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" Assessment Of the Clinical Outcomes of Selective Dorsal Rhizotomy in Treatment of Children with Spastic Cerebral Palsy"

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ABSTRACT:

Spastic cerebral palsy is a prevalent motor disorder that impacts mobility and quality of life. Treatment options, including physiotherapy, medications, and surgery, aim to alleviate spasticity and improve motor function. Selective dorsal rhizotomy is a surgical procedure for children with diplegic spastic cerebral palsy, which reduces spasticity by cutting specific dorsal roots. Effective case selection is crucial for achieving favorable outcomes, as selective dorsal rhizotomy can significantly improve children's quality of life.

Objective: This article aims to evaluate the clinical motor outcome in spastic cerebral palsy children who are treated with selective dorsal rhizotomy.

Methods: In this study, twenty patients underwent selective dorsal rhizotomy. Patient outcomes were assessed preoperatively and compared postoperatively using clinical evaluations of muscle tone (Ashworth scale) along with subjective functional assessments via the Gross Motor Function Classification System.

Results: The majority of patients showed significant improvements following selective dorsal rhizotomy. minor postoperative complications observed and most of them were treated conservatively, and most of patients maintained positive outcomes during follow-up.

Conclusion: Selective dorsal rhizotomy is a safe and effective treatment for children with spastic cerebral palsy that requires a multidisciplinary team for optimal results. Success factors include careful case selection, comprehensive care planning, complication management, and family education on follow-up and physiotherapy.

Key words: Cerebral palsy, selective dorsal rhizotomy, spasticity

Introduction:

Cerebral palsy (CP) occurrence rate is 2-3 in every 1000 livebirths. Spasticity affects 60% of children with cerebral palsy. (Velnar et al., 2019) Spasticity is a motor disorder which could be defined as increase in muscle tone making it resistant to motion. It becomes stiffer and tighter after a specific threshold motion speed or angle is reached. (Sanger et al., 2003) Thus, it interferes with free mobility and hampers the life good quality. (Lieber et al., 2017)

Children with CP experience reduced descending inhibitory input to spinal motor neurons, resulting in increased activity of alpha motor neurons that has stimulatory input through dorsal roots. This imbalance leads to heightened muscle reflexes and spasticity. Selective dorsal rhizotomy (SDR) aims to reduce the stimulation of alpha motor neurons by cutting certain dorsal root sensory fibers, thereby addressing the spasticity associated with CP. (Tubbs et al., 2015) It's very important to treat spasticity in order to decrease muscle tone, reduce pain and deformations. Thus, improve the life quality. (Velnar et al., 2019)

Spasticity could be treated by many options; medical drugs like diazepam and baclofen could be used. Also, baclofen may be injected into CSF space, affected muscles may be injected with botulinum toxin, procedures of peripheral neurectomy; and dorsal nerve roots sectioning. Proper type of treatment is selected based on grade of function, plan of care, and current stage of the child spasticity. (Tubbs et al., 2015)

Assessing the real contribution of spasticity in patient`s disability is the corner stone in choosing patients for selective dorsal rhizotomy, patients must have adequate useful muscle power to get the benefits of the deducted spasticity. Although selective dorsal rhizotomy will lower spasticity, it shall not raise the muscle strength. (Steinbok, 2007) This study assesses the efficacy of SDR in improvement the clinical motor function in children with cerebral palsy.

In this study, clinical motor function was assessed using the gross motor function classification system (GMFCS) and the muscle tone was assessed by the Ashworth scale. GMFCS categorizes the gross motor function of children with CP based on their abilities and limitations in movement. It has five levels, ranging from Level I, where children have the least impairment and are able to walk without limitations, to Level V, where children have severe limitations and require assistance for mobility. The Ashworth scale assesses resistance to passive movement and ranges from 0 (no increase in tone) to 4 (affected parts rigid in flexion or extension). (Rosdiana & Ariestiani, 2021)

Aim of the work:

The aim of the work is to evaluate the efficacy of SDR surgery in improvement clinical motor outcome in children with CP.

