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# Parental experience of hypoglossal nerve stimulator implantation in adolescents with Down Syndrome and obstructive sleep apnea

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## ABSTRACT

**Purpose:** The purpose of this study is to explore the experience of parents in deciding whether to participate in a clinical trial of the insertion of the Hypoglossal Nerve Stimulator (HNS) to treat their adolescent with Down Syndrome (DS) and Obstructive Sleep Apnea (OSA).

**Design and methods:** A qualitative descriptive design with interviews was used to gather parental experiences from those who consented to HNS for their adolescent with DS and OSA. Interviews were conducted, audiotaped, and transcribed. Basic content analysis was followed to interpret the data. Using a process of data debriefing/engagement, codes were generated, and field/reflective notes were used to assure trustworthiness of the data.

**Results:** Parents, 13 mothers/2 fathers, participated. Three themes were identified: Parents experience desperation about acceptance of standard of care for their adolescent with DS and OSA. This desperation led parents to seek information/insights from social media and they came to rely on those sites to explore options, ultimately leading them to HNS clinical trial. Finally, parents had a desire to share experience with HNS implantation.

**Conclusions:** Parents described being desperate at acceptance of standards of care for OSA. They shifted reliance on experts and parents by searching social media pages to explore options for treatment of OSA. Parents wished to share experiences with the HNS implantation.

**Practice implications:** Nurses play a role in preparing for the HNS by instructing/educating parents. Nurses can identify supportive social media sites for parents during the HNS decision and suggest ways to measure outcomes of HNS.

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Down syndrome (DS) is the most commonly occurring chromosomal disorder, affecting approximately 50,000 adolescents in the United States (Presson et al., 2013). The Republic of Ireland reports the highest number in Europe with 1 in 444 live babies with DS and in the United States it is estimated at 1 in 1499 live births (De Graaf et al., 2017; Loane et al., 2013; Ni She & Filan, 2014). It is accompanied by declining learning capacity, speech and language delay and impairment in cognition as well as a myriad of other medical conditions that can have adverse outcomes on quality of life (Nadel, 2003).

Obstructive sleep apnea (OSA) is one of the most commonly occurring of these other conditions. It affects greater than 60% of adolescents with DS and is confirmed by abnormal sleep studies (Hoffmire et al., 2014). Children and adolescents with DS have a higher prevalence of OSA due to anatomical abnormalities that contribute to airway

obstruction. These abnormalities include macroglossia, adenotonsillar hypertrophy and midface hypoplasia, each placing them at risk for upper airway obstruction (Maris et al., 2016). OSA occurs due to airway obstruction that is caused by repeated episodes of decreased airflow, hypoxemia, and hypercapnia. Lack of treatment for OSA can result in cardiovascular problems, learning disabilities, behavioral problems, excessive day time sleepiness and increased morbidity (Hsieh et al., 2019; Nation & Brigger, 2017).

Adenotonsillectomy is the primary treatment for OSA in children and adolescents and has been shown to provide therapeutic benefit in those with tonsillar hypertrophy (Shott et al., 2006). However, after adenotonsillectomy approximately 50% of children and adolescents with DS will continue to experience disrupted sleep requiring positive pressure airway support with continuous positive airway pressure (CPAP) or bi-level positive airway pressure for persistent airway obstruction (Diercks et al., 2016). Unfortunately, although these treatments are effective, they are often poorly tolerated (Diercks et al., 2016). There are also other surgical therapies for those individuals who cannot tolerate positive airway pressure. However, these

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procedures, which include tongue-based surgery, oral maxillofacial surgery, and tracheostomy, have varying degrees of risk (Diercks et al., 2017) that may be unacceptable.

The hypoglossal nerve stimulator (HNS) is a novel approach that was introduced for treatment of adults with OSA that was first described by Schwartz et al. (2001). The HNS is an implanted device that generates electrical impulses to stimulate the protrusion of the tongue, which is timed to inspiration. The device was found to decrease obstructive apneic and hypopneic events in adults without disrupting sleep, but many technical problems occurred, and no further studies were undertaken for the next ten years (Eastwood et al., 2011). With resolution of technical problems, the HNS has been recently proven to be safe and effective for treatment of adults with OSA and prospective studies demonstrate that HNS is well tolerated (Strollo et al., 2014).

