVA DE DEDO EM MARTELO ÓSSEO TRATADOS PELA FIXAÇÃO COM BLOQUEIO DE EXTENSÃO PELO MÉTODO DE ISHIGURO

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ABSTRACT

Objective: This study aimed to evaluate the outcomes of the Ishiguro technique in the treatment of flexion deformity of the interphalangeal distal (IFD) joint, known as bony mallet finger (BMF) Albertoni Type C2, addressing a common condition that may cause functional impairment if not adequately treated. Methods: All cases of patients with Albertoni Type C2 bony mallet finger (BMF) who underwent the Ishiguro method between March 2018 and February 2023 were included, with a minimum postoperative follow-up of five months; cases of patients who did not complete the minimum five-month postoperative follow-up were excluded. Epidemiological data (age, sex, dominance) and fracture-related information (affected finger, fragment size, time from trauma to surgery, follow-up duration) were analyzed. Outcomes included anatomic reduction, union, return to activities, pain, flexion deficit, passive extension of the affected finger, passive extension of the contralateral finger, and Crawford criteria. Results: None of the analyzed characteristics—such as age, affected finger, interval between trauma and surgery, fragment size, and union—significantly influenced anatomic reduction ($p > 0.05$). The Crawford scale, as well as anatomic reduction, showed no significant impact ($p > 0.05$). However, all patients with positive results on the Crawford scale demonstrated union ($p = 0.063$). Conclusion: In our series, more than half of the patients achieved good or excellent results, predominantly characterized by anatomic reduction. No significant associations were identified with variables such as age, affected finger, time from trauma to surgery, fragment size, bone union, or anatomic reduction. These findings suggest that the Ishiguro technique may be effective in the treatment of BMF Albertoni Type C2. ***Level of Evidence III; Retrospective Study.***

Keywords: Fractures, Avulsion; Intra-Articular Fractures; Osteosynthesis, Fracture.

RESUMO

Objetivo: este estudo teve como propósito avaliar os resultados da técnica de Ishiguro no tratamento da deformidade em flexão da articulação interfalângica distal (IFD), conhecida como dedo em martelo ósseo (DMO) do Tipo C2 de Albertoni, visando abordar uma condição comum que pode causar déficit funcional se não tratada adequadamente. Método: foram incluídos todos os casos de pacientes com dedo em martelo ósseo (DMO) do Tipo C2 de Albertoni que foram submetidos ao método de Ishiguro entre março de 2018 e fevereiro de 2023, com acompanhamento mínimo de cinco meses pós-cirurgia; excluídos casos de pacientes que não completaram o acompanhamento mínimo de cinco meses pós-cirurgia. Dados epidemiológicos (idade, sexo, dominância) e informações sobre a fratura (dedo afetado, tamanho do fragmento, tempo entre trauma e cirurgia, tempo de seguimento) foram analisados. Os desfechos incluíram redução anatômica, consolidação, retorno às atividades, dor, déficit de flexão, extensão passiva do dedo afetado, extensão passiva do dedo contralateral e critérios de Crawford. Resultado: nenhuma característica analisada, como idade, dedo afetado, intervalo entre trauma e cirurgia, tamanho do fragmento e consolidação, influenciou significativamente a redução anatômica ($p > 0,05$). A escala de Crawford, assim como a redução anatômica, não apresentou impacto significativo ($p > 0,05$). Contudo, todos os pacientes com resultados positivos na escala de Crawford exibiram consolidação ($p = 0,063$). Conclusão: na nossa série, mais da metade dos pacientes obteve resultados bons ou excelentes, predominantemente caracterizados por redução anatômica. Não foram identificadas relações significativas com variáveis como idade, dedo acometido, tempo entre trauma e cirurgia, tamanho do fragmento, consolidação óssea ou redução anatômica. Estes achados sugerem que a técnica de Ishiguro pode ser eficaz no tratamento do DMO Tipo C2 de Albertoni. ***Nível de Evidência III; Estudo Retrospectivo.***

Descritores: *Fratura Avulsão; Fraturas Intra-articulares*; *Osteossíntese.*

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INTRODUCTION

The flexion deformity of the interphalangeal distal joint (IFD), known as the hammer finger, is common and can cause functional deficit if not properly treated.[1,2] The diagnosis of the deformity is clinical, but radiography is necessary to differentiate between tendon lesions and bone lesions with a fracture of the dorsal margin of the base of the distal phalanges.[3]

Although conservative treatment with thistles shows good results in some cases, the maintenance of the joint step can lead to post-traumatic arthrosis and pain. Thus, when there is compromising at least one third of the joint surface of the distal phalanx or subluxation of IFD[4], there is the need for anatomical reduction, which can be obtained open or closed, followed by fixation with Kirschner wires, pull-out suture,[4,5] tension band,[6] mini interfragmentary screw[7] , or even a hook plate.