Methods:

This prospective interventional study – single arm trial was conducted at Suez Canal University Hospital from July 2023 until December 2024. During this period, total of 20 children (11 males and 9 females) were enrolled in the study. Patients who met the inclusion criteria were offered SDR as treatment option for spasticity. (table 1)

The study protocol was approved by the Research Ethics Committee of the Faculty of Medicine at Port Said University, written informed consents were obtained from all participants families prior to enrollment. The aims of the study, the methods used, possible complications, and other surgical options were reviewed with each patient. Confidentiality was maintained throughout the research, and patients were informed of their right to refuse participation. They were also provided with written consent regarding the use of their images. Additionally, patients had the right to withdraw from the study at any time without jeopardizing their treatment rights or affecting their relationship with their care provider.

Pre-operative assessment:

A detailed medical history was obtained for each participant, especially perinatal circumstances, and details of any previous surgical operations. Degree of spasticity was classified using the Ashworth scale, while GMFCS score was also determined. Imaging studies; MRI of the brain and spinal cord, along with X-rays of the hip and spine were performed to identify any abnormalities that may contraindicate the procedure.

Table (1): Inclusion and exclusion criteria for patient selection

Inclusion Criteria	Exclusion Criteria
• Spastic CP children whose family understand, accept, and agree to goals of care and treatment • Static spasticity or very slowly progressing spasticity • Young patients (ages 3 to 16 years) • Preserved strength in lower extremities • Capable of learning new behaviors • Non-ambulatory patients with severe spasticity that impedes their care	• Individuals with other movement disorders (e.g., dystonia) OR Spasticity in upper limbs that does not affect lower limbs • Age outside the specified range • Inadequate underlying strength • children with idiocy or severe intellectual delays • Long-standing spasticity leading to joint contractures or fixed deformities without potential for orthopedic surgery correction

Operative description:

During the operative procedure, all patients received total intravenous anesthesia (TIVA) with an endotracheal tube, carefully avoiding the use of long-acting muscle relaxants. Neuromonitoring was applied for dermatomes and myotomes from L1 to S2, and each patient was positioned in a prone posture. Intraoperative imaging, including an X-ray of the lumbosacral spine, was performed to accurately identify the level of L1, indicating the expected level of the conus. (figure 1)

We performed our technique derived from Peacock et al. with slight modifications as the following description to maximize the benefit for patients; The surgical approach began with a midline skin incision, followed by muscle separation, and proceeded with a laminotomy from L1 to L5, ensuring good exposure of the dural sac while preserving the facet joints to maintain spinal stability. The posterior roots of the 1st lumbar to the 1st sacral spinal nerves were isolated on both sides. (Peacock et al., 1987).

