Review

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Alessandra Pantaleo , Alessandra Murri , Giada Cavallaro , Vito Pontillo , Nicola Quaranta *

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Article

Hearing Rehabilitation in Single Side Deafness: CROS vs BCD vs CI

Alessandra Pantaleo [1], Alessandra Murri [1], Giada Cavallaro [2], Vito Pontillo [1] and Nicola Quaranta [1,*]

[1] Otolaryngology Unit, Department of BMS, Neuroscience and Sensory Organs, University of Bari, Bari, Italy

[2] Otolaryngology Unit, Madonna delle Grazie Hospital, Matera, Italy

* Correspondence: nicolaantonioadolfo.quaranta@uniba.it

Abstract: Single-sided deafness (SSD) refers to a condition where there is severe to profound Sensorineural hearing loss in one ear, while the other ear has normal or near-normal hearing. SSD can significantly impact an individual's ability to localize sounds, understand speech in noisy environments, and maintain spatial awareness, often resulting in a reduced quality of life and increased social isolation. In children with unilateral deafness, it can impede spoken language development and potentially impact cognitive abilities. Historically, there was a misconception that the unaffected ear could adequately support speech development in early cases and provide acceptable hearing function in adults. Consequently, SSD was frequently undertreated. However, advances in technology and a deeper understanding of the psychological and developmental impact of SSD have led to a shift in management strategies. Various treatment options are available for addressing SSD, but ongoing debate surrounds the most effective approach. These options include cochlear implants (CI), contra-lateral-routing-of-signal (CROS) devices, and bone conduction devices (BCDs). The aim of this study is to review the scientific evidence on different treatment options for SSD and compare their impact on the quality of life of individuals with this condition.

Keywords: single-sided deafness (SSD); cochlear implants (CI); contra-lateral-routing-of-signal (CROS) devices; bone conduction devices (BCD)

1. Introduction

Single-sided deafness (SSD) is defined as severe to profound Sensorineural hearing loss in one ear and normal or near-normal hearing in the other[1]. However, there is currently no consensus on standardized audiological criteria for defining SSD. According to Van de Heyning et al.[1], SSD is characterized by a pure-tone average (PTA) of ≥70 dB HL in the worst ear, a best ear PTA of ≤30 dB HL, and an interaural threshold gap of ≥40 dB HL In contrast, Ramos Macías et al.[2] suggest that the primary criterion for defining SSD is the lack of improvement in the weaker or "affected" ear with conventional acoustic aid, and the "better" ear having a PTA (0.5, 1, 2, and 4 kHz) of 20 dB HL or higher.

Approximately 1% of the adult population is affected by unilateral deafness, with an estimated incidence of 200 new cases per million per year[3]. On the other hand, the prevalence of single-sided deafness (SSD) in the pediatric population remains unclear due to various factors, including the use of different definitions, diverse methods employed to determine hearing thresholds, and sampled populations that do not fully represent all children at birth or later in life. Historically, the diagnosis of SSD was frequently delayed. According to a study conducted by Everberg et al.[4] in 1960, 52% of cases of unilateral deafness were not identified until after the first year of school, with an average age of 6 years at the time of symptom recognition. However, this situation has evolved with the introduction of universal hearing screening programs. Currently, unilateral deafness occurs in approximately 0.6-0.7 cases per 1000 live births in the United States (according to the Centers for Disease Control Early Hearing Detection and Intervention [CDC] Database). As children reach school age, the number of affected individuals increases, estimated to be between 2.5% to 6%[5–7].

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SSD can result from various etiopathological mechanisms and may vary between pediatric and adult populations. In children early-onset congenital SDD is frequently secondary to cochlear nerve deficiency (cochlear nerve hypoplasia or aplasia) [8], infections such as CMV or meninigitis [9–11] and inner ear abnormalities (e.g., incomplete type I partition, common cavity) [12]. Only a small number of early-onset SDD cases are attributable to genetic causes (e.g., Waardenburg syndrome), which are instead more frequently associated with bilateral sensorineural hearing loss[10] [11]. Some forms of deafness can be attributed to perinatal causes such as jaundice, hypoxia, prematurity, and the use of ototoxic drugs in neonatal intensive care units. Additionally, some cases that initially present as unilateral may progress to bilateral deafness over time. According to a study conducted by Fitzpatrick et al. [15] involving 537 children, 20.1 % were diagnosed with unilateral deafness at birth. Among these cases, 42.2 % experienced a deterioration in their hearing over the years, and in 19 % of the cases, it eventually developed into bilateral deafness.

