

FAMILY EDUCATION STRATEGIES IN CARING FOR CHILDREN WITH CONGENITAL ADRENAL HYPERPLASIA (CAH): A LITERATURE REVIEW

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Abstract

Families having children with congenital adrenal hyperplasia (CAH) may have low knowledge and awareness in some aspects of CAH management. Information and education regarding caring for children with CAH is essential for families so that they can provide proper care and treatment to their children. This study aims to describe the strategy of family education about caring for children with CAH from the existing literature. A literature review was performed by investigating relevant studies from online databases of Google Scholar, Science Direct, PubMed, and BioMed Central. Keywords of "education" and "children" and "congenital adrenal hyperplasia" and "family or parents" were used in searching the data. The results of the review showed various strategies that can be applied to provide education to families, especially parents. These strategies include providing detailed care instructions about adrenal crisis management, encouraging families/ parents to use positive and useful coping strategies, increasing knowledge and awareness on the use of medical alert IDs, providing Psychosocial Education Program (PEP), increasing nurses' knowledge and comfort in providing education to families, and suggestions for improving the provision of family education. The provision of education to families or parents having children with CAH should consider some individual aspects such as emotional states, health literacy, learning styles, the amount of information, and knowledge assessment of each family/parent.

Keywords: Congenital adrenal hyperplasia, education, family

INTRODUCTION

Congenital adrenal hyperplasia (CAH) is a chronic inherited disorder with an autosomal recessive mode of inheritance. This disorder causes a deficiency in one of the five enzymes needed to synthesize cortisol in adrenal glands (Dhyani et al., 2016). The incidence of classic CAH ranges from 1:10,000 to 1:20,000 births (Speiser et al., 2018). CAH leading to adrenal crises can occur both in male and female infants and is potentially life-threatening. Hormonal imbalances due to adrenal crises can cause electrolyte imbalance, dehydration,

vomiting, cardiac arrest, and shock (Utari, 2017).

A study by Bhakri and Jain on 28 parents having children with CAH found a lack of knowledge and awareness among parents in some aspects of CAH management. The results showed that 14.28% of respondents did not realize adjustments needed during life, and two respondents considered short life expectancy and difficulties for children with CAH. There was a misunderstanding about the use of steroid substitutes as routine drugs (6 of 28; 21.42%) and the risk of addiction due to long-term use (5 of 28; 17.8%). Twelve out of 28 (42.8%)

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respondents did not know the most appropriate reasons for administering increased glucocorticoid doses during stressful conditions. Six respondents (21.42%) perceived that regular visits to the doctor for continuous monitoring were not important for children with CAH (Bhakhri & Jain, 2011).

Caregivers or parents having children with chronic diseases experience significant psychological distress, as well as financial and social burdens. While medical and non-medical costs contribute to financial problems, families may also experience disturbances in their daily routines, altered leisure times, and changes in family dynamics (Khanna et al., 2015).

Timely medical treatment, counseling, and education are essential for families having children with CAH. Medical treatment involves the replacement of glucocorticoid and mineralocorticoid hormones. Clinical management of CAH has been described as "difficulty in balancing hyperandrogenism and hypercortisolism" (Speiser et al., 2018). Parents of female children may have difficulty in determining appropriate genital surgery due to genital abnormalities in children.

Lim and Zebrack stated that the concept of normalization in families with chronic diseases is realized by changing lifestyles that support treatment process. Management of CAH requires high vigilance and self-efficacy of caregivers in the administration of drugs before an adrenal crisis occurs. In newborns who suffer from CAH, it is expected that parents understand routine medical management immediately and during emergency situation so that they can care for their children at home. Therefore, adequate knowledge and understanding are essential in maintaining life for families in general and become a vital

component for medical management and within families (I Mitchelhill et al., 2013).

Caregivers or families receiving adequate and comprehensive knowledge may have an opportunity to adjust their lives and make the illness a part of everyday life, and hence they can reduce psychological burdens and instill a sense of control in the management of CAH (I Mitchelhill et al., 2013). Research findings indicate the necessity to provide information and education about CAH, as well as resources that can be used by families so that patients can be taken care of appropriately by a multidisciplinary team (Bhakhri & Jain, 2011). This study aimed to describe the provision of health education to families in caring for children with congenital adrenal hyperplasia through a literature review.