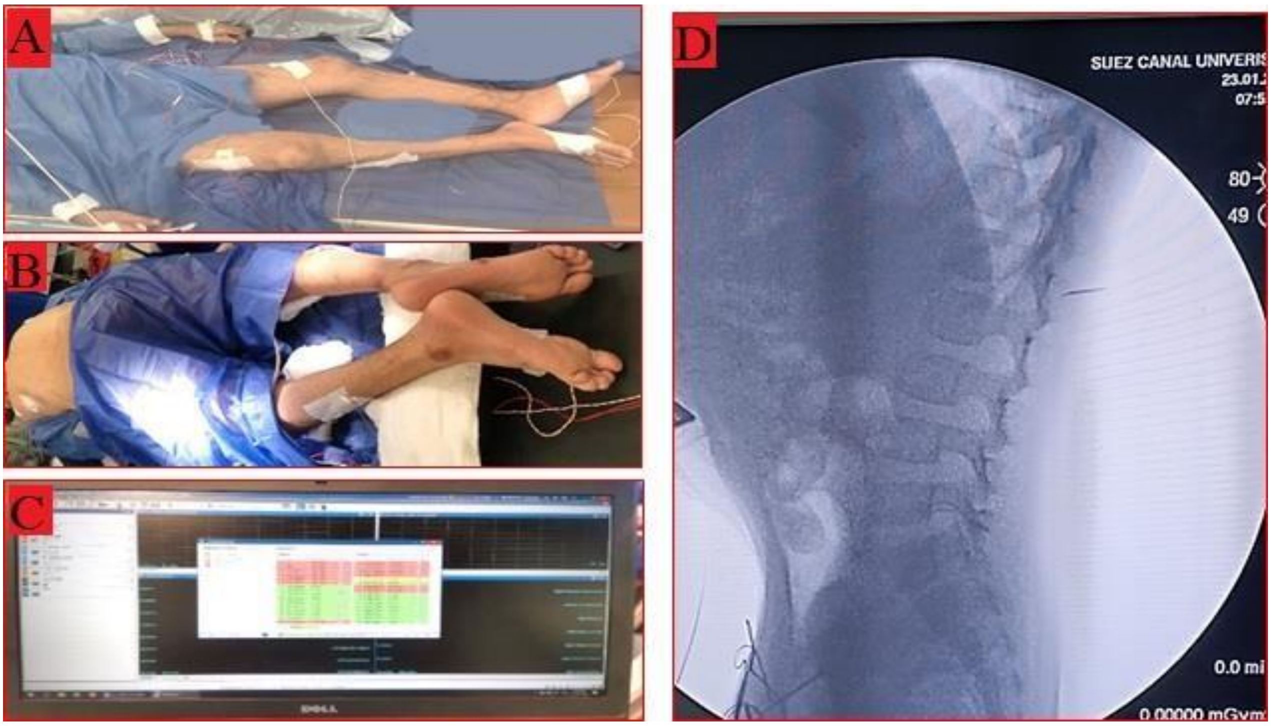


Figure (1): Operative preparations;

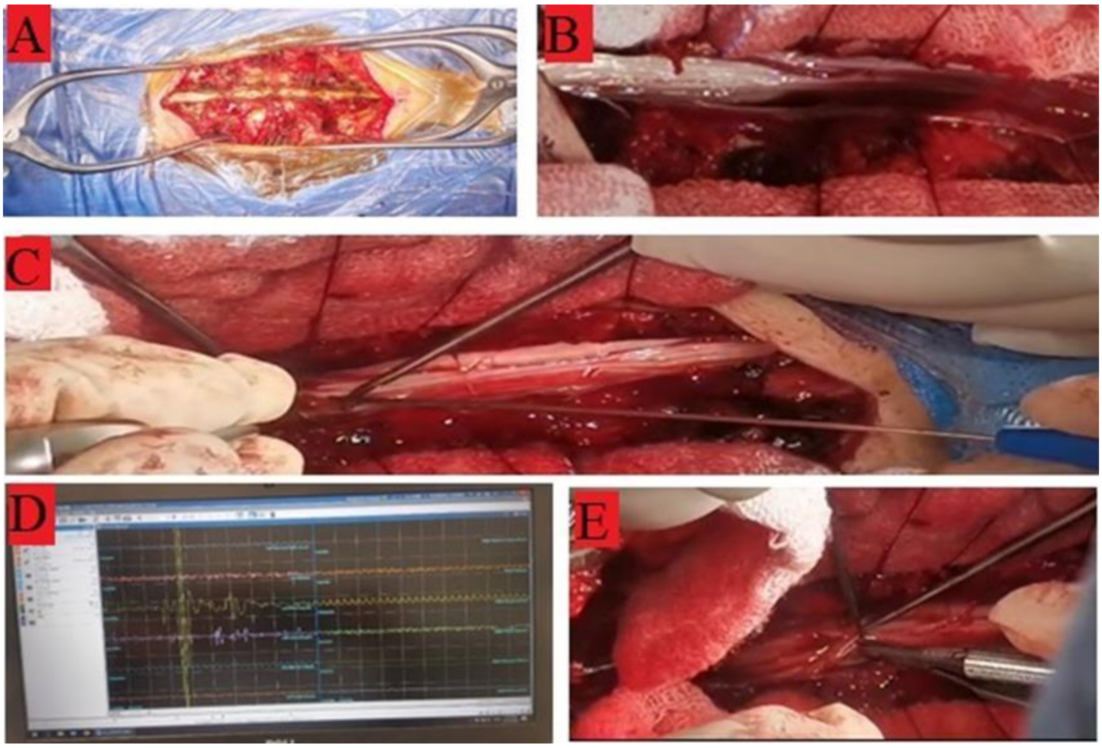
A: Neuromonitoring probes being attached for dermatomes and myotomes, B: Patient in prone position, C: Neuromonitoring being checked pre-operatively, D: Intraoperative x-ray of Lumbosacral spine with a mark identifying L1 level.

