

CASE REPORT

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Biomimetic shoes use in a child with epidermolysis bullosa: Case report

Liria A Okai-Nobrega, Patricia S Tossato, Ana Carolina RE Rezende, Jeanine A Magno, Clarissa CS Couto Paz, Ana Paula P Lage, Ana Cristina R Camargos

ABSTRACT

Introduction: Epidermolysis bullosa (EB) is a rare disease with skin fragility that develops blisters with a minimum trauma. Feet blisters are a challenge to develop walking milestone. Shoes are the primary interface between the body and the ground, protecting feet and affecting how ground reaction forces are applied to the foot and transferred to the entire body. The effect of shoes on children's gait is poorly understood, especially in children with EB. The selection of appropriate footwear may help reduce blisters and improve foot function in EB.

Case Report: This case report investigated the use of Biomimetic Shoe over EB's child gait. We tested a commercially available shoe for toddlers that used sand as a model to develop its midsole. As a nature-inspired design, this shoe claims to respect the morphology of

the toddler's feet and provide better conditions for foot development.

Conclusion: Offering walking experiences similar to barefoot conditions improved the child walking pattern, empowering the child to stay mobile and allowing the foot to function as it would naturally.

Keywords: Case report, Child, Epidermolysis bullosa, Shoes

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Liria A Okai-Nobrega^{1,2}, Patricia S Tossato³, Ana Carolina RE Rezende^{1,2}, Jeanine A Magno⁴, Clarissa CS Couto Paz⁵, Ana Paula P Lage¹, Ana Cristina R Camargos^{2,6}

Affiliations: ¹Anamê Science and Technology in Child Health Products, Belo Horizonte, Brazil; ²Physical Therapy Department, Universidade Federal de Minas Gerais, Belo Horizonte, Brazil; ³SOS-EB Kids, New York, USA; ⁴Pediatrics Department, Universidade Regional de Blumenau, Blumenau, Brazil; ⁵Physical Therapy Course, Universidade de Brasília, Brasília, Brazil; ⁶Graduate Program in Rehabilitation Sciences, School of Physical Education, Physical Therapy and Occupational Therapy, Universidade Federal de Minas Gerais, Belo Horizonte, Minas Gerais, Brazil.

Corresponding Author: Liria A Okai-Nobrega, Department of Physical Therapy, EEFETO - Escola de Educação Física, Fisioterapia e Terapia Ocupacional, Av. Presidente Antônio Carlos, Belo Horizonte 6627, Brazil; Email: okainobrega@gmail.com

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INTRODUCTION

Epidermolysis bullosa (EB) comprises a group of rare diseases characterized by skin and mucous membrane fragility, developing a blister from a minimum trauma [1, 2]. The EB classification is complex because mutations in the same gene may be inherited in an autosomal dominant or recessive manner and may result in different clinical phenotypes. However, foot blistering is a common problem in most subtypes of EB [2–4]. The selection of appropriate footwear can help reduce blisters and improve foot function. Evidences that support how to choose adequate footwear is beneficial for EB [2–4]. A dystrophic EB study reported reduced pain on walking in six of the seven subjects

by correcting the foot's balance and eliminating areas of abnormal weight bearing [5]. The biomimetism is the design of nature inspired products [6]. Walking in natural terrains provides a diverse experience that affects the mechanics and energy of locomotion and generates constant adjustments to maintain stability [7–10]. Considering the benefits of walking in natural environments the aim of this case report is observe the effect of a Biomimetic designed Shoes (BS) over the gait of a child with epidermolysis bullosa at the gait development phase.

CASE REPORT

The participant of this study was a 3-year-old girl (13.3 kg and 102 cm) with recessive dystrophic EB confirmed by genetic exam. Her immunologic map showed antibodies against laminin, collagen IV, collagen VIII protein characteristic in RDEB. Hereafter, the child will be called as MFA, her name initials. MFA presented extensive and confluent ulcerations and blister on her foot, covered by exuberant granulation tissue (Figure 1A), with intense walking difficulty.