METHODS

This study is a literature review that searched for published articles from the online databases of Google Scholar, Science Direct, PubMed, and BioMed Central. Keywords of "education" and "children" and "congenital adrenal hyperplasia" and "family or parents" were used in the searching process. The inclusion criteria were articles published within the last ten years (2010-2020), relevant to the research question, original research studies, and involved families having children with CAH or nurses caring for children with CAH as subjects. The selection of the articles is presented in Figure 1.

RESULTS

The categorization of research papers was performed through the preparation of paper synthesis matrix. Eight articles that met the criteria was included in this review, as presented in Table 1.

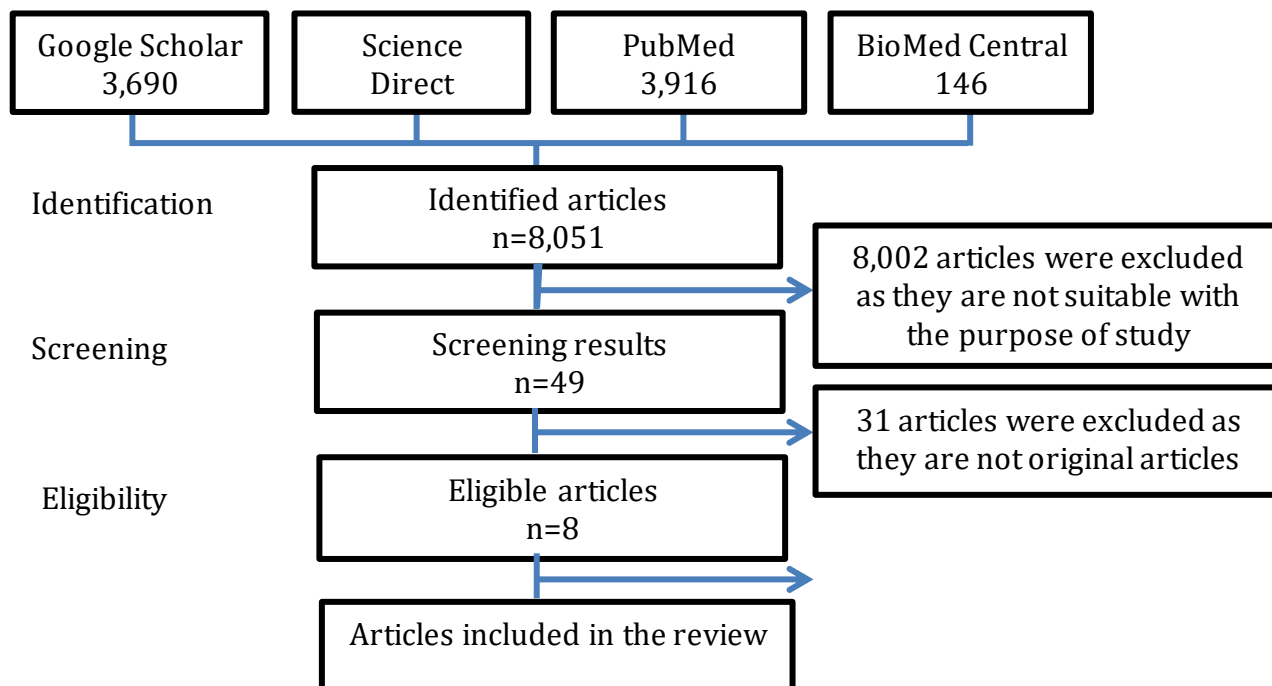


Figure 1. Search flow and article selection.

DISCUSSION

The results of this study showed a variety of strategies in the provision of education to parents. These strategies include providing detailed care instructions of adrenal crisis management in parents, encouraging parents to use positive and useful coping strategies, increasing knowledge and awareness on the use of medical alert IDs, providing Psychosocial Education Program (PEP), increasing nurses' knowledge and comfort in providing education to families, and suggestions on improving the provision of education to parents.

