Caring for Children With Congenital Upper Extremity Differences

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The purpose of this article is to provide information about the changing landscapes in research, treatment, civil rights' protection, disability awareness, and accepted terminology in the care of children with congenital upper limb differences. This knowledge can guide clinical and nonclinical conversations between patients and their families. *(J Hand Surg Am. 2021;46(12):1105–1111. Copyright © 2021 by the American Society for Surgery of the Hand. All rights reserved.)*

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CONGENITAL UPPER LIMB DIFFERENCES (CULD) were reported in approximately 1 in 1,900 births in the United States from 2010 to 2014.[1] There are thousands of people with CULD living in the United States today. Limb fetal differences are identified earlier using intrauterine advanced imaging.[2] Parents and patients have been connecting and learning from each other online in unparalleled volumes. Advocacy has driven civil rights' legislation and the development of financial agencies that protect and support children with CULD.

CLASSIFICATION AND ETIOLOGY

When surveyed, new parents of children with CULD identified information as a critical aspect of an initial interaction with health care providers.[3] Specialists may be consulted when a patient is *in utero* and an upper limb difference has been identified using 3-dimensional ultrasound or magnetic resonance imaging. Hand surgeons may play the critical role of correcting inaccurate diagnoses and referring patients for evaluations of associated syndromes.[4] Hand surgeons should have practiced thoughtful communication strategies using currently accepted terminology and language in the care of children with CULD (Table 1).

Human limb formation science has advanced with fetal imaging; however, it is still largely based on animal research. The models of limb development currently remain focused on 3 axes and unique proteins that provide either signals for early specification or positional information to mesenchymal cells in discrete zones. The limb is fully differentiated by 7 weeks of gestation and over the next 33 weeks grows only in size. Fetal limb development can be influenced by intrinsic or extrinsic factors.[4]

Congenital upper limb differences were recently reclassified into the internationally adopted Oberg-Manske-Tonkin system. It has proven useful in diagnostic, prognostic, and therapeutic functions because it incorporates both etiology and phenotype.[5,6] The Oberg-Manske-Tonkin system is flexible for diagnoses that overlap, and it leaves room for advances in genetics and human fetal limb development (Table 2).

Many syndromes have been identified using emerging global banks of genetic material.[5] Currently, if a child is nonsyndromic and has a

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TABLE 1. Preferred and Nonpreferred Terminologies

Preferred Terminology	Nonpreferred Terminology
"Congenital upper limb difference," "congenital hand difference," "congenital limb difference," "limb difference"	"Birth defect," "club," "deformity," "deficiencies," "broken," "missing," "little," "atypical," "abnormal," "anomaly"
"I am going to ask you questions about your family history and pregnancy to be complete, not necessarily because they are connected to your child's limb difference."	"Did you take any medications or drugs during your pregnancy?"; "Could this be passed down in the family?"
Person-first: descriptive without ability Eg, "Child with CULD," or "6-year-old with 1 arm/3 fingers on left hand"	Avoid assigning ability or loss Eg, "Child missing his arm"; "Disabled 6-year-old"
"There are indeed times when having an arm difference may mean things are different for you AND you will be able to do the things you need and want to do. How you do things may look a little different and that is ok."	"You are just like everyone else." "Don't treat your child any differently."
"In my opinion," "in my experience," or "what other parents and children have shared with me"	"Your child should have surgery"

TABLE 2. Diagnoses and Definitions

Diagnosis	Definition
Malformations	- Entire limb or isolated hand plate: axis specific Examples: Symbrachydactyly, duplication, longitudinal hypopolasias, syndactyly, and physis structural abnormalities (clinodactyly, and Madelung deformity)
Deformation	- Insult to the fully developed limb Example: Amniotic band syndrome - Circulating striations of the amniotic sac at ~15 weeks of gestation, form constricting scars, cause acrosyndactyly, and/or amputate limbs or partial to complete digits
Dysplasia	- Limb (bone, muscle, nerve, or vascular) growth driven by a tumor gene Examples: Macrodactyly, neurofibromatosis, or bone diseases such as arthrogryposis
Associated syndromes	- With and without known genetic anomalies or inheritance patterns Examples: TAR syndrome, SHSF, and Poland syndrome

SHSF, split hand/split foot; TAR, thrombocytopenia absent radius.