A clinical trial to evaluate the safety and efficacy of the HNS in adolescents and young adults with DS and OSA was approved in August 2016 (A Pilot Study to Evaluate the Hypoglossal Nerve Stimulator in Adolescents with Down Syndrome and Obstructive Sleep Apnea (NCT02344108)). For the first few years, as parents were deciding whether to participate in the trial, little was known about the experience of adolescents with DS who had the HNS inserted to treat OSA to help parents make an informed decision. This study aims to explore the experience of parents in deciding whether to participate in this clinical trial.

### Protection of human subjects

Prior to commencement of this study, institutional review board (IRB) approval was obtained. Confidentiality of the participants was assured by using pseudonyms in the collection and reporting of the findings. All findings were kept on a password protected, encrypted computer to which only the study team had access. Privacy was assured by interviewing participants at a time and place that allowed them to be away from others who could hear the conversation.

### Study design

The investigators used an open ended qualitative descriptive design with a semi-structured interview guide. This is the method of choice when little is known about a topic and straight description of a phenomenon is needed (Sandelowski, 2000). The interview guide was developed by the investigators. The questions that were used were: How did you hear about the Hypoglossal Nerve Stimulator as a possible treatment option for your adolescent? What, if any, concerns did you have about the Hypoglossal Nerve Stimulator? If you were concerned, what information and who did you seek to have your questions answered? If you spoke with another family whose child had gone through the procedure before, was this helpful? And have you noticed any changes in your adolescent now that the HNS has been implanted?

### Reflectivity

In qualitative research, it is important to state up front how and why the research team is interested in a topic. In the setting where one member of the team was employed, a pilot study was initiated to assess the safety and efficacy of the HNS in adolescents with Down Syndrome and severe OSA (Diercks et al. 2016). As part of the initial engagement with the pediatric nurse supporting the clinical trial, parents asked many questions but also shared about how they came to find out about the study and how this supplemental information was informing their decision whether to participate in the trial. Rather than rely on these insights, a formal study team was formed to interview as many parents as possible regarding how they came to make their decision.

The investigators interviewed parents of the first 15 trial participants that had HNS inserted from 2017 and 2018.

### Data collection procedure

The participants recruited for this study had enrolled their adolescent with DS into a study in which 15 adolescents received a HNS device at a tertiary care setting in the northeast United States. The nurse caring for the adolescent who had the HNS device implanted approached the parents to ask if they were willing to participate in an interview about their experience. If the parents agreed to be interviewed, the PI obtained verbal consent as indicated by the IRB. The interviews were conducted by telephone and recorded digitally. The recordings were transcribed verbatim.

### Data analysis

Basic content analysis as described by Hsieh and Shannon (2005) was followed to interpret the descriptive data. We used a process that consisted of reading and re-reading of the data to assure accuracy against the recordings. Field notes were recorded during this process and included comments about parents' emotions, tone of voice, pauses during the interview process as well as the investigator's thoughts and reflections. After reviewing the data, the team noted words and phrases that were repeatedly used and organized these into meaning units or codes. In re-reading the data, the codes/meaning units were considered in light of the context of the ideas being conveyed by parents. This allowed the team to collapse the codes into categories. The team reflected on these categories and developed these categories into themes that captured the overall gestalt of the experience.

### Results

Parents, 13 mothers and 2 fathers, participated in the interviews. The parents were predominately Caucasian, and one parent was Hispanic. They were from multiple states throughout the United States. Their adolescent, 6 females/8 males, mean age 16.4 years, were the first adolescents to have the HNS procedure in the United States. Many of these adolescents had tried alternative treatments including having their tonsil and adenoids removed, tongue reduction, unsuccessful trials of Continuous Positive Airway Pressure (CPAP), and some had tracheostomies.

Interviews lasted 20–45 min. The interview guide was easily understood by parents and the questions as posed did not require re-wording. Three themes were identified from the data:

1. Parents experience desperation about the acceptance of the current standard of care (e.g., adenotonsillectomy, continuous airway pressure, or tracheostomy) for their adolescent with DS.
2. This desperation led many parents to seek out information and insights from social media (e.g., Facebook sites for parents of children with DS) and came to rely on those sites to explore options, ultimately leading them to awareness of the clinical trial.
3. Parents want to share their experience and educate others about the benefits of HNS implantation.