Provision of instructions on adrenal crisis management in parents

The incidence of adrenal crisis is a challenge for many parents, especially if health care providers do not prepare parents to manage the crisis. Unpreparedness in managing adrenal crises will cause parents to rely on medical emergency services. In a study by Fleming et al, it is reported that parents who had received an explanation on how to give an injection from health care providers

showed an increased perception of management ability; parents also expressed satisfaction when nurses or doctors prepared them to handle adrenal crisis incidents through direct explanation (Fleming et al., 2011).

In another study by Fleming, Rapp, and Sloane, it was revealed that parents who received written instructions and demonstration by doctors about drug administration procedures, both orally and by injection, had high self-efficacy scores (Fleming et al., 2011). This is also in line with a review discussing the Multidisciplinary Evidence-Based Models for Patients and Families Affected by Congenital Adrenal Hyperplasia in the chapter related to the development of educational rights for patients, families, and school personnel. Families received folders containing general information about CAH, Cortes, fludrocortisone administration, and national support groups.

In addition, the folders also provided contact information for clinics and emergency management. Oral and intramuscular doses

of glucocorticoids were given in the initial teaching session, and a summary of this information was also provided in the folders. At the first education session, patients and parents were given an emergency room (ER) card, which contains contact information for pediatric endocrinologists, service times, and instructions for administering intravenous fluids and glucocorticoids (IV). The families were also given a copy of the Hsu and Rivkees' Congenital Adrenal Hyperplasia: A Parent's Guide to take home. Parents were also provided with "CAH emergency kits," which contained a thermometer and all things needed in providing an emergent intramuscular injection of hydrocortisone hemisuccinate (Solu-Cortef) (Schaeffer et al., 2010).

Use of positive and useful coping strategies

Parents having children with CAH often feel confused in perceiving their child's sex due to abnormal appearance on the external genitalia. Also, information about disease conditions, misunderstandings and stigma about CAH, and parents' concerns about the child's future and the child's life in adulthood are some things that burden the parents and cause stress (Gilban et al., 2014). One of the ways to reduce stress is to use positive coping strategies.

The use of coping strategies aims to change how parents perceive or feel stressful situations that come from a child's condition. These coping strategies can be adaptive or maladaptive. A study by Joshi et al. reported that most parents used emotion-focused coping strategies (adaptive). Meanwhile, maladaptive coping strategies were used by a few parents, including mental disengagement, humor, rejection, substance abuse, and releasing behaviors. Health workers need to encourage parents to use positive and useful (adaptive) coping strategies, such as focusing on problems and emotions (Joshi et al., 2017). This is in line with the 2002 consensus statement, which states that "psychological assessment and

support of patients and families should be a routine component of comprehensive care and management of patients with CAH. Parents or patients should be offered psychological counseling options based on age and sex at the time of initial diagnosis." Patients and families should meet with child psychiatrists at least once a year, and the meetings can focus on discussing issues such as depression and anxiety (Schaeffer et al., 2010).

In a review paper, it is reported that as an effort to provide support and continuing education to patients and families, the Oklahoma Children's Hospital offers local group support for CAH management three times a year. The meetings focus on discussion and fun activities designed to facilitate socialization among group members (Schaeffer et al., 2010). Adult endocrinologists also attend the meetings so that patients/families can make some interactions before receiving the treatment. Support between parents and online or face-to-face education is also available on the CARES website (<http://www.caresfoundation.org/>) and the MAGIC Foundation (<http://www.magicfoundation.org/>) (Schaeffer et al., 2010).

Increasing knowledge and awareness in using medical alert IDs

Patients with CAH need to use or carry a medical identification card (ID) to inform health care providers of steroid replacement needs. Most adrenal crises occur outside home, with low levels of awareness of family and healthcare providers of the principles of emergency glucocorticoid administration. Thus, effective use of medical alert IDs can identify adrenal crises early and anticipate suitable interventions immediately (Husebye et al., 2014).

