

Public health and disability – The importance of keeping up the good work

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Abstract: Secondary and tertiary prevention strategies targeting people with disabilities to improve their health and wellbeing is important. In Sweden, >95% of all children with cerebral palsy (CP) participate in a follow-up program, where one of the goals is the prevention of hip dislocations. We reviewed the incidence of hip dislocations from 2010 to 2019 and the number of children who underwent different types of hip surgeries. The number of hip dislocations was reduced from 8.8% before introduction of the program to 0.3-0.4% up to 2015, followed by a gradual increase to 0.8% in 2019. The proportion of children who underwent adductor-psoas lengthening as their primary preventive surgery decreased from 50% in 2017 to 37% in 2019 with a corresponding increase of children undergoing femoral osteotomy, indicating more children underwent surgery at a later stage. Reasons for the increased number of children with hip dislocation may be that more children have moved to Sweden in recent years without corresponding compensation in health care resources and increased waiting times for surgery due to, among other things, a shortage of nurses. The results highlight the importance of constantly monitoring follow-up programs.

Keywords: Disability, quality of life, cerebral palsy, surveillance.

1. Introduction

All children should have the right to health and quality of life (QoL) [1]. However, disability, irrespective of which of the numerous definitions available is used, affects millions of people of all ages. These individuals are at increased risk of reduced participation, limited involvement in everyday activities, and reduced QoL [2,3]. This may be due to any combination of body function and structure, personal factors, or notably, the environment, as identified in the World Health Organization's (WHO) *International Classification of Functioning, Disability and Health* [4]. Worldwide, individuals with disabilities are among the most marginalized in society, with less access to education, work, and financial assets [5].

Like disability, public health is defined in different ways. The definition used by the *American Public Health Association* states that “public health promotes and protects the health of people and the communities where they live, learn, work and play” [6]. In contrast to regular healthcare, which targets unique individuals and tends to be reactive in nature, public health is concerned with population-based health and the thrust is on prevention. In brief, public health uses surveillance to investigate the scale of a problem; epidemiology to identify specific groups, develop measures, and apply rigorous research methods; and intervention to improve health outcomes [7]. Traditionally, public health has been charged with preventing morbidity, mortality, and disability [8]. Given the broad heterogeneity of disability, combined with the goal of preventing it, there has been confusion as to where people with disabilities fit in the context of public health [7,9]. This could partially be explained by the inaccuracy of equating disability with poor health [7]. Nevertheless, primary, secondary, and tertiary prevention are all cornerstones in public health and the need for and the importance of secondary and tertiary prevention strategies targeting people living with disabilities should not be understated.

Cerebral Palsy and the Cerebral Palsy Follow-Up Program (CPUP)

One of the most common lifelong disabilities is cerebral palsy (CP). CP is caused by non-progressive brain damage that occurs before the age of two years [10], oftentimes already in utero. There is great variability in functioning, however, motor function is always affected and challenges in cognition, perception, sensation, behavior, and comorbidities such as epilepsy are frequent [10,11]. Hip displacement, which means that the femoral head is being laterally displaced within the joint, is common in individuals with CP, due to altered muscle forces across the hip joint. Hip dislocation, which is defined as the femoral head being out of the joint, is a severe problem, with high risk of pain, development of severe contractures, windswept deformity, and scoliosis (Figure 1), resulting in problems with positioning, sitting, standing, and walking [12,13]. The risk of hip dislocation is estimated to 15–20% in the total population of individuals with CP and the risk is highest in those with severe limitations of gross motor function [14]. Hip dislocation in CP is often preventable. Evidence show that at a population-based level, hip displacement can be prevented if young children with CP are included in a surveillance and follow-up program that includes repeated radiographic and clinical examinations and preventive treatments for hips that are displacing [15,16]. Non-surgical management to prevent hip dislocation consists of positioning the hip in abduction and extension in lying, sitting, and standing. Surgical treatments of hip displacement in CP, on the other hand, consists of adductor-iliopsoas lengthening (APL) or varus- derotation osteotomy of the proximal femur (FO), sometimes in combination with pelvic osteotomy. APL is performed primarily on younger children with a lesser degree of displacement, while osteotomy is performed on children with more severe displacement.