#### Theme 1

Parents experience desperation about the acceptance of the current standard of care (e.g., adenotonsillectomy, continuous airway pressure, or tracheostomy) for their adolescent with DS.

With continuing and escalating problems with sleep disturbances in their adolescent with DS, parents expressed increasing concern after trying the standard options. *'We had gotten to the point where our son wasn't sleeping past 2 in the morning, so we needed to do something'. 'There were times when she could have rocked the house with her snoring'. Parents were also observing regression in cognitive development. One parent shared that their adolescent 'was experiencing regression in behaviors that she, I mean, she had cognitive language delays that were actual regressions from where she had been in a year or 2 before'.*

Primary care physicians typically respond to parental concerns referred the parents to an ear, nose, and throat (ENT) specialist. The ENT provider ordered more sophisticated testing including a sleep study. For some parents, the sleep study was their first exposure to actual concrete data to identify the severity of their adolescent's sleep issues and the dangers of OSA. *'Every night...he was choking on his tongue every three minutes'*. Sleep studies confirmed or identified significant OSA. One parent learned for the first time that the OSA was severe enough that it could require, *'...surgical intervention up to including a tracheostomy'*. Based on the sleep study results, the parents pursued the suggested series of options to treat their adolescent's sleep apnea, voicing concern that each treatment step led to more invasive options. These included pallet expanders, removal of tonsils and adenoids, tongue reduction surgery, use of a CPAP mask, and permanent tracheostomy.

Some parents, after consulting with their physician, had already arrived at a conclusion that a tracheostomy, the last resort, was necessary. Parents that had experience with a tracheostomy actively searched for less invasive options. *'I would do anything to get this trach out at the time, because it was, like 11 years, dealing with the trach.'* *'We tried the CPAP machine with her, four times without success'*. Still other parents lacked access to physicians that had experience treating children and adolescents with DS and OSA.

Parents shared that having a child with OSA that persisted over years had severe impacts on both their child from sleep deprivation (development, behavioral, etc.) and on the entire family. Parents were also sleep deprived and faced with making choices among care options that did not solve this fundamental problem. The invasive character of the available options contributed to the concerns that they already faced having an adolescent with DS. Parents reported adverse impacts on siblings. These parents had reached a point of desperation and thus began to look at HNS implantation as a viable option.

It should be noted that many of the parents that identified HNS as an option and subsequently verified that their adolescent met the criteria to participate in the HNS study, had the financial means to participate in the clinical trial including adequate health insurance and the ability to incur out-of-pocket expenses for travel. However, this was not always the case. There were out of pocket expenses that strained some family resources for travel to the tertiary care facility for the HNS insertion.

### Theme 2

This desperation led many parents to seek out information and insights from social media (e.g., Facebook sites for parents of children with DS) and came to rely on those sites to explore options, ultimately leading them to awareness of the clinical trial.

Several parents learned about the HNS from attendance at DS conferences or physicians with a specialty in DS or with an ear, nose, and throat specialty. For some parents, this was their first exposure to the HNS and awareness of the pilot study testing its use in adolescents with DS and OSA. These parents followed up with their existing health care providers. Currently, many health care providers were not aware that HNS was an option for adolescents. *'We certainly talked to the doctors at the clinic and the doctor at her pediatricians, you know just getting perspective'*.

A few parents turned to their extended family when there was a health professional in that group. *'I first heard about it (HNS) through my brother ...a pulmonologist'*. Physicians were also learning about the HNS option by conducting their own research and subsequently informing parents of their patients with DS. *'My child's physician referred us. We were so desperate'*. Parents then reached out to the Ear, Nose and Throat (ENT) specialist that was performing the HNS at this time.