Table 1. Matrix of article synthesis

No	Title (Year)	Authors	Research Purpose	Methods and Samples	Instruments	Results
1	Parental management of the adrenal crisis in children with congenital adrenal hyperplasia (2017)	- Louise Fleming - Kathleen Knafl - George Knafl - Marcia Van Riper	- Describe the incidence of adrenal crisis in children with CAH - Explore parents' perceptions of the consequences of having children with life-threatening conditions - Determine the relationship between parental perceptual management abilities and the impact of CAH on the family.	Methods - Using the Family Management Style Framework (FMSF) to explore the family's response to children's chronic condition - Phase 1: using a mixed-method, cross-sectional study - Phase 2: semi-structured interviews Samples: - 77 parents of children with CAH (60 mothers, 2 grandmothers, and 15 fathers)	- Demographic information questionnaire - Pediatric Quality of Life-Family Impact Module (PedsQLFI M) - Family Management Measure (FaMM) - View of Condition Impact and Management Ability - Interview guides	- There was a positive and significant relationship between the provision of detailed care instructions about adrenal crisis management in parents with the perceived management abilities (p=0.02) - The stronger management ability is perceived, the less the impacts of CAR on families are (p<0.001). - Parents reported that most adrenal crises occurred when children are 0-5 years old. - Parents caring for children aged 0-5 years old perceived less ability to manage the child's condition than parents in older children (p=0.009)
2	Evaluation of a psychosocial education program for families with congenital adrenal hyperplasia (2013)	- Irene Mitchelhill - Grant Andrew Betts - Jennie King - F Murray - J Crisp - C Briggs	- Evaluate the developed Psychosocial Education Program (PEP) to meet the needs of families having children with CAH - Explore the impacts of CAH on children and families.	Methods : - Participants attended a one-day workshop which included the provision of information about CAH, injection administration training, group discussions and the provision of useable resources Samples: - 202 participants from 68 families	- Child Behavior Checklist (CBCL) and Child Health Questionnaire (CHQ) were used to explore the effects of CAH on children and families. - Knowledge Assessment Questionnaire (CAHKAQ) and formal evaluation.	- Participants' knowledge increased after the administration of PEP, which was maintained over time. - Sick day management was perceived as a major challenge for families. - Father and mother perceived that children with CAH have lower quality in the aspects of body comfort, general child health, and emotional impact on parents when compared to their siblings

No	Title (Year)	Authors	Research Purpose	Methods and Samples	Instruments	Results
3	Development of an educational resource to increase nursing staff knowledge of adrenal insufficiency Management in the Neonatal Intensive Care Unit (2020)	- Aimee L. Hastriter - Meghan Potthoff	- Assess and increase the knowledge and comfort of staff nurses in providing education to families with a diagnosis of adrenal crisis in children	Method: - Pre-post survey Sample: - 71 nurses from the NICU and 7 associate nurses at pediatric endocrinology clinics completed the questionnaires before and after the survey	- A survey of 10 questions was completed before and after the PowerPoint presentation. - Evidence-based educational resources were developed in the framework for the PowerPoint presentations, which were given to NICU staff nurses and associate nurses from endocrine clinics.	- There was a significant difference in the score of knowledge about adrenal crisis before given the intervention (M=5.18; SD=1.501) and after given the intervention (M=7.37; SD=0.775) [t (77) = -10.919; p=0.000]. - Staff nurses reported increased levels of comfort in providing education about the management of adrenal insufficiency to caregivers.
4	From Knowing Nothing to Knowing What, How and Now: Parents' Experiences of Caring for their Children With Congenital Adrenal Hyperplasia (2017)	- Tove Lundberg - Anders Lindstrom - Katrina Roen - Peter Hegarty	- Determine various knowledge used by parents in caring for children with CAH	Method: - Semi-structured qualitative interviews Sample: - 20 parents of 22 children with CAH	- Parents' interview guide about the general description of the children, parents' experience in receiving the diagnosis, the perception of parents and children about health care, communication to others about the child's condition, and parents' thoughts about their children's futures	- Parents emphasized the importance of knowledge about things supporting children's needs - The importance of knowing how to deal with and understand new situations - Paying attention to the children's medical needs and how to communicate with children. - Challenges in social relationships, emergency care, how to help children be independent, and the ability to respond to unique incidents appropriately
5	"It Was an Overwhelming Thing": Parents' Needs After Infant Diagnosis With CAH (2014)	- Kyla L. Boyse - Melissa Gardner - Donna J. Marvicsi	- Describe parents' experiences and needs after their baby is diagnosed with CAH	Method - This study employed a qualitative descriptive design with a retrospective approach	- Semi-structured telephone interviews that lasted for 45-90 minutes	- The encoding of interview transcripts revealed several themes, including the importance of communicating health problems, lack of support of care at home and