The patient consent to participate in this case report and whole project received ethical approval from the University's Committee (4.891.224). The child was oriented to use BS every day for at least four hours. MFA was evaluated three times: Mo (Moment 0) before the biomimetic shoe's use, M1 (Moment 1: 12 weeks after the use of biomimetic shoe), and M2 (Moment 2: 33 weeks after the use of biomimetic shoe). One of the authors measured gross motor activities using items of dimensions D (standing) and E (walking, running, and jumping) of Gross Motor Function Measure—version 88-item (GMFM-88) [11].

She was required to walk in two conditions: barefoot (BA) and using biomimetic shoes (BS - Noeh (www.noeh.com.br, Brazil)). The gait bidimensional kinematics parameters were evaluated using software Kinovea (www.kinovea.org) from bidimensional video analysis. Data were analyzed at four gait phases: Initial Contact (IC), MidStance (MS), PreBalance (PB), and MidBalance (MB). Figure 2 shows the lower limb angles. Three strides were considered and exported to be processed under MatLab software (<https://www.mathworks.com/>). The spatiotemporal parameters were: walking speed, stride length, and foot height. Kinematic variables were angular position at hip, knee, and ankle joints.

At age 3, MFA did not present independent walking but she was able to stand up. According to the caregiver, she could only walk supported for a short period of time and do side walk with support. Considering she was not able to give few steps at Mo, it was not possible to measure the kinematic before BS's use.

MFA presented GMFM scores at Mo of 17 D (dimension D) and 14 E (dimension E), M1 21 D and 28 E and M2 29 D and 44 E.

The spatial temporal parameters at M1 and M2 are show in Table 1. By M1 evaluation, MFA walked with support and by M2 she walked independently. At M1, compared to BA, MFA presented a slower walk with BS ($\approx 6\%$) but a fast speed walk at M2 with BS ($\approx 56\%$). Between BAs (M1 and M2) there was also a gait speed increase ($\approx 110\%$). The stride length also showed an increase. At M1, with BS MFA presented 24.4 cm at BA and 26.7 cm using BS, representing around 9% of augment of stride length. At M2 she presented an increasement of stride length of 20% (BA=34.2 and BS=41.4). Between BAs (M1 and M2) she increased the stride length by 40%. The foot height had an increase, comparing to BA, at M1 with BS of $\approx 18\%$ and $\approx 70\%$ at M2. The parent noticed fewer crusts and lower hypersensibility on both feet after the use of BS (Figure 1B).

Table 1 shows the average values of angles. Figure 3 illustrates the biomechanical behavior of these joints in all four situations: M1-BA, M1-BS, M2-BA, and M2-BS.

The upper graphic of Figure 3 illustrates the ankle joint. Observing M1-BA, at IC, ankle presented plantar flexion (angle higher than 90°) in all conditions, more at M1 moment. At MS, MFA started a tendency to dorsiflexion after the use of BS. PreBalance shows a tendency to plantar flexion important to the body impulse forward. At MB, the ankle tended to dorsiflexed specially at M2. With shoes (M2-BS) she tended to a neutral position (90°).



Figure 1: Images of MFA feet plantar surface (A) before and (B) after the use of BS.

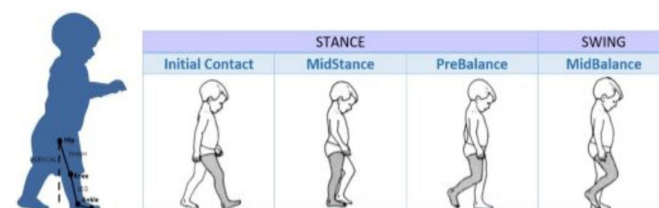


Figure 2: Lower limb angles and the gait phases used to measure the angles (based on Sutherland et al., 1988).