Rootlet stimulation occurred to observe muscle responses; roots were confirmed both anatomically through their exit through the foramina of the vertebral segment and by using neurophysiological monitoring. Any posterior rootlet that exhibited low thresholds, leading to sustained muscle contractions or contractions diffusing to unrelated muscle groups, was divided, rootlets with high thresholds, normal muscular distribution, and no diffusion of contractions were left intact. (figure 2)

Special attention was given to the S2 root, which was divided into two fascicles. Pudendal action potentials were evaluated while cutting the part of the fascicle exhibiting lower electrical activity, ensuring minimal harm to the sphincter. The surgical team conducted meticulous hemostasis, sutured the dura, re-placed the lamina, and performed

Figure (2): Intra-operative steps;

A: Muscle separated, B: dura opened, conus exposed with nerve roots, C: Nerve root test using IOM, D: IOM, nerve root testing; 50 Hz Stimulation with Grade 3/4 responses, E: Rootlet fascicle divided after confirmation closure in anatomical layers



Post-operative assessment:

Post-operative assessment focused on evaluating changes in the Ashworth scale for spasticity and the GMFCS score.

Results

The clinical outcome measured was the motor function and was determined by the grade of spasticity using the Ashworth score and also the GMFCS. 20 children (11 males and 9 females) were enrolled in the study, with a mean age of 9.55 years.

The pre-operative assessment of patients' findings using GMFCS and the Ashworth scale. In the GMFCS evaluation, 30% of patients were level 3, 55% were level 4, and 15% level 5. The Ashworth scores for muscle tone indicated that 50% of patients had scores of 3 and 50% scored 4 for the hip joint; the knee joint showed a similar trend with most patients scoring 3 and 30% scoring 4; while for the ankle joint, 50% scored 3 and 50% scored 4. An overall summary of pre-operative motor function demonstrated a mean GMFCS level of 3.85 (SD ± 0.67) and mean Ashworth scores of 3.50 (hip), 3.30 (knee), and 3.50 (ankle), with a total Ashworth mean of 3.43 (SD ± 0.31). 6 months post operative assessment was done; GMFCS level was 3 for 50% of patients, and level 2 for 40% of patients. Most patients have got scores (3 and 2) on the Ashworth scale. The mean GMFCS level was 2.75 (SD ± 0.79) and the mean total Ashworth scores was 2.62 (SD ± 0.56). Means of outcome assessment were compared before and after SDR, we found that the scores significantly improved. (table 2)

Table (2): Association between clinical outcome mean assessment before and after SDR

Clinical outcome assessment	**Pre-SDR**	**6 Months follow up**	Difference in **Mean**	P - value
GMFCS m**ean ± SD**	3.85 ± 0.67	2.75 ± 0.79	1.1	**0.001***
Ashworth m**ean ± SD**	3.43 ± 0.31	2.62 ± 0.56	0.81	**0.001***
Ashworth (hip joint) m**ean ± SD**	3.5 ± 0.51	2.80 ± 0.62	0.7	**0.002***
Ashworth (knee joint) m**ean ± SD**	3.3 ± 0.47	2.6 ± 0.68	0.7	**0.003***
Ashworth (ankle joint) m**ean ± SD**	3.5 ± 0.51	2.45 ± 0.83	1.05	**0.001***

Wilcoxon Signed Ranks Test

****stared are statistically significant***

There was a moderate positive correlation between the change (improvements) in GMFCS levels and the change in Ashworth scores (reduction of spasticity). This correlation is statistically significant (table 3). This suggests that patients who had a greater reduction in Ashworth score (i.e., a larger reduction in spasticity) tended to have a greater reduction in their GMFCS levels (i.e., a larger improvement in gross motor function).