However, many desperate parents, hoping to find another option to these invasive treatments for OSA, turned to social media as either their first stop or as a supplement to more traditional sources of information. For parents enrolling later in the clinical trial after approximately five

patients had the HNS implanted, social media and internet-hosted websites was the principal source of information. These sources including the website of the manufacturer of the HNS (Inspire Medical Systems Inc), a DS physician's social media site that discussed the HNS, and a closed Facebook page established by a parent of an adolescent with DS who had the HNS implanted. Another parent created a video of her adolescent with DS demonstrating how the adolescent managed his HNS device and describing how it helped him sleep. These two parents indicated that they were concerned about health equity as it related to access to information about HNS. Parents of adolescents with DS that were considering an HNS were able to show this video to their adolescent.

*'So, we did do our research, printed up a lot of information, and we did call another family who had HNS done, and they raved about how well their son was doing after getting it. There are no DS physicians in my state...so I posted to social media, which all these people follow me..., and someone in Boston posted about HNS on a Facebook page'. 'Yeah, it was very helpful... we spoke with the parents, but we spoke with the adolescent too', 'I reached out to... two parents that I got information from that...made me feel confident'*.

### Theme 3

Parents want to share their experience and educate others about the benefits of HNS implantation.

Once adolescents meet the criteria for participation in the clinical trial, the HNS is implanted during a one-day hospital stay followed by a period where the adolescent, parents, and medical team worked together to fine-tune the stimulator setting to address the experience of the adolescent. After this period, parents begin to understand that what they had accepted as "normal" was in fact, not normal. Parents started to note changes that they were seeing in their adolescent. As first, it was that their adolescent was sleeping! The insertion of the HNS was described as a 'game changer'. *'She is getting oxygen all night for the first time in her life'*. Over time, parents began to notice other changes such as speech, cognition, and behavioral improvements.

Parents shared observations regarding changes in their adolescent's sleep pattern, speech, and behavior. These observations informed the identification of the need for further investigation leading to more objective measures that could be used by other specialists (e.g., speech and language therapists and neuropsychologists) to measure the outcome of the HNS.

First, the parents noted changes in sleep patterns. Before HNS, parents recounted that their nightly routine was interrupted sleep to get up to place CPAP mask back on their adolescent, checking that oxygen source was still on, and getting a smaller number of hours of sleep.

After the HNS, adolescents with DS were sleeping without breathing assistance and oxygen and were easier to get upon the morning. *'She sleeps through the night now', 'The sleep study after the HNS averaged 4 episodes of sleep apnea down from 45 apnea episodes per hour before', she's sleeping well', 'He is waking up easily and happily and his blankets are still tucked around him', 'it's easier to get her up'*.

Parent also noticed speech, academic skills/language, and behavioral changes over and above the improved sleep patterns. Parents communicated observed changes in speech. *'She was talking for 10 minutes. And you know, prior to surgery she used only two to three words', 'Even the tone in the muscles in his face are improving', 'he is trying to say more words...and the words he is using are clear'*. Speech therapists at school were also noting changes in the adolescent after HNS.

There were noted changes that occurred both at home and at school by the parents and the teachers. *'You know, doing so much better academically. I just get reports from school; she does her work, a lot'. 'I think in terms of attention and ability to, to power through and do schoolwork and things like that is really where we see the biggest change', 'Teachers all say he is ...improving scholastically...even with a decrease in his ADHD medication by 50%', 'Their IQs come up a bit'*.



Parents offered comments noting improvements in emotions and behavior. *'Yep, he's happier, when we walk in his room he smiles and gives us a hug and he's much quicker to do what we ask him to do and happier to do what we ask him to do, 'he able to go swimming', 'she has OCD... the rituals she would do, those have definitely improved', 'he is less moody and irritable'.*

Once again, these parents turned to social media to share their experiences, leading to personal phone contact between parents that were trying to make a decision to have the HNS implanted, and these early trailblazers. Many of them became mentors to new parents.

### Trustworthiness

The standards of Lincoln and Guba (1985) were used to establish trustworthiness of the data. The standards are credibility, dependability, confirmability, and transferability. Credibility was established by using an iterative process that supported prolonged engagement with the data. Codes, categories, and themes were identified, reviewed, and revised as needed to assure they captured the overall parental experience. To assure dependability, an audit trail was maintained to record ideas, reflections, interviews and coding decisions. Confirmability was assured by each member of the team being involved in the analysis, recognizing, and documenting personal biases. This allowed us to question our interpretations of the data and reach consensus on the findings to assure that they represented the parents' voices. Transferability was achieved through member checking the themes with several of the parents who participated in these interviews who agreed that these findings did reflect their experiences.