Idiopathic sudden sensorineural hearing loss (SSNHL) [12,13], chronic otitis media with and without cholesteatoma [18], and cerebellopontine angle tumors [19] are significative causes of SSD among adult patients.

Unilateral deafness (SSD) is often associated with various symptoms that can have a significant impact on a person's daily life. Among these, tinnitus, characterized by the perception of noise without an external source, is a common problem: between 54 and 84 percent of patients with SSD have debilitating tinnitus [20,21]. Hyperacusis, auditory fullness, and dizziness are other symptoms commonly associated with SSD.

Despite being undertreated in the past with the misconception that the unaffected ear was enough for general speech development in early prelingual cases and adequate for acceptable hearing function in adults, it is now widely recognized that the management of unilateral hearing loss is crucial for both children and adults. Single-sided deafness can have significant negative effects on an individual's ability to localize sounds [22–25], understand speech in noisy environments [26–28] and maintain spatial awareness, leading to decreased quality of life and increased social isolation [29–31]. The lack of binaural information and reduced spatial abilities in children, especially in complex sound environments (e.g., classrooms, schools, playgrounds), can result in impaired language, cognitive and learning functions [32].

Diagnosis of unilateral deafness, like its bilateral counterparts, involves several steps. Initially, a detailed medical history must be collected, which should include details of the pattern of onset and progression of hearing loss, any environmental, familial, and genetic risk factors, existing comorbidities, associated symptoms, and any past surgeries or infections. After a thorough objective examination, audiological evaluation should be performed, which includes immittance measurement, pure-tone audiometry, and speech audiometry. Hearing thresholds are measured over frequencies ranging from 125 to 8000 Hz, potentially including frequencies of 3 and 6 kHz, especially in the ear with better hearing. In addition, an assessment of speech intelligibility in noise with tests in noisy environments (e.g., the Italian matrix sentence test) should be done. In the pediatric population, the battery of tests should include age-appropriate behavioral assessments [33]. However, obtaining reliable results from behavioral tests in very young children can be difficult. Therefore, objective assessments of the auditory system, such as brainstem-evoked auditory potentials, are considered the gold standard[34]. In addition, taking a multidisciplinary approach, including assessments of language and communication skills, along with neuropsychiatric evaluations, is essential to ensure a comprehensive understanding of the patient's overall condition and needs.

While various treatment options are available for managing SSD, there is still ongoing debate regarding the most effective approach. Cochlear implants (CI), contra-lateral-routing-of-signal (CROS) devices, and bone conduction devices (BCDs) are possible treatment options.

The aim of this review is to revise the scientific evidence about the different treatment options for SSD and compare the outcomes on the quality of life of people with this condition.

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2. Sound Localization And Speech Perception

Binaural hearing, with the integration of auditory information from both ears, is essential in facilitating spatial awareness and speech perception. The precise and dynamic processing of interaural differences are the main clues for the auditory system to determine the spatial position of sound sources. Differences in the length of the acoustic path from a sound source to the two ears cause an interaural time difference (ITD), while an interaural level difference (ILD) is mainly caused by the acoustic shadow of the head on the contralateral ear[35]. Other advantages of binaural listening are the "binaural loudness summation" [36] and a reduction of competing noise for better speech perception in noise, referred to as the "squelch effect"[37].

Consequently, patients with unilateral profound deafness typically encounter challenges in localizing sound and comprehending speech in noisy environments, resulting in several functional limitations. These include safety risks such as not hearing a vehicle or bicycle approaching from the deaf side, as well as an elevated cognitive load necessary for processing auditory information[38]. This added cognitive effort is particularly evident when attempting to concentrate on a specific speaker or decipher meaning from unclear or ambiguous speech signals. Individuals with Single-Sided Deafness (SSD), in fact, necessitate a 3-10 dB increase in signal intensity to attain the same perceptual improvement as individuals with normal hearing[39]

3. Psychological, Social Consequences, and Impact on Well-being

The literature extensively documents the psychological and social repercussions of unilateral deafness. Hearing loss is linked to increased levels of anxiety, depression, and social isolation [30,40] as decreased self-esteem, especially in environments with background noise, which can lead to feelings of frustration and isolation [31].