Table 1: Average of spatial temporal and kinematic parameters during gait at M1 and M2. BA corresponds to barefoot and BS corresponds Biomimetic Shoes. M1 (Moment 1: 12 weeks after the use of BS) and M2 (Moment 2: 33 weeks after the use of BS).

Parameter		M1		M2	
		BA	BS	BA	BS
Speed (cm/s)		23.4 ± 4.8	22.0 ± 2.0	49.4 ± 7.0	77.3 ± 5.2
Stride length (cm)		24.4 ± 3.9	26.7 ± 4.7	34.2 ± 3.2	41.4 ± 4.3
Foot height (cm)		7.2 ± 1.1	8.5 ± 0.6	7.0 ± 0.2	11.9 ± 1.2
Gait phase	Joint	M1 (°)		M2 (°)	
		BA	BS	BA	BS
Initial contact 	Ankle	99.2 ± 6.1	99.7 ± 4.0	93.9 ± 2.2	94.5 ± 2.8
	Knee	32.2 ± 6.8	33.2 ± 2.0	32.2 ± 3.3	28.1 ± 2.0
	Hip	30.2 ± 4.7	35.7 ± 0.1	30.7 ± 2.4	26.3 ± 0.9
Mid stance 	Ankle	85.0 ± 4.7	89.6 ± 0.8	84.2 ± 1.6	82.3 ± 1.5
	Knee	34.2 ± 7.3	36.3 ± 3.9	31.0 ± 1.1	35.4 ± 3.1
	Hip	20.1 ± 5.6	23.2 ± 3.5	18.3 ± 0.2	18.5 ± 1.8
Pre-balance 	Ankle	99.6 ± 1.6	97.3 ± 6.2	93.5 ± 4.2	86.2 ± 2.9
	Knee	52.2 ± 3.9	60.8 ± 5.4	52.9 ± 2.3	60.9 ± 1.8
	Hip	16.0 ± 2.0	23.8 ± 1.3	16.4 ± 1.3	16.6 ± 3.1
Mid balance 	Ankle	92.4 ± 3.5	94.7 ± 1.3	87.8 ± 4.1	92.4 ± 3.5
	Knee	58.8 ± 6.4	57.8 ± 3.8	61.0 ± 1.6	80.6 ± 4.4
	Hip	31.6 ± 2.7	34.2 ± 2.8	26.0 ± 4.7	33.6 ± 1.4

The middle and lower graphics of Figure 3 illustrate the knee and hip joints respectively. The shapes of the curves in all conditions are similar. The lower knee angle happens in the beginning of stance phase (IC and MS) but does not reach the extension (near 0°). The higher flexion happens at MB specially at M2, helping to clear off the foot during swing phase. At hip with BS initially (M1-BS), MFA presented the higher values of hip flexion. The M2-BS was more similar to other conditions but presented the lower hip flexion at IC and is one of the highest hip flexions at MB.

DISCUSSION

The use of BS in a child with EB showed changes over her gross motor activities and lower limbs kinematics during gait. Even in barefoot condition (M1 and M2) there were graphical differences, showing better positioning during stance phase and clearance at swing. MFA, in addition to acquiring independent gait, was able to perform important gross motor for her independence.

All these activities influence how the child interacts with the environment. The possibility to have mobility with free hands allows children to explore all the environment and learn [12–14].

Only about one quarter of all children with EB are totally independent in walking [15]. The independent upright walking asks propulsive forces to displace the body forward and different gait strategies [16]. Being able to walk is a motor milestone very important for children with disabilities as EB.

The interest in children's footwear is growing [17]. Biomimetic design provides more protection of the foot, may maintain natural flexibility, and sustain foot arches [18]. Besides that, the development of the morphology of the child's foot, specially at gait acquisition phase has been associated not only because neuromuscular development but also as a confluence of several external factor. This natural development usually does not happen smoothly in children with EB. Blisters, pain, and foot deformations prevent the exposure of these children to stand up and walk postures, impacting the natural development [19].