unilateral CULD, it is less likely that a genetic study will demonstrate findings. Genetic syndromes may affect the whole child and increase the fatality risk of a child with CULD. In Australia, 18% of reported children with CULD die before 6 years of age, predominantly because of associated syndromes.[4] A thorough physical examination may prompt a referral for the appropriate hematopoietic, cardiovascular, or genetic evaluations. Early identification of associated syndromes can save the lives of children with CULD. For example, children with symbrachydactyly should be evaluated for pectoralis hypoplasia or chest wall and cardiac differences, and children with radial longitudinal deficiency and thumb hypoplasia should undergo appropriate testing for vertebral, anal, cardiac, tracheoesophogeal, renal, limb (VACTERL) associations and Fanconi anemia (CBC, platelet count, mitocycin C and diepoxybutane analysis).

The most accepted etiology theory about unilateral CULD is that the limb never fully forms if the branching of arteries from the heart toward the arm, typically the subclavian artery, is smaller, clotted, or disrupted.[7] Supporting this theory is the connection between the congenital formation of the limb and heart, spine, and chest wall development, such as that in patients with Poland syndrome or Mobius and Klippel-Feil syndromes. However, it is unclear why

only 57% of children with chest wall differences have CULD if the disruption occurs proximally.[8] In addition, in these theories, the arm, hand, or fingers are apparently never formed; yet, phantom feelings and pain have been reported in 20% of children and adults with congenital absence.[9,10]

Since the thalidomide era in the 1960s, despite consistent efforts to understand links between exposures or environmental factors and limb differences, other causes have not been definitively identified. Warfarin, phenytoin, valproic acid, misoprostol, recreational drugs, and other likely teratogens may cause poorly defined associated skeletal anomalies, such as vascular disruption, delayed ossification, and muscular contractures.[11] A maternal viral infection during pregnancy may result in fetal limb differences, as recently demonstrated by fetal amyoplasia and microcephaly following prenatal exposure to Zika virus. While providing information to parents and children with limb differences, the goal is to differentiate between conditions to provide anticipatory guidance about the condition's etiology, associations, and inheritance. Symbrachydactyly, a malformation according to the Oberg-Manske-Tonkin classification, can occur at the limb or hand-plate level and can be distinguished from an amniotic band syndrome deformation based on nubbins or vestigial apical ectodermal ridge. In contrast, patients with amniotic band syndrome may present with multiple limb involvement, lymphedema, and digit syndactyly with fenestrations.[12] A third type of unilateral hypodactyly without nubbins or bands has been described, which is of an unknown origin.[13]

TREATMENT DECISION-MAKING

The roles of surgery, hand therapy, and mental health resources for the optimized care of children with CULD are being rigorously studied. Substantial challenges exist in measuring the effectiveness and outcomes of interventions.

Surgical decision-making is no longer considered straightforward for children with CULD. For example, only some studies have reported that surgery to straighten the wrists of children with radial longitudinal deficiency leads to improved limb function and quality of life.[14] For children with radial longitudinal deficiency, the wrist position correlates less with dexterity than with finger stiffness, for which treatment options are limited.[15] However, multiple other longitudinal studies comparing children and adults with operatively straightened arms and those without straightened arms have found little or no differences in activity level and participation in societal roles.[16]

Children with more than 2 opposable digits did use their hand more in functional testing, yet, were not found to have differences in overall self-care skills, psychosocial adaptation, or activity participation.[17,18] Therefore, surgery to create opposability may change how the child uses the hand, but not whether they participate or thrive.

The discrete effect of surgery in altering the cosmesis or appearance of the limb is also challenging to measure because of variations in child and parent reporting.[19–22] Findings have indicated that children with more major visible differences, such as transverse deficiency below the elbow or bilateral arm differences, undergo fewer surgeries and have overall higher body image scores and peer relationship scores than children with more subtle or unilateral differences.[23] Ylimäinen et al[24] reported in 2010 that "unexpected attention and perceived physical appearance" has a significant impact on outcomes in children with CULD.