No	Title (Year)	Authors	Research Purpose	Methods and Samples	Instruments	Results
		- David E. Sandberg		- Data were gathered through interviews about how they learned about CAH and its management, followed by qualitative data analysis Sample - Six parents (four mothers and two fathers)		support in making decisions, and the willingness to obtain social support among parents - Research implications indicate the need for family-centered health care that is delivered after a newborn is diagnosed with CAH
6	Caregiver Knowledge and Self-Confidence of Stress Dosing of Hydrocortisemia in Children with Congenital Adrenal Hyperplasia (2011)	- Louise Kathleen Fleming - Carla Gene Rapp Rick Sloane	- Assess caregivers' knowledge of CAH, adrenal crises, and the administration of hydrocortisone doses during the acute period - Assess caregivers' self-efficacy in managing adrenal crises and administering stress doses to children with CAH	Method: - An exploratory study using a survey method Sample: - Sixty (60) caregivers of children aged 0-16 years old with classic salt-wasting CAH	- The questionnaire consisted of 32 questions to collect demographic data, evaluate CAH knowledge, and 10 questions to evaluate self-efficacy.	- Caregivers who received written instructions on doses and demonstration of injection techniques by trained health workers had a higher self-efficacy score in managing general adrenal crises (p=0.03) and self-efficacy of injection procedures (p=0.006)
7	Improved medical-alert ID ownership and utilization in youth with congenital adrenal hyperplasia following a parent educational intervention (2018)	- Alaina P. Vidmara - Jonathan F. Webera - Christina M. Koppin - Roshanak Monzavi - Mimi S Kim	- Evaluate the use of medical marker IDs by adolescents with CAH - Evaluate the ownership and awareness of medical marker IDs in parents - Evaluate the effects of educational interventions in	Method: - A prospective, longitudinal cohort study design that refers to the CAH Comprehensive Care Center Sample: - 50 families having children with classic CAH	- A preliminary survey was conducted to assess parents' needs in the clinics. An educational intervention was then delivered and an evaluation via telephone was conducted to assess the needs for follow-up one month after the intervention.	- In the initial survey, 20/50 (40%) families with CAH had a medical alert ID - 10/20 (50%) ID owners reported the ID use of >3 days a week - 26/50 (52%) of parents aware of the medical alert IDs - After the intervention, the number of ID ownership had doubled to 39/50 (78%; p <0.05)

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			clinics on the use of IDs.		- The quality improvement (QI) framework was used with a cycle of increasing the plan-do-study-act (PDSA) process.	- The use of ID among ID holders was 100% (39/39), and awareness increased to 42/50 (84%; p<0.05) - The obstacle reported by five Spanish-speaking families was the inability to order a medical alert ID online.
8	Knowledge, stress and adopted coping strategies of parents of children having congenital adrenal hyperplasia: An exploratory survey (2017)	- Poonam Joshi - Babita Yadav - Vandana Jain - Shilpa Sharma	- This study aimed to determine the knowledge, stress, and coping strategies used by parents having children with CAH	Method: - A descriptive cross-sectional survey with a mixed method approach Sample: - 30 parents who had children with CAH aged 0-18 years old	- The CAH Knowledge questionnaire - Parental Stress Scale - COPE inventory - In-depth interview guides	- The majority of parents (67%) had low knowledge scores (<7) and moderate stress (42-65). - Knowledge and stress had a negative relationship (r=-0.39, p=0.035). - Coping strategies used by parents to deal with stress were problem-focused and emotional-focused.

A team in European states had developed general emergency cards for patients with adrenal deficiency designed to provide health workers with quick and easy information in emergency situations (Quinkler et al., 2015). Educating patients and /or parents about adrenal crisis management with emergency glucocorticoids, followed by medical alert IDs that explain needs for care to health workers can prevent adrenal crisis complications with timely interventions.

A study conducted by Vidmar et al. found that teaching interventions to parents by providing a full page of medical alert IDs can be implemented by health workers without disrupting their workflow. The effect of this intervention can be seen from the significant improvement in the use, awareness, and utilization of medical alert IDs and in the care of the patients. The provision of education to increase knowledge and awareness in the use of medical alert IDs includes materials on detailed six types of medical alerts and other choices of medical alerts, an explanation of how to obtain a medical alert ID, and the use of emergency medical alerts on the iPhone® that is designed to be visually appealing (Vidmar et al., 2018).