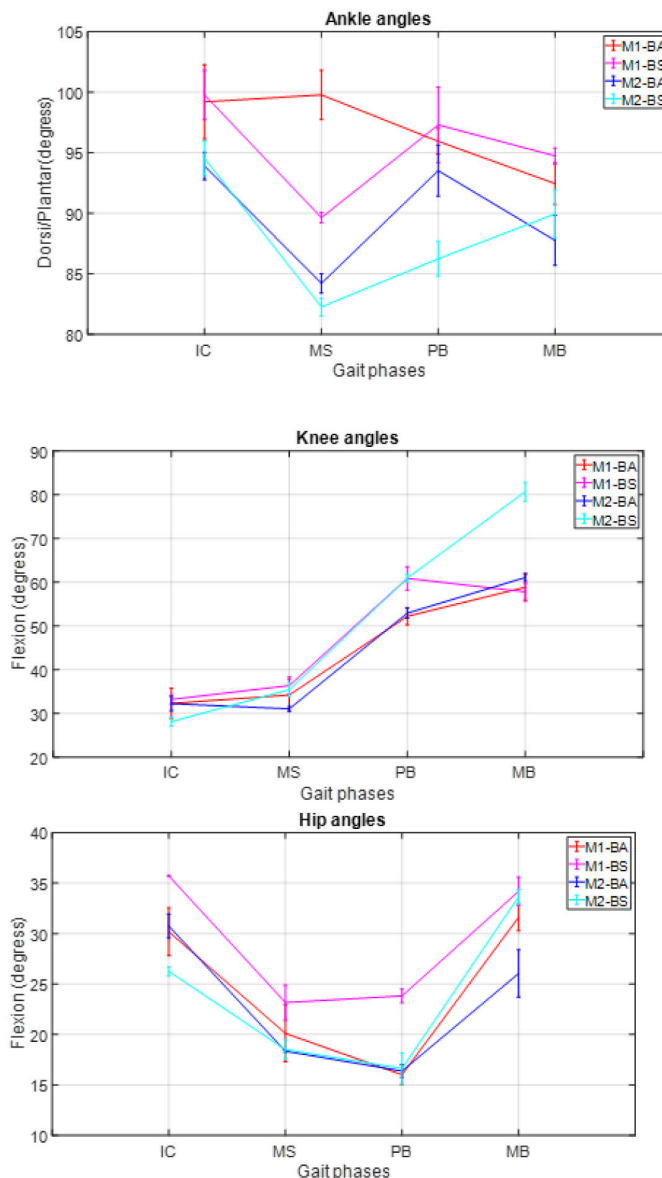


Figure 3: Biomechanical behavior of ankle, knee, and hip joints during specific gait phases and conditions: M1-BA (barefoot—red), M1-BS (Biomimetic Shoes—magenta), M2-BA (dark blue), and M2-BS (light blue). Ankle: more than 90° are plantar flexion and less than 90° are dorsiflexion. The angles of knee and hip represent flexion.

Patient perspective

Before this case report, the caregiver reported difficulties finding a comfortable and safe shoe and the constant fear of causing more pain. The use of BS improved MFA locomotor skills, did not hurt the feet, and gave comfort and security that she was able to walk.

CONCLUSION

The reduced mobility and, eventually, muscular atrophy present in many EB patients can end up in wheelchair use. Preventing these problems with

biomimetic shoes can help children with all subtypes of EB to stay mobile. Movement like walking is an essential development landmark that impacts the emergence of other skills. The environment exploration in a bipedal position improves their quality of life.

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Author Contributions

Liria A Okai-Nobrega – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Patricia S Tossato – Conception of the work, Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ana Carolina RE Rezende – Acquisition of data, Analysis of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Jeanine A Magno – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Clarissa CS Couto Paz – Conception of the work, Design of the work, Interpretation of data, Drafting the work,

Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ana Paula P Lage – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ana Cristina R Camargos – Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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The corresponding author is the guarantor of submission.