### Discussion

This is the first study to report on the parent's experience of participating in a clinical trial of the insertion of the HNS for OSA in their adolescent with DS. The diagnosis and treatment of OSA in adolescents with DS is important since OSA has been implicated as a factor in the development of pulmonary hypertension, cardiovascular complications, and behavioral problems (Brooks et al., 2015). Worried about escalating symptoms of OSA, the parents looked to their adolescent's pediatrician for more information, assessment of severity and an examination of potential treatments.

Sleep problems in adolescents with DS have been reported in up to 60% in population-based samples, however 66% of parents of children and adolescents with DS did not perceive that their child/adolescent had trouble sleeping. This suggests that sleep problems maybe under-recognized by parents and not brought to the attention of primary care physicians (Diskin et al., 2020; Hoffmire et al., 2014). Symptoms such as snoring, daytime fatigue, bedtime resistance, and behavioral changes were problems reported by parents of children with sleep disordered breathing without DS (Boss et al., 2017). These symptoms were not always recognized by those parents of adolescents with DS (Maris et al., 2016). Therefore, support for the supplementation and/or revision of health care guidelines to include broader screening of sleep in children and adolescents is needed to decrease the impact of sleep abnormalities.

The use of the diagnostic sleep study, polysomnography, to define the degree of OSA in adolescents with DS is recommended by the American Academy of Pediatrics (Bull and Committee on Genetics, 2011). This test informs parents of the severity of their adolescent's OSA and offers details of the changes in physical and behavior health that could be attributed to OSA. When parents are provided this information regarding their child or adolescents sleep problems and the risk associated with OSA, they face the systemic barriers within the episodic care model of trying to access specialists, making decisions on treatment, and finding social and financial services for support (Currie & Szabo, 2020).

In children and adolescents with Autism, sleep problems are highly prevalent and are related more to biopsychosocial etiologies than anatomical abnormalities that are seen in DS. The parents of children with Autism are faced with multiple pharmacologic and behavioral options and recommendation, most without evidence, to treat their child's sleep problems (Carnett et al., 2019; Owens, Rosen, Mindell, Kirchner, 2010). In contrast, parents of children and adolescents with DS are faced with risky surgical interventions or use of difficult devices to treat OSA in children and adolescents with DS (Diercks et al., 2017).

Parents of children and adolescents have traditionally relied on their pediatricians and other medical professionals to advise and direct care. This has been a source of inequity with respect to knowledge of alternative and particularly, novel treatments, including clinical trials. A pediatrician with a significant population of patients with DS improves the odds that parents will learn about a novel treatment or clinical treatment during a doctor's visit (O'Neill et al., 2018).

Parent populations of all races, genders, and income levels have become accustomed to looking to the internet to start a research effort. In recent years, Facebook pages devoted to rare diseases have become prevalent. When one combine's desperation with access and expediency (speed and ease of gathering information), it should come as no surprise that Facebook pages devoted to children with DS have proliferated. These pages make it easy to ask open-ended (has anyone found an answer to sleep deprivation for OSA) and more precise questions (has anyone heard of Dr. "X" at hospital "Y")? This often leads to the opportunity for parents to connect directly offline and via the telephone if they reside in different parts of the country. Parents remain responsible for assessing the quality of information that they receive but they are used to performing this function with respect to a wide variety of internet searches. Social media does not fully address equity issues since many families in the United States do not have access to the internet in their home.

Research on the use of social media for medical answers is consistent with the HNS experience. A review examined the use of digital media to obtain health information. The researchers concluded that parents are supplementing information from printed materials and health care providers with digital sources increasing the use of social media to inform health care decisions for their children and adolescents (Frey et al., 2022). Fathers, in particular, perceive digital media as a good source of health information (Jaks et al., 2019).

We found that connecting parents during decision-making to other parents and their adolescent who had agreed to insertion of the HNS was a useful technique. The overwhelming majority of parents were happy to talk and share their experience and offered their unique perspective. A video of an adolescent speaking about how he turned the HNS on and off and how the HNS improved his sleeping is available for prospective parents to share with their adolescents. With guidance from the nurse, parents were able to address the needs of parents during the shared decision-making process involving the parents, the adolescent with DS, and the ENT specialists.