The techniques of reduction and open fixation, in general, present some risks, which include infection, nail deformity,[8,9] hypertrophic scar formation,[10] prominence of the synthesized material,[7,9] comminution of the bone fragment during the fixation attempt,[7] etc. Thus, the percutaneous Kirschner wire is frequently employed in fracture osteosynthesis, for which the most diverse configurations have been described.[11-14] Ishiguro et al.[15] introduced a method for closed finger reduction in bony mallet finger (BMF), in which the extension of IFD with Kirschner wire is performed (Figure 1). Since then, some authors have modified the configuration of the wires,[16,17] while retaining the fixation concept of the original technique.[15]

Although it is a technique that has become popular since its publication, because it is considered simple, fast, easily reproducible, and with fewer complications than the open methods,[18] few papers in the literature evaluated the results of Ishiguro et al. Inoue et al. reported success rates of 84% in closed surgical treatments.[19] Lee and Hyun observed that there was no statistical difference in the results when comparing closed and open techniques, but in the open-air reduction group 22% of complications of skin or nail bed, sensitivity at the site of incision, plus longer surgical time were observed.[20]

This work aims to evaluate the results of the technique of Ishiguro et al,[15] using clinical and radiographic parameters, of BMF Type C2 of Albertoni et al.[21]

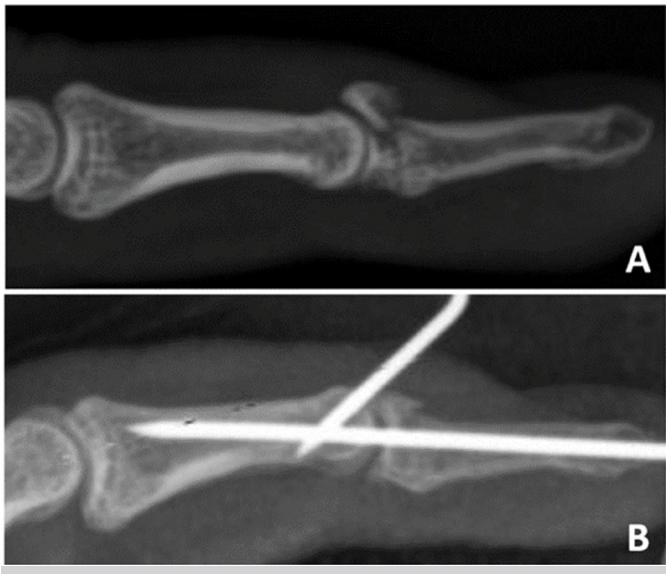

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Figure 1. BMF lateral X-ray (A) and after fixation by Ishiguro method (B).

MATERIALS AND METHODS

For the purposes of this study, 16 fingers were evaluated from 16 patients with BMF Type C2 of Albertoni et al.[21] (Figure 2) treated by Ishiguro et al. Method[15] between March 2018 and February 2023, with a minimum follow-up time of five months after surgery, with the approval of the ethics and research committee (opinion 5.438.893). All participants signed the Free and Informed Consent Clause, with the approval of the Ethics and Research Committee (CAAE 44575121.9.0000.5479)

We evaluated epidemiological data (age, gender, dominance), fractural data (affected finger, fragment size, time between trauma and surgery and follow-up time) (Table 1) and outcomes (anatomical reduction, consolidation, return to activities, pain, flexion deficit, passive and passive extension of the affected finger, passive and passive extension of the corresponding contralateral finger and Crawford criteria.[22] (Figure 3)

A: pure tendon lesion	1: Fall of the distal phalanges < 30°
	2: Fall of the distal phalanx > 30°
B: injury with bone avulsion	1: Fall of the distal phalanges < 30°
	2: Fall of the distal phalanx > 30°
C: Falange base fracture	1: Stable joint
	2: Unstable joint
D: epiphyseal disembarkation	1: Isolated epiphyseal separation
	2: Epiphyseal decay associated with fracture

Figure 2. Classification of Albertoni et al.[21]

Table 1. Descriptive statistics of the variables age, gender, dominance, affected finger, fragment size, time between trauma and surgery, and follow-up time.