Table (3): Correlation between **changes in** mean measurements in both GMFCS and Ashworth scale before and after SDR

Scoring system	No	Pre-operative ± **SD**	Post-operative ± **SD**	Change in score ± **SD**	Correlation coefficient	P - value
GMFCS	20	3.85 ± 0.67	2.75 ± 0.79	1.10 ± 1.02	0.52	**0.016***
Ashworth scale	20	3.43 ± 0.31	2.62 ± 0.56	0.82 ± 0.66		

* *Spearman's rank correlation coefficient (Spearman's rho)*

*****stared is statistically significant***

5 patients suffered complications; 2 patients of hypoesthesia and 1 patient for wound dehiscence, wound infection (table x), CSF leak each. All were treated conservatively except wound dehiscence who needed surgical debridement & re-closure. (table 4)

Table (4): Postoperative complications (n = 20)

Postoperative Complications	Number	Frequency %
wound dehiscence	1	5%
Wound infection	1	5%
CSF leak	1	5%
Hypoesthesia	2	10%
Total Complicated	5	25 %
No complications	15	75%

There weren`t statistically significant association between either age, cause of CP or gender and the clinical outcome.

Discussion:

Treatment of spasticity in cerebral palsy is a major concern in neurosurgical practice, it is essential to improve the muscle tone and consequently reduce pain and deformities. Although the effectiveness of SDR for treatment of spasticity have been highlighted in previous studies in literature, the safety and effectiveness of this approach has not been confirmed. To our knowledge, this is the first study to be done concerning SDR as a treatment of spasticity in Suez Canal Area.

Selection criteria for eligible patient for SDR surgery remain controversial. (Grunt et al., 2014), (Mittal et al., 2002) However, it was suggested that movement disorders other than spasticity such as ataxia, dystonia and absence of good antigravity strength were reported to be exclusion criteria to SDR. (Buckon et al., 2004)

In this study, none of patients required post operative intensive care place, mean length of stay at hospital postoperative was 4.4 days. Steinbok et al. mentioned that patient to be discharged in the 4th or 5th day postoperative. (Steinbok, 2007) While Park et al. favored the 5th day to discharge patients after SDR. (Park & Johnston, 2006)

We experienced 5 (25%) of patients (4 males and 1 female) presented with hypoesthesia (10%), wound dehiscence, wound infection, and CSF leak (5% each). Most complications were treated conservatively, except for wound dehiscence, which required surgical intervention. Farmer et al. classified complications into; intra operative (bronchospasm and aspiration pneumonia), early and late post operative; (CSF leak and infection, sensory deficits are the major early complications while bladder dysfunction, transient retention, weakness could still happen). Late complications include; spinal deformities and long-term spinal degenerative diseases. (Farmer & Sabbagh, 2007) In this study, no statistically significant association between occurrence of complications and other factors such as gender and cause of CP. Also, there wasn't statistically significant association between complications and improvements happened in the clinical motor functioning.

At 6 months follow-up, the mean change in GMFCS level was 1.1, indicating a reduction or improvement of approximately 28% of baseline scores. We have found a statistically significant improvements in the GMFCS affirming that SDR has a meaningful impact on improving gross motor function. Yet, there wasn`t a strong linear relationship between the patients' motor function status before and after the procedure. Lack of correlation between pre-operative and post-operative scores suggests that the improvements are not merely a continuation of existing patterns but represent a distinct positive change as a result of the intervention. Farmer et al. observed that patients with better preoperative motor function tended to achieve greater postoperative improvements. (Farmer & Sabbagh, 2007) In the study of Tedroff et al. the preoperative motor function did not correlate with the changes in GMF measurements 10 years after SDR. (Tedroff et al., 2011)

Ashworth scale measurements shew similarities with GMFCS in patient improvements. Improvement was 23.6% from the mean base score in the form of reduction of the mean Ashworth by 0.81. Out of the 20 patients in our study, we had 18 patients (90%) showing improvements (reduction of Ashworth score); (55% showing reduction in the mean Ashworth score 1.0 and more, and 35% showing reduction in the mean Ashworth score less than 1.0) and only 2 patients 10% of the study shew worsening of spasticity in the picture of increase the mean Ashworth score. 90% of patients` Ashworth scores were 3 and less after 6 months. Dudley et al. found that long-term outcomes after SDR for spasticity of hip adductors, hamstrings, and ankle plantar flexors as measured by the modified Ashworth score comparing preoperative values to 1-, 5-, 10-, and 15-year follow-up values shew statistically significant improvements as spasticity measurements dropped from (2-3) to below 1. (Dudley et al., 2013)