Specifically, unilateral deafness causes individuals to feel excluded from conversations involving multiple speakers, prompting withdrawal from social situations among those with SSD [40] resulting in an impact on personal and professional relationships [41,42].

The impact of SSD also extends to overall well-being. Studies have shown that people with unilateral hearing loss are more likely to report poor health, dissatisfaction, and loneliness than those with normal hearing [43,44]. Furthermore, even with the use of hearing aids, patients with SSD often experience a decline in health-related quality of life.[44,45].

4. Rerouting Solutions

4.1. CROS

Conventional approaches to hearing rehabilitation designed for Single-Sided Deafness (SSD) typically encompass rerouting the auditory signal from the affected ear to the unaffected or better-functioning ear to facilitate further processing. Contralateral Routing of Signal (CROS) devices provide a non-surgical approach and represent the least invasive solution currently available. CROS hearing aids involve a microphone and transmitter in a hearing aid worn on the impaired ear, which transmits sound to a receiver in a hearing aid worn on normal hearing ear [46] determining a pseudo-bilateral hearing. In cases of asymmetric hearing loss (sensorineural, conductive or mixed), the aid on the better hearing ear can also provide amplification in addition to the CROS input, creating a configuration known as Bilateral Contralateral Routing Of Signals (BiCROS). BiCROS is typically recommended for individuals with mild to moderate hearing loss in the better hearing ear [47]. Recently it is proposed a new technology for subjects with SSD, suffering from bothersome tinnitus in the poorer ear in addition to difficulties on speech in noise and sound localization. The new device combines the ability to reroute the sound from the poorer ear to the good ear (CROS system) while still stimulating bilaterally with conventional amplification (StereoBiCros). This strategy seems to reduce tinnitus handicap and loudness for subjects with AHL/SSD/ and tinnitus.[48] The underlaying mechanism of the positive effect of StereoBiCros on tinnitus is unclear; probably it is due to the

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acoustic masking of tinnitus produced by the acoustic stimulation of the poorer ear. Furthermore this stimulation could reverse the tinnitus-related central plasticity[49].

4.2. Bone Conduction Devices (BCD): Surgically Implanted Devices

Bone Conduction devices (BCD) function similarly to CROS devices and transmit signals from the ear with SSD to the better ear through bone conduction bypassing the air-conduction pathway [50]. Since the late 1970s in were implanted the first bone-anchored hearing aids (BAHAs), several improvements and variations have been introduced. Surgically implanted bone conduction devices transform sound waves into mechanical vibrations through direct skull contact, facilitating transmission to the inner ear and encompass percutaneous bone conduction implants (pBCI) and transcutaneous bone conduction implants (tBCI).

Transcutaneous bone conduction implants and percutaneous bone conduction implants share essential components, including a titanium screw surgically implanted into the mastoid bone and an external processor[51]. The main difference between these two implants lies in how the external processor connects to the implant. Percutaneous bone conduction implants typically use an external pillar to attach the processor (e.g., Oticon Pronto and Cochlear™ Baha® Connect), whereas transcutaneous bone conduction implants utilize magnets to pair the speech processor with a fully subcutaneous abutment that transmits sound to an internal transducer (e.g Cochlear™ Baha® Attract and Medtronic Alpha 2 MPO ePlus™).