Variable	Description (N = 16)
Age (years)	
Average SD	41.7 ± 12.8
median (p25; p75)	44 (33; 47)
Gender, n (%)	
Female	8 (50)
Male	8 (50)
Dominance, n (%)	
Right(a)	16 (100)
Finger, n (%)	
Left index	2 (12.5)
middle left	2 (12.5)
right ring	1 (6.3)
left ring	4 (25)
little right	3 (18.8)
little left	4 (25)
Fragment size (%)	
Average SD	53.8 ± 8.2
median (p25; p75)	53 (50; 60.8)
Time between trauma and surgery (days)	
Average SD	9.6 ± 10.3
median (p25; p75)	7.5 (2.3; 14)
Tracking time (months)	
Average SD	11.1 ± 5.1
median (p25; p75)	11 (6; 13.8)

Qualitative characteristics evaluated in all patients using absolute and relative frequencies were described, and quantitative characteristics were described for all patients using summary measurements (average, standard deviation, median, and quartile).[23]
The good and excellent results, according to the Crawford criteria, were grouped, as well as the bad and regular ones. Still in relation to the Crawford criteria, we calculated the movement deficit of the IFD relative to the counter-side finger. (Figure 4)
Success in reduction and the best result on the Crawford scale were described according to the qualitative characteristics of interest, and the association of these parameters was verified with the use of Fisher Exact Tests or Tests of the Ratio of Probability Similarities. The ages and size of the fragment were described according to the outcomes and compared with the use of non-pairing t-Student tests, since the time between trauma and surgery was described according to the outcomes and compared with the use of Mann-Whitney tests, because the distribution of the data did not present normal distribution.[23] For all variables the odds ratios (OR) not adjusted with the respective intervals were estimated with 95% confidence using simple logistical regressions.[24]
The analysis was performed using the IBM-SPSS *software for Windows* version 22.0, and the data tabulation was performed using the Microsoft Excel 2013 software. The tests were conducted with a significance level of 5%.

Grade	Lost extension	Flexion	Pain
Excellent	None	Total	None
Good	0-10 degrees	Total	None
Regular	10-25 degrees	Some loss	None
Poor	greater than 25 degrees	Some loss	Persistent

Figure 3. Crawford's criteria for the result of the hammer finger treatment.

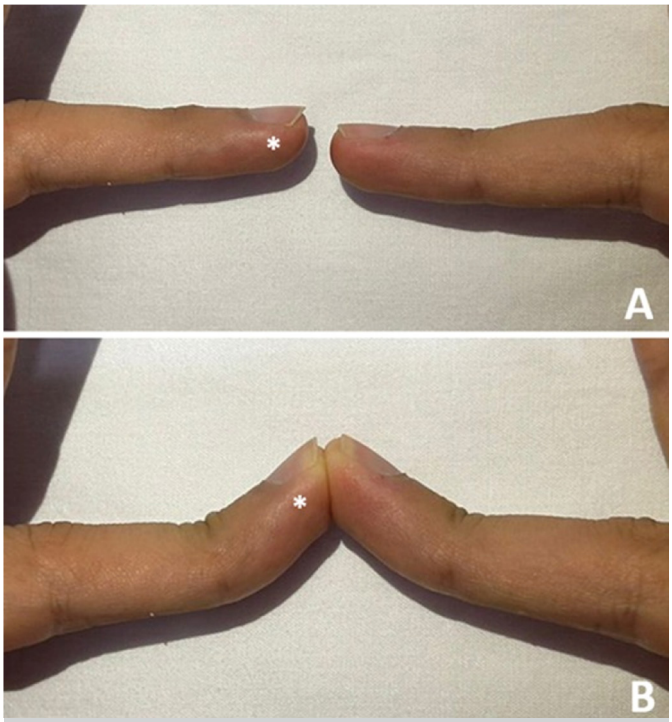

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Figure 4. BMF image after 2 months of rehabilitation, comparing active extension (A) and passive extension (B) of the affected finger (*) with the contralateral finger.

RESULTS

None of the characteristics of interest (age, affected finger, time between trauma and surgery, fragment size, and consolidation) statistically influenced the anatomical reduction ($p > 0.05$), and the results found are described in Table 2.
From Table 3, it is shown that, as well as the anatomical reduction, the Crawford scale did not present a statistically significant influence on any characteristic of interest evaluated ($p > 0.05$), but all patients who presented positive results on the Crawford scale presented consolidation ($p = 0.063$).