Figure 1. Pelvic radiograph of 8-year-old boy with cerebral palsy and left-sided hip dislocation.

The cerebral palsy follow-up program (CPUP) started in 1994 in the Region of Skåne in southern Sweden to address the problem of painful hip dislocations in children with CP [17]. Children with CP would show up to clinic with painful difficult to treat hip dislocations requiring extensive surgery. In some cases, it was too late to perform surgery at all. The multidisciplinary team at the time consisting of a pediatric orthopedic surgeon (author GH), a physical therapist, a pediatric neurologist, in collaboration with the professionals at the child re/habilitation units decided to adopt a multidisciplinary preventative approach to prevent hip dislocations. The decision was made to implement a population-based follow-up program where all children with a suspected diagnosis of CP would be followed over time irrespective of if they had any indications of hip dislocation. Standardized assessments were developed and the data collected were compiled into a secure database. To-date, 95% of all children and adolescents with CP in Sweden participate in CPUP as well as many adults. Given that CPUP started in southern Sweden in the 1990s, we have access to a pre-CPUP cohort of individuals; with those born 1990-1991 constituting a historical pre-surveillance cohort and individuals born 1992 or later who have been followed in the surveillance program. Results from southern Sweden were published after 10- and 20 years respectively, showing that hip dislocations in children with CP had been reduced from 8.8% to 0.4-0.3% [15,16].

1.1 Purpose and aims

The purpose of this report was to

- (1) review the incidence of hip dislocations in children with CP residing in Sweden from 2010 to 2019,
- (2) review the number of children who underwent APL and FO during 2017-2019,

- (3) outline plausible hypotheses of the development of hip dislocations over time, and
- (4) discuss the significance of well-functioning infrastructure and adherence over time in public health.

2. Materials and Methods

This was a retrospective registry study based on data from the CPUP. In CPUP, the internationally used gross motor function classification system (GMFCS) is used to determine GMFCS levels [18,19]. All children in GMFCS levels III-V are radiographically examined with a pelvic anteroposterior view annually up to 8 years of age and those in GMFCS level II at 2 and 6 years of age. Children in level I are not examined radiographically provided the child has a normal pain free range of hip motion. From 8 years of age, the examination intervals are determined individually based on the results of previous examinations and clinical reports. The degree of displacement of the femoral head is measured with the Reimer's Migration Percentage (MP), where MP 100% defines hip dislocation (Figure 2). Children 16 years or younger with a diagnosis of CP in GMFCS levels I-V reported to CPUP were included. Children entering the program with hip dislocation (MP 100%) on the first radiographic examination were excluded from the analyses.

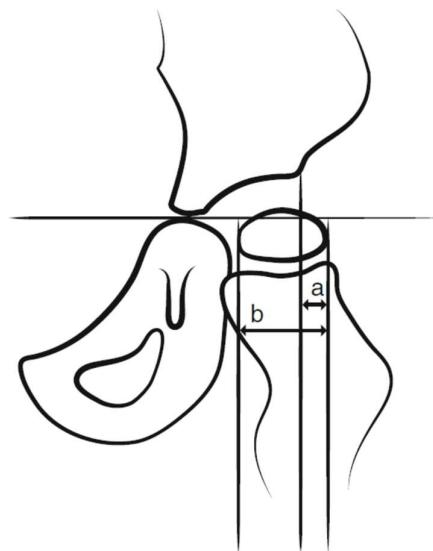


Figure 2. Measurement of hip Migration percentage (MP). $MP = a/b \times 100$

2.1 Statistics

The number of individuals with a recorded hip dislocation as of December 31, for each individual year 2010-2019, was registered and related to the total number of children in each respective GMFCS level the during the same time span. Historical comparisons were made with the results from the control group of children born 1990-1991 (before CPUP) and with the results presented after the 10 and 20-year follow-ups of CPUP. Data were presented descriptively as absolute numbers and percentages. The proportion of children who underwent surgery to prevent hip dislocation with APL and FO was registered for the years 2017 - 2019.