The contribution of parent mentors in sharing their lived experience with parents of children with diabetes demonstrated that the parent mentors were empowered by serving in this role (Sullivan-Bolyai & Lee, 2011). A subsequent study found that parents that help others help themselves by helping them accept the complex diagnosis of their own child (DeHoff et al., 2016).

The parent insights have led to the development of a larger study to evaluate speech, behavior, and changes in learning as proxy measures of the effectiveness of the HNS, *Effects of hypoglossal nerve stimulation on cognition and language in Down Syndrome* (5UO1DC029379–02).

### Practice implications

Parents describe the decision-making with considerable thoughtfulness due to the lack of experience applying this procedure to their adolescent, the experience of others, and the outcomes of the implantation

of HNS. Nurses, both inpatient and outpatient, recognize parental and adolescent needs can play an important role in preparing the parents and the adolescent with DS for this novel therapy. For example, nurses can point parents to social media sites about DS and HNS.

Health information resources about the HNS need to be simple and be available in multiple formats (e.g., videos) to enable revisiting and downloading as part of the journey to a decision to pursue HNS insertion in adolescents. Health literacy needs to support comprehension and recall of risks, benefits, and alternative treatments for OSA in those challenged with medical terminology. Parental decision aids that provide simplified information have been shown to increase knowledge of options and outcomes (King & Moulton, 2006).

We found that connecting parents during decision-making to other parents and their adolescent who had agreed to insertion of the HNS was a useful technique. Some parents and adolescents after HNS were happy to talk and share their experience and offered their unique perspective. With guidance from the nurse, parents were able to address the needs of parents during the shared decision-making process involving the parents, the adolescent with DS, and the ear, nose the throat specialists.

The nurse can suggest, besides sleeping, other ways to assess the measurement of outcomes after HNS. Assessments by parents, teachers, and others that the adolescent interacts with can provide additional factors that may be improved by better sleep. Speech, cognition, mood, and behavior are areas that can measure the impact of the HNS that parents are able to assess.

The most recent evidence from the HNS clinical trial (42 subjects at multiple sites) reveals significant improvements in sleep as measured by polysomnogram, that the procedure is safe and sustainable (no patients required removal of the device or experienced longstanding morbidity post-operatively (Caloway et al., 2020; Yu et al., 2022)). This suggests that prospective parents and their health care providers will have more evidence to rely on in the future when making their decision regarding the HNS for their adolescent with DS and OSA.

Finally, having determined how valuable information is to the parents of adolescents with DS and OSA, it is appropriate to identify ways for nurses, nurse practitioners, health centers, pediatric practices, and family medicine practices to work individually and together to spread awareness of HNS as a viable option.

### Limitations

There are limitations to this study. The parents were identified from a clinical trial that had only a small number of actual adolescents with DS that received the HNS. The sample was predominately Caucasian females. Due to the lack of diversity in the sample, the findings are limited. In addition, this precludes us from assessing equity issues including the access of low-resourced families to ENT referrals or other resources that will identify HNS as a viable option.

### Conclusions

The Hypoglossal Nerve Stimulator offers a less invasive and effective alternative for parents that are desperate to address sleep apnea for their adolescent with DS and OSA. Many parents became aware of this new treatment through social media sites for families with children and adolescents with DS and came to rely on those sites to explore options, ultimately leading them to awareness of the clinical trial. After insertion of the HNS, parents reported their observations to study investigators regarding improvements in sleep pattern, speech, and behavior. Nurses can play a pivotal role in making parents aware of HNS, educating, and supporting them in decision making for the HNS for their adolescent with DS and OSA.

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### CRedit authorship contribution statement

**Kevin Callans:** Conceptualization, Formal analysis, Resources, Visualization, Writing – review & editing. **Diane L. Carroll:** Conceptualization, Data curation, Formal analysis, Methodology, Supervision, Validation, Visualization, Writing – review & editing. **Annette McDonough:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Writing – original draft.

### Declaration of Competing Interest

The authors declare no conflict of interest.

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