DISCUSSION

There is no consensus on whether the anatomical reduction achieved through surgical treatment is essential for the patient to have a good functional recovery. When there is subluxation or fracture affecting more than one-third of the joint surface, most authors suggest that the best treatment is surgical with reduction of the avulsed fragment.[1,4,5,14,16,18,19,25,26] In these cases, the technique of percutaneous fixation with extension blockage is a quick and easy technique. However, there is little information on the long-term functional outcome of patients undergoing the technique described by Ishiguro et al.[15]
In our study, we analyzed the anatomical reduction of the fracture with age, gender, affected finger, time between trauma and surgery, fragment size, and consolidation; and none of the characteristics evaluated presented a relationship with statistical significance.
We believe that gender, dominance, and affected finger could vary due to patient demand for their activities. Male patients in our service tend to perform tasks that require greater physical strength, and female patients, on the other hand, have a demand for tasks that require greater precision. Similarly, the dominant limb and the affected finger influence the representativity that the injury region exerts in the patient's daily work and exercises.
The average age of the patients who presented anatomical reduction was 38.3 years, being lower than the average of the others of 51.8 years. This could suggest that the lower age predisposes to a better postoperative radiographic alignment immediately, and may be due to better bone quality in younger patients, but due to the low number of cases, we cannot confirm such a statement.
Another factor evaluated was the time between the patient's trauma and the surgical fixation. The process of consolidation begins at the initial moment with the formation of a hematoma and subsequent organization in a bone callus.[27] It was believed that the delay in treatment caused the organization of fibrosis and callus in the area of the focus of fracture and the retraction of the tendon[28] to make it difficult to reduce the fragment. However, in this series, we did not find a relationship between the result and the delay in treatment, as one would expect; on the contrary, the patient with the longest time between injury and surgery developed an excellent result.
Taking into account the follow-up time, after removal of the wires, patients needed rehabilitation to improve finger mobility. The minimum follow-up time was five months, which was sufficient to observe the result after stabilizing the possible function gain. On the other hand, we cannot infer whether these lesions will evolve with post-traumatic arthrosis as a late complication, leading to long-term impairment of function.
Because it is a bone lesion, following the principles of the treatment of fractures, when there is joint impairment, anatomical reduction and rigid fixation are necessary.[29] Larger bone fragments are most often better reduced due to ease in intraoperative manipulation; however, the cases with a higher percentage of affected joints were not associated with better reduction. In twelve cases, anatomical reduction was achieved, and in thirteen cases, there was consolidation (Figure 5); however, we did not observe a correlation

Table 2. Description of the anatomical reduction according to the characteristics of interest (age, affected finger, time between trauma and surgery, fragment size, and consolidation) and the result of the unadjusted analyses.

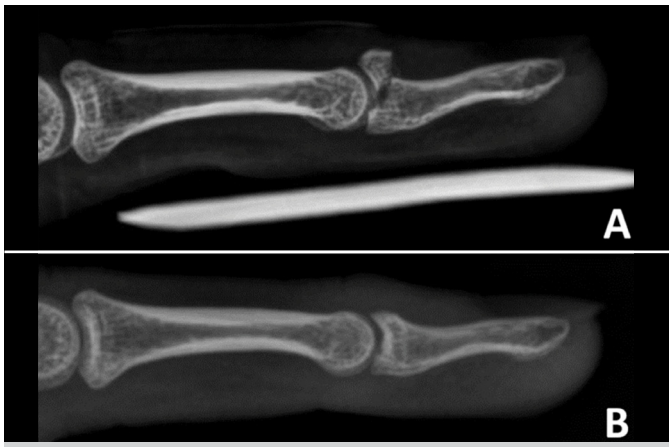
Variable			OR	CI (95%)		p
	No (N = 4)	Yes (N = 12)		Lower	Upper	
Age (years)			0.87	0.73	1.04	0.068**
Average ± SD	51.8 ± 15	38.3 ± 10.7				
Median (p25; p75)	54.5 (36.8; 64)	44 (29.3; 45.5)				
Gender. n (%)						>0.999
Female	2 (25)	6 (75)		1.00		
Male	2 (25)	6 (75)	1.00	0.10	9.61	
Finger. n (%)						0.129#
Index	0 (0)	2 (100)			1.00	
Middle	1 (50)	1 (50)			&	
Ring	0 (0)	5 (100)			&	
Little	3 (42.9)	4 (57.1)			&	
Time between trauma and surgery (days)			1.01	0.90	1.14	0.953£
Average ± SD	8.8 ± 8.3	9.9 ± 11.2				
Median (p25; p75)	7.5 (1.8; 17)	7 (2.3; 14)				
Fragment Size (%)			1.04	0.90	1.20	0.639**
Average ± SD	52 ± 9.2	54.3 ± 8.2				
Median (p25; p75)	53.5 (42.5; 60)	53 (50; 61.5)				
Consolidated. n (%)						0.136
No	2 (66.7)	1 (33.3)	1.00			
Yes	2 (15.4)	11 (84.6)	11.00	0.65	187.17	