Considering correlation analysis between changes in mean measurements in both GMFCS and Ashworth scale before and after SDR; Improvements in gross motor function (GMFCS) may be associated with reductions in spasticity (Ashworth Scale). Although, for Wright et al. there was previously some debate about whether the improvement in spasticity will improve the gross motor function, their study clearly suggested that there is significantly greater functional motor improvement at 1 year following spasticity improvements. (Wright et al., 1998) Farmer et al. documented a statistically significant clinical improvement in total Gross Motor Function Measure, conversely, a small group of severely disabled, non-ambulatory patients showed little improvement in motor function, even with effective reduction of lower extremity spasticity. This group was too small for statistical analysis. (Farmer & Sabbagh, 2007) Mittal et al. study found significant improvements in spasticity and functional muscle strength one year after SDR. The Gross Motor Function Measure (GMFM) scores showed progressive improvement over time. The greatest gains were seen in lower-extremity motor function, with mean improvements of 10.1% at 1 year, 19.9% at 3 years, and 34.4% at 5 years compared to the baseline value. (Mittal et al., 2002) With Abou Al-Shaar et al. team, almost all patients with spastic diplegia, have shown improvement from the procedure, observations of patients who received SDR alongside physiotherapy over a follow-up period of 24 months, patients continued to experience ongoing benefits from the treatment. (Abou Al-Shaar et al., 2017) For Dudley et al. groups I, II, and III of the GMFCS exhibited statistically significant improvements at the 5- and 10-year follow-ups compared to their preoperative scores. In contrast, GMFCS group IV showed no notable changes in total GMFM scores from preoperative values at any assessed time point. (Dudley et al., 2013)

In this study, the findings suggest that while age may affect baseline functional status, it does not dictate the outcomes of SDR in terms of GMFCS or spasticity as measured by the Ashworth scale. To Wright et al. there wasn`t any significant differences between groups in the terms of both age and gender.**خطأ! الإشارة المرجعية غير معرّفة.** (Wright et al., 1998) A study by Kim et al. found that children who had a favorable response to SDR, as indicated by improved lower limb tone, motor function, and daily life activities one-year post-procedure, tended to be younger than those with a poor outcome. However, the age at which SDR was performed did not significantly differ between the two groups. (Kim et al., 2006)

The comprehensive analysis across the tables suggests that the cause of CP does not have a significant impact on GMFCS scores or Ashworth Scale measurements before or after SDR in most cases. Grunt et al. mentioned that regarding the cause of spasticity, no studies showed a significant correlation with the outcomes of SDR. (Grunt et al., 2014)

the limited number of cases restricts our ability to draw definitive conclusions, necessitating a cautious interpretation of the results. The follow-up period for the study was relatively short, lasting only six months. During this time, we encountered instances where families did not attend their follow-up appointments, resulting in gaps in communication.

Recommendations:

The findings from this study emphasize the importance of a multidisciplinary team approach in managing spasticity in patients with CP undergoing SDR. With careful patient selection, good surgical practices, implementing a structured postoperative monitoring protocol, conduction of more research on SDR surgeries, equipping hospitals with enhanced tools and resources for performing these procedures, establishing a training program to support talented neurosurgical trainees in gaining expertise in SDR techniques. Adequate intra and post-operative neurophysiological monitoring during follow up, and a specialized rehabilitation center for spastic children and young adult after surgery is recommended. Furthermore, A comparative study between SDR and a recent CAPR (Abdel Ghany et al., 2016) is advised.

Conclusion:

SDR is a relatively safe treatment option for children with spastic CP. Proper selection along with comprehensive and multidisciplinary care team consisting of neurosurgeon, neurophysiologist, physiotherapist, and anesthesiologist is essential for achieving optimal short-term and long-term outcomes. Our research demonstrates that this collaborative approach has resulted in significant functional improvements and objective outcomes for our patients. By sharing our knowledge and experience, we can help a greater number of patients who would benefit from this procedure and ultimately improve their quality of life.

Abbreviations:

CP: Cerebral palsy; SDR: Selective dorsal rhizotomy; CSF: Cerebrospinal fluid; GMFCS: Gross Motor Function Classification System; MRI: Magnetic resonance imaging; TIVA: Total intravenous anesthesia; GMFM: Gross Motor Function Measure; CAPR: Combined anterior and posterior rhizotomy.

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Conflicts of interest:

There are no conflicts of interest.

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