New registries with longitudinal follow-up, patient-reported outcomes, and transparent recurrence and complication rates after surgery in children with CULD can be used for shared decision-making discussions.[14,25–28] The outcomes of surgery are also likely affected by the psychosocial health of the patient and the parents of patients undergoing these types of surgeries.[3,19,29] The time taken to address the psychosocial health and needs of the parents, who may be coping in a dysfunctional behavioral pattern, may be integral to the surgical care of these children.[30] In a case study of prenatal diagnoses of symbrachydactyly, parents who received prenatal, multidisciplinary, psychological support declined postnatal consultation with a hand surgeon completely.[31] Once the children themselves have gained self-awareness, the effect of medical decision-making on the child's psychosocial health may be more critical than surgery.[32–34] Mental health professionals, such as pediatric psychologists, should be consulted for the child and/or the parents as needed.[35]

PROSTHETICS

Upper extremity prosthetic devices have been reported as an advantage socially rather than functionally in older children and adults with CULD.[36] Prosthetics cover up the sensation of the terminal end of the residual limb, and patients have reported a

need to perform tasks without them as well. "Sit to fit" or the early introduction of prosthetics was not associated with later child satisfaction or functional use of the prosthesis.[37] Modern, 3-dimensional, printed and multiarticulating myoelectric prosthetics for children with CULD have a wide range in terms of costs, efficacy, need for replacement parts, training therapy sessions, and insurance coverage.[38] The funding source and the cost of the prosthesis directly result in usage differences.[39] Without clear evidence to guide the treatment of CULD with prosthetics, hand surgeons are likely needed to revisit the topic over time as the patient continues to explore the role of prosthetics in their health and functioning.

QUALITY-OF-LIFE, EVIDENCE-BASED STUDIES

Several, but not all, quality-of-life studies have demonstrated similar levels of self-confidence and self-esteem in children with CULD compared with their peers.[9,17,19,20,24,32,34,40,41] Children with complete congenital limb absence below the elbow have reported higher scores on adjustment and peer relationship scales than those with differences isolated to digit formation.[23,42] Empowerment and participation may be the most relevant outcomes to study. Results have suggested that surgeries to add digits, straighten the arm, or wear prosthesis are not significant factors for a child's scoring of the quality of life, but a child's own personal coping mechanisms have far more impact.[42,43] Single-nation registry studies in this field, using all patients from a single country, may help elucidate an integral cultural component.[9,21,24,44] Studies analyzing existing scoring systems (eg, Patient-Reported Outcomes Measurement Information System [PROMIS], Perceived Quality of Life [PQoL] scale, Health-Related Quality of Life [HRQoL] scale, Prosthetic Upper Extremity Functional Index (PUFI), 36-Item Short Form Survey [SF-36]) suggest that there is not yet a normalized, validated instrument that can be used to successfully measure a complete, unbiased evaluation of psychosocial health and outcomes for children with CULD.[45]

Although most children with hand differences have positive levels of self-concept, there are many who struggle with psychosocial adjustment or pain and may benefit from being routinely assessed.[45] A person's perception of their difference may shift across their life span as they encounter various milestones, and the need for support will likely change with age, environment, identity development, and personal experiences. Semistructured interviews with trained mental health professionals may be the most sensitive method of gathering tailored information to connect patients with appropriate resources.[34]

COMMUNITY

Having a child with a limb difference is uncommon enough that the parents may find themselves feeling isolated. Parents have reported increased feelings of support and empowerment through child-to-child, parent-to-parent, as well as child-to-mentoring adult connections.[29] Communities can generally fall into a few broad categories: patient parents and close family members; resources and advocacy; and children or adults themselves. Depending on internet access, language barriers, and proximity to in-person services, patients may have a variety of options: medical and patient communities, group therapy, social media blogs, personal accounts and groups, online forums, resource and advocacy organizations, camps, or in-person events or gatherings. Many hand surgeons participate in parent-based or hospital-based virtual and physical limb difference communities.

CURRENT THEORIES OF DISABILITY IDENTITY AND TERMINOLOGY

Children with CULD and their families are learning to navigate social implications and messages related to disability and of being visibly different. The following models are an introduction to the current relevant research on disabilities in children: the person, environment, and occupation model; Goffman's theory of stigma; and medical, social, and intersectionality models (Table 3).[23,34,35,40,46] Parents and adults living with children with CULD have reported that they prefer an authentic representation of the child's disability rather than an "is just like everyone else" narrative. When trying to express positivity and support, physicians may downplay the negative aspects of a limb difference and fail to provide adequate education regarding the hardships, which research has demonstrated, that exist for the child and their family.[29]

The terminology most currently used in the literature and in the care of children with CULD is focused on being positive and descriptive without attributing stigma. Surveyed parents of children with CULD have reported that "attitude" and "emotional and psychological support" are critical components of health care interactions.[3] Parents and children