Exact Fisher test; #Test of the likelihood ratio; **Unmatched t-Student test; £ Mann-Whitney test; & Can't be estimated.

Table 3. Description of the Crawford scale according to the characteristics of interest (age, affected finger, time between trauma and surgery, fragment size, consolidation, and anatomical reduction) and the result of the unadjusted analyses.

Variable	Crawford		OR	CI (95%)		p
	Bad/Regular (N = 7)	Good/Excellent (N = 9)		Lower	Superior	
Age (years)			0.97	0.89	1.05	0.470**
Average SD	44.4 15.2	39.6 11.1				
median (p25; p75)	44 (39; 54)	44 (28.5; 47)				
Gender. n (%)						>0.999
Female	4 (50)	4 (50)	1.00			
Male	3 (37.5)	5 (62.5)	1.67	0.23	12.22	
Finger. n (%)						0.204#
Index	2 (100)	0 (0)	1.00			
Middle	1 (50)	1 (50)	&			
Ring	1 (20)	4 (80)	&			
Little	3 (42.9)	4 (57.1)	&			
Time between trauma and surgery (days)			1.06	0.93	1.20	0.252£
Average SD	7 8.4	11.7 11.7				
Median (p25; p75)	2 (1; 16)	8 (5.5; 14)				
Fragment Size (%)			0.95	0.84	1.08	0.490**
Average SD	55.4 7.6	52.4 8.9				
Median (p25; p75)	57 (50; 62)	50 (45; 60.5)				
Consolidated. n (%)						0.063
No	3 (100)	0 (0)	1.00			
Yes	4 (30.8)	9 (69.2)	&			
Anatomic reduction. n (%)						>0.999
No	2 (50)	2 (50)	1.00			
Yes	5 (41.7)	7 (58.3)	1.40	0.14	13.57	

Exact Fisher test; #Test of the likelihood ratio; **Unmatched t-Student test; £ Mann-Whitney test; & Can't be estimated.



Source: Author.

Figure 5. Initial lateral radiography (A) and after 3 months of surgery (B).

between these two variables as expected, since two cases that did not present anatomical reduction had consolidation, and one reduced anatomical case did not show consolidation.

The patients with better results, according to the Crawford criteria, classified as good and excellent, all presented consolidation of the fracture. Among these factors, what led our study patients to be classified as lower results was predominantly the deficit of extension, which corroborates that the most statistically significant factor is the consolidation of the fragment. The hammer finger bone fragment is adhered to the terminal extensor tendon,[30] and its non-consolidation suggests that the action of this tendon on the distal phalanges will be impaired. Thus, more than the reduction achieved intraoperatively, it is conceivable that fracture consolidation has greater relevance for long-term functional outcome.
Due to the limited number of cases, more studies are needed to determine which factors would actually have the greatest influence on long-term functional outcomes.

CONCLUSION

We concluded that, in our series, just over half showed good or excellent results, that the majority of cases had anatomical reduction, and that there was no relationship with age, affected finger, time between trauma and surgery, fragment size, presence of bone consolidation, or anatomical reduction.

CONTRIBUTIONS OF THE AUTHORS

Each author personally and significantly contributed to the development of this article: AAV: Conception, conception, drafting of the original draft, design, methodology, visualization, investigation, literature review, analysis and preparation of the manuscript; AGM and SMPA: visualization, investigation, literature review, analysis and preparation of the manuscript; YAA and ACC: Conception, conception, drafting of the original draft, design, methodology, revision and editing, analysis and preparation of the manuscript, recommendation of the journal for publication.

DATA AVAILABILITY DECLARATION

The contents underlying the research are available in the manuscript.

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