Reported complications of pBCI include skin overgrowth, granulation tissue formation, keloids, infections, and other soft tissue issues[52]. Transcutaneous systems were developed to address cosmetic and skin issues associated with percutaneous devices, though they can still cause discomfort due to the magnetic force[53]. Reducing magnet pressure and limiting device usage can alleviate symptoms. However, excessive pressure may lead to skin necrosis if it surpasses capillary pressure[54]

To maximize the benefits of both percutaneous and transcutaneous devices, active transcutaneous BCIs like the BonebridgeTM (MED-EL, Innsbruck, Austria) and The CochlearTM Osia system were development. These devices feature an external component equipped with a sound processor featuring dual microphones and a magnetic network with an external coil for transmitting the transcutaneous signal. The internal component, which is surgically implanted and osseointegrated into the mastoid, includes a magnetic receiver that directly stimulates the bone conduction implant [55]. Active transcutaneous BCIs like the BoneBridgeTM are associated with fewer skin complications when compared to pBCI, primarily because they lack a percutaneous stump[56].

Evidence suggests that rerouting devices are successful at reducing the negative effects of the acoustic head-shadow and enhance the awareness of sound and the Signal-to-Noise Ratio (SNR)[57] [58] when sounds are directed toward the affected ear, but are not successful in restoring access to binaural cues [59,60].

In a review of four controlled studies, Baguley et al. [61] uncovered limited evidence supporting the efficacy of BCD for treating Single-Sided Deafness (SSD). However, they did observe that BCD offered advantages over both CROS devices and the unassisted condition, particularly in terms of speech discrimination in noisy environments and subjective questionnaire-based assessments of hearing ability (Abbreviated Profile of Hearing Aid Benefit APHAB questionnaire). Agterberg et al. [44] suggest that BCD neither improves nor worsens the localization abilities of patients with SSD. Similarly, no significant differences were found between CROS and BCD in terms of speech perception in noisy environments and sound localization in the study by Peters et al. [62]. This is in contrast to previous studies that instead reported better sound localization for SSD listeners with a BCD [45] or a conventional CROS hearing aid[46].

4.3. Bone Conduction Devices (BCD): Extrinsic Devices

Non-implantable bone conduction hearing devices are also available, which transmit sound vibrations through the intact skin to which they are attached by means of bands, soft bands,

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adhesives, and glasses. The main advantage of these devices is that they do not require surgery for their application. However, these devices experience signal attenuation, especially at high frequencies, and their time of use may be limited because of the force required to secure them in place[63].

Non-implantable bone conduction devices include the adhesive bone conduction device (ADHEAR) and the dental device (SoundBite), designed for patients with unilateral deafness or conductive hearing loss with a bone conduction THA better than or equal to 25 dB HL.

ADHEAR, developed by MED-EL[64], is a bone-conduction hearing solution known for its distinctive adhesive attachment method. Specifically, the device is affixed to the skin over the mastoid bone using a specialized adhesive, ensuring secure placement without causing discomfort or pressure-related issues. In a study by Mertens et al.[65], 17 SSD patients were involved in a prospective randomized crossover study comparing an adhesive hearing system with a CROS hearing aid. Group A started with the adhesive device, and Group B with the control device, followed by a crossover test after 2 weeks. The results showed that 70% of SSD participants found the adhesive system partially useful or better, with satisfaction levels similar to those using the control device according to the Audio Processor Satisfaction Questionnaire (APSQ). While sound localization improved with the adhesive system, there was no significant improvement in speech perception in noisy environments.

SoundBite developed by Sonitus Technologies, consists of a behind-the-ear microphone (BTE) placed in the damaged ear, which captures sound that is then processed by digital audio equipment inside the BTE. Additionally, there is a removable device inside the mouth (ITM), specially designed to fit comfortably in the upper back teeth, which receives the signals processed by the BTE device, converting them into vibrations, which stimulate the cochlea and allow the user to perceive the sound. According to the study conducted by Lou et al. [66] on nine patients with SSD, SoundBite has been shown to lead to improved speech recognition and overall quality of life in both quiet and noisy environments.

5. Cochlear Implants (CI)

Cochlear Implant (CI) is a surgically implanted electronic device that contains an array of electrodes which is placed into the cochlea, and stimulates the cochlear nerve, bypassing the injured parts of the inner ear. Initially suggested as a treatment for severe tinnitus in adults with single-sided deafness (SSD), cochlear implant provision and rehabilitation has now become the clinical standard for SSD. Effectively, CIs are the only solution that can restore binaural input to the auditory pathways.