TABLE 3. Theories of Disabilities

Theory/Theoretical Model of Disability	Definition
Person, environment, and occupation model	Child exists in multiple contexts beyond anatomy. Interventions must consider the child's personal factors and environmental (physical, cultural, social, and temporal) context
Stigma theory	Stigma is the negative association of socially unfavored traits. "In" group perceives disabled people as "out" group, practicing ableism through discrimination and microaggressions. Risk: Internalized ableism—child embraces assigned limitations, isolates Concealment, avoidance, try to "pass as normal"; anxiety of being discovered, lower self-worth Goal: Acceptance or pride, educate or mentor—significant emotional and mental energy used; more common in older children and adults, and not every person will attain this
Medical	Deficiency in the biomechanics of the human body should be corrected using interventions whenever possible to assimilate to the expected norms
Social	Social and environmental constructs determine disability, not physical functionality
Intersectionality	The unique experience felt by people who have multiple minority identities. Children with CULD navigate microaggression associated with the disability simultaneously with other minority-group affiliations (racial, cultural, socioeconomic, mental health, behavioral or learning issues, gender identity, sexual preference, religious, etc.)

specifically have reported comfort in the language of "differences."[47] The term is closer to the concept that CULD is a part of human variation (Table 1).

Word order may also matter to families and children with CULD. Some prefer "person-first language," indicating that people are people first

TABLE 4. Federal Legislation on Disabilities

Federal Legislation	Definition of Disability	Summary
Americans with Disabilities Act of 1990	"A physical or mental impairment that substantially limits 1 or more major life activity" "Applies to someone who is regarded as having a disability"	Protects individuals from discrimination in employment, transportation, public programs access, buildings, and communication • https://www.ada.gov/pubs/adastatute08.htm
Section 504 of the Rehabilitation Act of 1973	"A physical or mental impairment that constitutes or results in a substantial impediment to employment" Documented disability required; students assessed to determine supports needed	- Provides resources to access learning environment - School teams annually review or modify - No reimbursement for school costs - States or school districts determine how policy is met, leading to wide variation • https://www2.ed.gov/policy/speced/leg/rehab/rehabilitation-act-of-1973-amended-by-wioa.pdf
Individualized Education Plan statue of Individuals with Disabilities Education Act (1975)	"A severe orthopedic impairment that adversely affects a child's educational performance." "Includes impairments caused by a congenital anomaly, impairments from disease, or impairments from other causes." Documented disability required; students assessed to determine supports needed	- Provides accommodation to access physical environment or curriculum - School teams annually review or modify - Funds available for reimbursement of school costs - States or school districts determine how policy is met, leading to wide variation • https://uscode.house.gov/view.xhtml?path=/prelim@title20/chapter33&edition=prelim
Social Security	"Musculoskeletal system -inability to ambulate effectively or perform fine and gross movements effectively must have lasted, or be expected to last, for at least 12 months." "Impairments substantially limit the ability to do basic work and or be employed."	- Financial support by Social Security Insurance—covered health benefits and/or supplemental security income. - Need to apply, document disability, and be approved to receive funding and services • https://www.ssa.gov/ssi/text-child-ussi.htm

and that the disability is secondary (eg, "an athlete who has a limb difference"), whereas others prefer "ability-first" terminology, wherein their disability is an adjective that helps describe who they are (eg, limb-different athlete). Because both are presently acceptable, hand surgeons can take cues for personalized terminology from individuals and empower families to develop the ever-evolving vocabulary they prefer.

LEGAL DEFINITIONS AND ADVOCACY

Parents and older children may report the instances of apparent discrimination to the surgeon or ask them about federal resources that fund care and help the child fully participate in school. It is useful to have a general knowledge of the current legal definitions of disability that apply to upper limb differences and of active federal and state legislation that protect civil liberties and provide resources at school for patients. Table 4 reviews the current federal legislation for publicly funded organizations that may apply to children with CULD. Legally qualifying the child's abilities or others' perceptions of their ability is not the responsibility of the hand surgeon; however, the diagnoses, details, and medical records are used by parents to apply for these services, and hand surgeons should be prepared to provide this documentation as needed.

SUMMARY

Providing information about and appreciating the changing landscape for children with CULD is paramount for successful outcomes of care. An early, accurate diagnosis helps guide children and families as they navigate their identity with the healthy acceptance of the limb difference, receive appropriate referrals for associated syndromes, seek supportive communities, and consider future progeny. It is important that hand surgeons encourage each child and family to individually explore the available treatment options and to make them feel empowered to participate in their medical care.

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