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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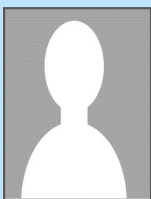
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ABOUT THE AUTHORS

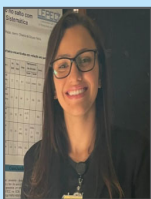
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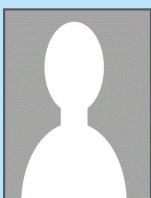
Liria A Okai-Nobrega is a researcher collaborator at Physical Therapy Department, School of Physical Education, Physical Therapy and Occupational Therapy, Universidade de Minas Gerais, and Researcher Associate at Anamê Science and Technology in Child Health Products, both in Belo Horizonte, Brazil. She earned the undergraduate degree in Physical Therapy from Physical Therapy Department, Sao Paulo State University, Brazil and postgraduate degree (PhD) from Neuroscience and Behavior from Neuroscience Department, Psychology Institute, University of Sao Paulo, Brazil. She has published ten research papers in national and international academic journals. Her research interests include motion analysis, biomechanics and gait.
Email: okainobrega@gmail.com



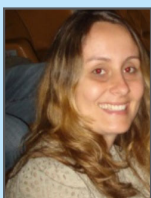
Patricia S Tossato is a dentist graduated from University of São Paulo, Brazil, and holds an Associate Degree in Dental Hygiene from University of Bridgeport, CT, USA. She is a Project Director of an American NGO called SOS EB KIDS, which helps kids with epidermolysis bullosa (EB). Her research interests include epidermolysis bullosa, pathology, and radiology.
Email: paty.tossato@sosebkids.com



Ana Carolina RE Rezende is an undergraduate Student in Physical Therapy Department at Physical Therapy Department, School of Physical Education, Physical Therapy and Occupational Therapy, Universidade de Minas Gerais, Belo Horizonte, Brazil. Her research interests include human movement analysis, biomechanics, physical therapy. She intends to pursue a Master's degree in Rehabilitation Science in future.
Email: anaprojetosufmg@gmail.com



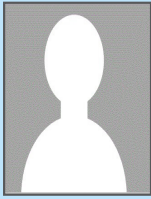
Jeanine A Magno is Professor at Dermatology Department at Fundação Universidade Regional de Blumenau, Brazil. She earned the undergraduate degree in Medicine from Medicine Department, University Federal do Paraná, Brazil and postgraduate degree as Specialist at Pediatric Dermatology from Hospital das Clínicas, University Federal of Paraná, Brazil. She has published (give number – if applicable) research papers in national and international academic journals and authored (give number – if applicable) books. Her research interests include deatology, pediatry and epidermolysis bullosa. He/she intends to pursue (Write future plans – for residents/PhD's/Postdocs) in future.
Email: jeanine.magno@terra.com.br



Clarissa CS Couto Paz is Adjunct Professor at Physical Therapy School, Universidade de Brasília, Brazil. She earned the undergraduate degree Physical Therapy from Physical Therapy Department, Universidade Federal de Minas Gerais, Belo Horizonte, Brazil and postgraduate degree in Neuroscience from Neuroscience Department, Biological Sciences Institute, Universidade de Minas Gerias, Belo Horizonte, Brazil. She has published 29 research papers in national and international academic journals and authored 3 chapters in books. Her research interests include neuroscience, pediatry, motion analysis.
Email: clarissacardososp@gmail.com



Ana Paula P Lage is CEO at Aname Science and Technology in Child Health Products, Belo Horizonte, Brazil. She earned the undergraduate degree in Design from Fashion Design Department, Fundação Mineira de Educação e Cultura, Belo Horizonte, Brazil and postgraduate degree from Design from Design Department, Universidade do Estado de Minas Gerais, Brazil. She has published one research papers in national and international academic journals. Her research interests include design, pediatry.
Email: anapplage@gmail.com