This study falls under the Ethics Board approval LU-433-99, dnr 2020 04511

3. Results

The total number of children with CP in CPUP increased from 2,186 in 2010 to 3,791 in 2019. There distributions according to sex and GMFCS levels during the years of interest were similar. The mean age increased from 7.25 years 2010 to 9.60 years in 2019 (Table 1).

Table 1. Sex, Gross Motor Function Classification System level, and mean age of participants, 2010-2019.

		Report year									
		2010	2011	2012	2013	2014	2015	2016	2017	2018	2019
Boys		1,253	1,388	1,530	1,654	1,797	1,928	2,021	2,078	2,155	2,206
		(57.3)	(57.1)	(57.1)	(56.9)	(56.9)	(57.2)	(57.2)	(57.3)	(57.7)	(58.2)
Girls		919	1,028	1,133	1,233	1,336	1,429	1,486	1,514	1,549	1,569
		(42.0)	(42.3)	(42.3)	(42.4)	(42.3)	(42.4)	(42.0)	(41.8)	(41.4)	(41.4)
Unknown		14	15	17	22	27	26	28	32	34	16
		(0.6)	(0.6)	(0.6)	(0.8)	(0.9)	(0.8)	(0.8)	(0.9)	(0.9)	(0.4)
Total		2,186	2,431	2,680	2,909	3,160	3,373	3,535	3,624	3,738	3,791
GMFCS	I	930	1,042	1,154	1,236	1,364	1,488	1,549	1,599	1,678	1,703
		(42.5)	(42.9)	(43.1)	(42.5)	(43.2)	(44.1)	(43.8)	(44.1)	(44.9)	(44.9)
	II	322	354	403	454	503	540	574	570	563	598
		(14.7)	(14.6)	(15.0)	(15.6)	(15.9)	(16.0)	(16.2)	(15.7)	(15.1)	(15.8)
	III	185	220	236	263	275	286	320	330	335	327
		(8.5)	(9.0)	(8.8)	(9.0)	(8.7)	(8.5)	(9.1)	(9.1)	(9.0)	(8.6)
	IV	296	342	386	424	450	482	524	517	529	527
		(13.5)	(14.1)	(14.4)	(14.6)	(14.2)	(14.3)	(14.8)	(14.3)	(14.2)	(13.9)
	V	332	383	421	458	507	548	568	608	633	636
		(15.2)	(15.8)	(15.7)	(15.7)	(16.0)	(16.2)	(16.1)	(16.8)	(16.9)	(16.8)
Unclassified		121	90	80	74	61	39	0 (0)	0 (0)	0 (0)	0 (0)
		(5.5)	(3.7)	(3.0)	(2.5)	(1.9)	(1.2)				
Mean age		7.25	7.61	7.93	8.27	8.67	8.96	9.27	9.43	9.52	9.60

The total number of children with hip dislocation ranged from 12 to 14 during the years 2010-2014 (0.4-0.5%). Starting in 2015, the number gradually increased over time to reach 29 in 2019 (0.8%). Of the 29 children with hip dislocations in 2019, four dislocated while waiting for spine surgery for scoliosis, which were to be performed prior to the hip surgery. An additional five children dislocated their hips while wait-listed for hip surgery. For some of these children, reductions of the displaced hips are planned and in one child, a salvage procedure will have to be undertaken.