Psychosocial Education Program (PEP)

The development of this innovative educational program aims to meet the needs of families having children with CAH. This program's contents include the provision of information about CAH, injection administration training, group discussions, and the resources that can be used by parents who have children with CAH. This PEP has been developed in the audiovisual DVD format to provide educational resources not only for patients and families but also for doctors and other healthcare professionals (Irene Mitchelhill et al., 2013)

Increasing nurses' knowledge and comfort in providing education to families

In a study by Hastriter & Potthoff, the Institute of Medicine states that health care improvement can be promoted by providing effective, patient-centered, and timely care. Nurses who play a role in providing education to patients and caregivers should have adequate knowledge and understanding of the diagnosis of adrenal insufficiency. Nurses need to prepare themselves in providing educational support to caregivers related to the diagnosis of complex adrenal insufficiency after discharge from the hospital. This is one of the efforts to promote effective and patient-centered care. Providing concrete education to increase nurses' knowledge of adrenal insufficiency care will facilitate timely care and management (Hastriter & Potthoff, 2020).

Hastriter and Potthoff also reported that after given a short educational session, nurses' knowledge of the adrenal insufficiency management and comfort in providing education to families also increased. During the session, the nurses received materials concerning the use of oral hydrocortisone during maintenance and administration of stress dosing, including situations where stress dosing would be recommended and whether stress dosing should be doubled or tripled from the maintenance one. The administration of parenteral hydrocortisone was reviewed, including in cases when caregivers had to give an intramuscular dose immediately. This short review covered the education that would be provided to families regarding the administration of intramuscular hydrocortisone. Complications of adrenal insufficiency were also discussed, including concerns over the inadequate management of routine drugs and the potential for an adrenal crisis.

Furthermore, the materials also contained recommendations of additional guidelines and resource information to be provided to caregivers and patients diagnosed with adrenal crisis (Hastriter & Potthoff, 2020). This is in line with the Comprehensive Care

Center (CCC) guidelines, which states that the function of the care coordinators and educators relies on the available nursing and administrative staff. In the implementation of CCC, nurses should have expertise and be responsible for educating patients and families (Auchus et al., 2010).

Suggestions for improving the provision of education

A study by Lundberg et al. reported that there were unfulfilled needs of parents during the treatment, such as encouraging parents' active involvement in decision making related to genital surgery, responding to unique circumstances and new situations promptly, explaining how to communicate with other people about the present condition, knowing when to seek emergency care for their children, and helping children to be self-reliant by developing the knowledge they have (Lundberg et al., 2017).

Parents emphasized the importance of knowing and understanding of the current situation. A majority of parents reported that they did not understand endocrinologist's explanation comprehensively.

Endocrinologists did not always consider parents' ability to receive complex and new information that is influenced by emotional states, the amount of information, and health awareness. Parents perceived that they are given too much information at once and therefore become overwhelmed. On the other side, parents also want to understand the daily care for their children and its relation to everyday life in the family (Hummelinck & Pollock, 2006).

Another study by Boyse et al. also showed that there are opportunities in improving health education provided to parents after their baby is diagnosed with CAH. The provision of family education should consider the emotional state of parents, health literacy, and learning styles, as well as the application of knowledge assessment

and use of reliable web resources, print media, and multimedia. Evidence-based multimedia web resources that are made and designed for parents are very appropriate for delivering educational materials that meet the needs of parents (Boyse et al., 2014).

CONCLUSION AND RECOMMENDATION

Conclusion

The results of this literature review show that some strategies can be used to provide education to families having children with CAH. These educational strategies include providing detailed care instructions about adrenal crisis management to parents, encouraging parents to use positive and useful coping strategies, increasing knowledge and awareness on the use of medical alert IDs, providing psychosocial education program, increasing nurses' knowledge and comfort in providing education to families, and suggestions in improving the provision of education to families/parents.

Recommendation

The provision of education to families/parents after their baby is diagnosed with CAH should consider some individual aspects, such as the emotional state of the parents, health literacy, learning styles, the amount of information provided, and the application of knowledge assessment in each family/parent.

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