Several studies have reported that CI in individuals with SSD provides significant improvement in speech perception, tinnitus control, localization, and QoL [67,68]. A recent systematic review conducted by Idriss et al. [69] that evaluated the effectiveness of cochlear implants in treating SSDs with severe tinnitus found that CIs are associated with high tinnitus suppression scores suggesting that CI can indeed be a viable option for patients with SSD who also experience disabling tinnitus.

A Dutch randomized controlled trial [70] involving 120 participants compared the short-term outcomes of IC with those of BCD, CROS aids and no intervention. The IC group showed improvements in speech perception in noise and sound localization, as well as improvements in tinnitus burden on the TQ and THI questionnaires. All treatment options improved disease specific QoL on most subscales of the questionnaires used.

Comparison of localization accuracy in patients with SSD between the conditions of unassisted aid, CI, BCD and CROS conducted by Arndt et [71], showed significantly lower localization error for patients treated with CI compared with the other treatment options.

The national, multi-centre, prospective study by Marx et al. [72], indicated that CROS hearing aids yielded inferior outcomes when compared to cochlear implants (CIs) in the treatment of single-sided deafness (SSD) or asymmetric hearing loss (AHL). The study found that cochlear implants were associated with significantly improved quality of life, especially among individuals with severe tinnitus combined with profound hearing loss, especially when trials of CROS and bone-anchored hearing systems (BAHS) proved to be ineffective.

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Advanced age is not a contraindication for cochlear implants (CIs), which have minor and major surgical risks and complications overlapping those of younger adults[73–75]. Several studies have shown a clear negative correlation between duration of deafness (DoD) and cochlear implant (CI) performance in individuals with postlingual deafness. Specifically, prolonged periods of auditory deprivation are linked to worse speech perception outcomes, attributed to neural degeneration and cross-modal plasticity[76,77]. However, Rader et al.[78] argue that duration of deafness (DoD) should not be considered a barrier to cochlear implantation (CI) in adults with unilateral postlingual deafness. Their study involving 36 adults with postlingual deafness revealed promising results in auditory rehabilitation for individuals with a self-reported duration of deafness of less than 400 months. For those with a longer duration, success is limited but still possible. These findings align with those of Nassiri et al.[79]., who found no disparity in hearing success among patients with unilateral deafness (SSD) with CI, regardless of whether their deafness lasted more or less than 10 years

6. SSD in Pediatric Population

Auditory deprivation resulting from monaural sensory input in children with unilateral deafness (SSD) may have a significant impact on the development of auditory pathways and brain networks associated with higher-order cognitive functions [80] [81]. Specifically, there is evidence of an "auditory preference syndrome" in which the developing auditory pathway in SSD children reorganizes with a dominance toward the NH ear and a weaker central representation of the hearing-impaired ear, involving spatial processing areas [82,83] This leads in SSD children to the loss of binaural advantages such as the binaural squelch effect[84] [85], the head shadow effect,[86] [87] and the binaural redundancy effect[88,89]. Overall, this results in greater difficulty in speech perception in noise and localization of sounds. It has been shown that unilateral deafness has implications in language development, cognition, and quality of life, with greater listening-related fatigue in children and difficulties in school learning compared with normal hearing children[82,90,91].

In addition to the auditory cortex, other studies have found reduced activation of networks deputed to attention in children with SSD, as well as changes in brain networks involved in executive functions, IQ and language comprehension. Children with SSD aged 9 to 14 years have reduced accuracy and efficiency in phonological processing and appear to have impaired executive control function[92]. Confirming this, MRI with diffusion tensor imaging (DTI) technique also shows non-integrity in auditory and associative nonauditory areas during the performance of executive functions in children with SSD compared with normo-hearing children of the same age[93]. Indeed, it has been hypothesized that children with unilateral deafness have different patterns of functional connectivity responsible for auditory and executive functions, which may thus explain behavioral and educational difficulties. Confirmation to this hypothesis comes from Resting-state functional connectivity MRI (rs-fcMRI) studies: in the cortical networks of executive functions of children with unilateral deafness, there are several areas of adaptive i.e., strengthened functional cortical networks and other maladaptive i.e., weakened areas with lack of predefined suppression of cortical networks[94]. These findings provide a possible explanation for the educational difficulties experienced by children with unilateral hearing loss. Although studies are few and there is bias in enrollment and etiology, there thus seems to be a direct link between unilateral deafness and cognitive development. Quality of life also appears to be impaired in children and adolescents with SSD in the domains related to school performance and social interactions compared with normals[95].