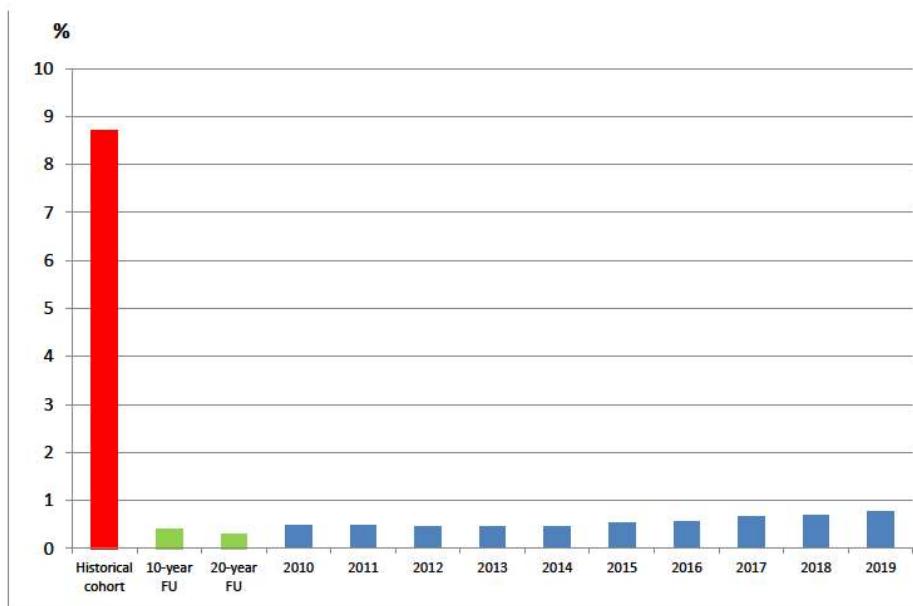


Figure 3. Proportion of children with hip dislocation during the years 2010-2019. For comparison historic cohort before CPUP (birth years 1990-1991) and 10- and 20-year follow-up.

The proportion of children who underwent APL as their primary surgery decreased from 50% in 2017 to 37% in 2019 with a corresponding increase of children operated with VDRO (Figure 4).

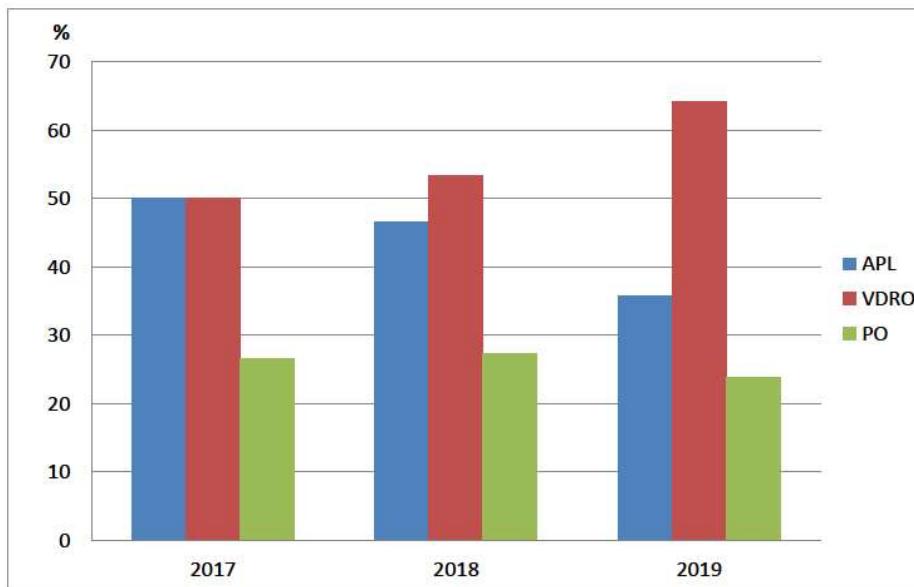


Figure 4. Proportion of children who underwent surgery with adductor-iliopsoas lengthening (APL), varus derotation osteotomy (VDRO) and pelvic osteotomy (PO) during the years 2017-2019.

4. Discussion

Hip dislocation is a secondary condition that primarily affects individuals with CP in GMFCS levels IV-V. Dislocations tend to be painful and are often indirectly negatively associated with QoL. Our earlier findings have shown that hip dislocations can be reduced substantially through surveillance and systematic preventive follow-ups [15,16]. The current report shows that an increase of hip dislocations in children with CP in Sweden has occurred in the last five years. Internationally, 0.8% of hip dislocations in children with CP is still a low percentage. However, that means that a number of young children might have experienced avoidable pain, have reduced their function — possibly permanently —, likely have had to undergo more invasive surgeries, and had longer rehabilitation periods than might otherwise have been needed. We know we can do better, because we have done so in the past.