Ana Cristina R Camargos is Adjunct Professor at Physical Therapy and Advisor in Graduate Program in Rehabilitation Sciences, School of Physical Education, Physical Therapy and Occupational Therapy, Universidade Federal de Minas Gerais, Belo Horizonte, Brazil. She earned the undergraduate degree in Physical Therapy from Pontificia Universidade Catolica de Minas Gerais, Belo Horizonte, Brazil and postgraduate degree (PhD) in Rehabilitation Science from Physical Therapy Department, Universidade Federal de Minas Gerais, Brazil. She has published thirty two research papers in national and international academic journals and authored one book. Her research interests include pediatry, physical therapy.

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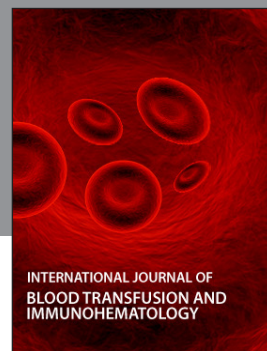
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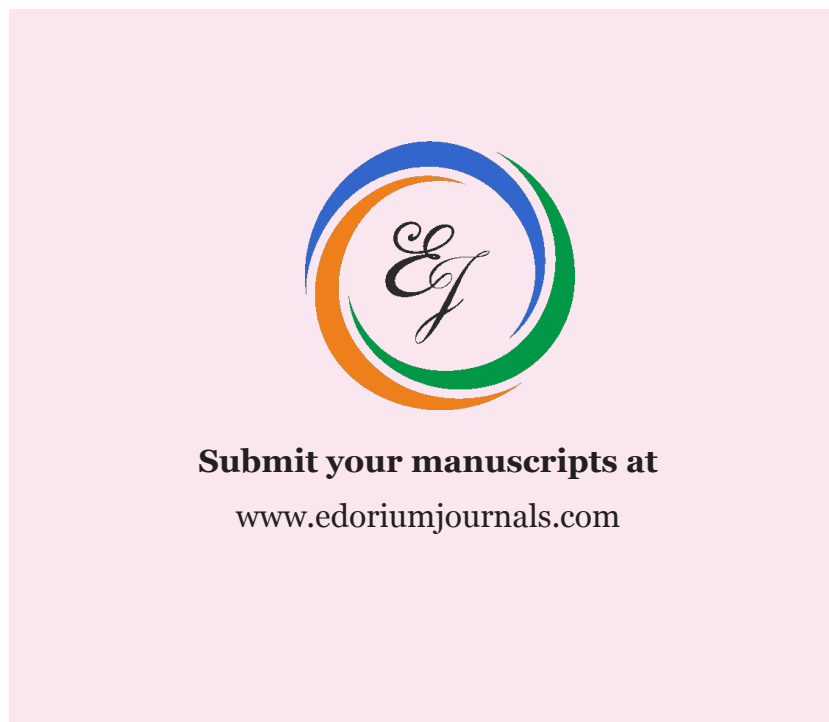
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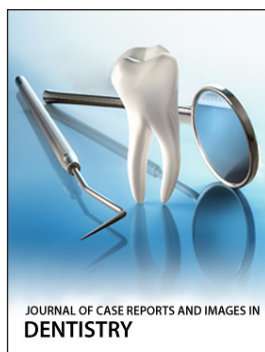
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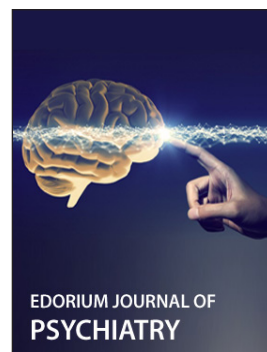
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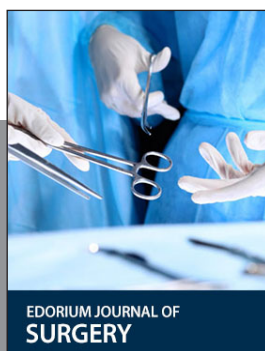
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