To address the challenges posed by unilateral deafness by enhancing communication skills, and to support educational progress and improve the overall quality of life of children with SSD, various therapeutic strategies have been proposed, including redirection technologies (CROS, BCD) and cochlear implants (CIs).

In children redirection technologies [96] such as CROS require the ability to handle the device and manage the surrounding environment to avoid transmission of weak signals from the deaf side to the ear with better hearing, as well as adequate ear canal dimensions to accommodate the device and prevent obstruction of the better ear. Bone conduction surgical devices (BCDs) are often not

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available for children under the age of 5 in many jurisdictions or states. Most importantly, both technologies do not facilitate binaural hearing because they stimulate only one auditory pathway. Therefore, they are generally not recommended in children with SSD[97].

Indeed, the only therapeutic option that can re-establish bilateral auditory stimulation is cochlear implantation. In 2019, the FDA approved the MED-EL CI in patients with SSD having a maximum of 10 years of hearing deprivation and a minimum age of 5 years. Nevertheless, as suggested by Park et al. [97] in the "American Alliance Task Force Guidelines for Cochlear Implantation in Children with Unilateral Deafness," the use of these devices at an early age may be advantageous, as neural plasticity is most significant during the early years of life [98]. Polonenko et al. [99] demonstrated rapid improvement in cortical reactivity, measured through electroencephalogram, after a few months of device use in children who received an implant before 3.6 years of age. Conversely, brain reorganization in response to SSD might impede central binaural integration after cochlear implantation, potentially as early as 2 years after the onset of HL [100], [101], [102]. Several studies showed worse outcomes in CI children with congenital or prolonged deafness SSD[82,103,104]. Furthermore, a recent systematic review and meta-analysis conducted by Benchetrit et al.[105] which included 12 observational studies evaluating 119 children, showed that most children had clinically significant improvements in speech perception, both in noisy and quiet environments, and in sound localization. A shorter duration of deafness was associated with more substantial improvements in auditory qualities of speech and space, emphasizing the urgent need to lower the current age limit for cochlear implant application in children with SSD. Since almost 50% of congenital SSD are due to inner ear and auditory nerve malformations, eligible patients should have a normal cochlear nerve[106]. In addition, special attention should be given to children with single-sided deafness (SSD) who are at risk of progressive hearing loss in their better ear. This includes children with conditions like cytomegalovirus infections and cochlear malformations. In such cases, as previously mentioned to minimize the duration of hearing deprivation and maximize the potential benefits of the implantation process, it is crucial to prioritize the implantation procedure before deterioration in the better ear occurs. Finally in the decision-making process for implantation, careful consideration must be given to the duration and etiology of deafness, as well as identifying the needs and goals of the family. Family counseling is critical to explain the risks and consequences associated with SSD and evaluate possible treatment options. [62].

7. Conclusions

SSD is a challenging condition that significantly affects an individual's life, from the ability to communicate effectively to overall well-being. Among the various treatment options, cochlear implants have emerged as a very promising solution that can significantly improve speech perception, sound localization, and quality of life. Importantly, age and duration of deafness are no longer a barrier, as cochlear implants demonstrate substantial improvements even in older adults and those with prolonged periods of SSD. In children, early cochlear implantation is critical, as it improves speech perception, especially in noisy environments, and promotes rapid adaptation to binaural hearing due to the neuroplasticity of the young brain.

Effective management of SSD involves comprehensive assessments, early intervention, and ongoing support, with a focus on children at risk for progressive hearing loss in the better ear. Family counseling and individualized interventions are essential. Although cochlear implants have become the gold standard, individualized assessment and intervention remain critical to ensure the best outcomes for both adults and children with SSD.

Ongoing research and technological advances in hearing rehabilitation offer hope for transformative solutions that can provide a fulfilling life with restored hearing function and improved quality of life for people with unilateral deafness.

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