The pillar stones of public health are important in this study in that surveillance, epidemiology, and interventions all play a part. To ensure the prevention of hip dislocations over time require all three. By monitoring hip dislocations in CPUP, it is clear that the incidence of hip dislocations is headed in the wrong direction. To implement an intervention to stabilize the incidence of hip dislocations in this population in Sweden, it is necessary to determine the reasons as to why it is happening in the first place. One important question to address is whether the observed increase is real. At times, changes in prevalence or incidence are artificial and due to, for instance, a change in a definition or criteria. The definition of CP was expanded in 2007 [10], clearly before the earliest year included in the current study. However, it did not change the prevalence of individuals with CP enrolling in CPUP. Furthermore, the definition of hip dislocations used in CPUP has remained constant, as have the assessment intervals, making it unlikely that hip dislocations are more likely to be found now than previously.

The total number of children in the surveillance program increased during the study period. Most regions included children born in 2000 and later, which means that the proportion of children aged 10-16 years gradually increased during the years 2010-2016. Most hip dislocations occur before the age of 10 years [14], meaning that most likely this did not affect the reported proportion of children with hip dislocation during these years. It is also possible that the increased number of children during the years 2015-2019 was due to an influx of children with CP who immigrated to Sweden. These children were more likely to be in GMFCS levels IV-V and thus have a higher risk of hip dislocation. Nevertheless, the distribution of GMFCS levels in the total population was stable during these years. However, the analyses do not include the children who had a complete dislocation (MP 100%) on arrival to Sweden. Children in the study cohort who have immigrated to Sweden also had the possibility to be followed and treated to prevent hip dislocation just like the children born in Sweden. Although this may sound equitable and fair, there are obstacles that might prevent that from actually happening. Arriving to a new country can be tumultuous and it is not easy to learn a new country's healthcare system, in particular not if there are language barriers. Hence, it is possible that it takes a longer time for individuals with CP who have immigrated to Sweden more recently to become part of CPUP than children born in Sweden. Consequently, the MPs might increase without anyone paying attention, resulting in hip dislocations. Furthermore, increased resources for medical care have not compensated the increased need now required with more children requiring services.

The steps that go into the chain of preventing hip dislocations need to be in place and the importance of timing is vital. The families have to be systematically scheduled and show-up to appointments for this to work. Physical therapists need to measure, record (surveillance), and intervene if needed. When problems are noted, swift referrals have to be made to an orthopedist who can decide on what actions need to be taken. It is possible that interventions or surgeries are delayed due to waiting lists and delayed surgeries allowing emerging hip displacements to become a hip dislocation. This also needs to be seriously considered in light of the foreseeable delay in many health care services that can be expected due to COVID 19. There is a shortage of nurses in Sweden, which has resulted in increased waiting times for surgery at several hospitals. Of the 29 children with hip dislocations in 2019, nine dislocated while on the waiting list for spine or hip surgery. Some of the children with hip dislocations were considered too weak to undergo surgery, and in some cases, the parents chose to refrain from surgery. However, that has always been the case, and the proportion of children who do not undergo preventive surgery has not increased over the years. In recent years, the proportion of children who have surgery with APL has decreased and the proportion that has had surgery with FO has increased, indicating that the children are undergoing surgeries at later ages. Terjesen analyzed a hip surveillance program in parts of Norway. Hip dislocation occurred in 4% of the children in the program. The main reasons were late inclusion in the program and long waiting list for preventive surgery [20], which support the hypotheses of why we are observing an increase in hip dislocations in Sweden.

There is also the importance of collective memory among professionals. There are plenty of examples of this. For instance, healthcare professionals who were working when polio was rampant and could observe the consequences first-hand likely had an easier time to understand the importance of population-based vaccination programs. Healthcare staff,

including clinic supervisors, in Sweden who regularly worked with children who were in obvious pain due to hip dislocations and who could tell, over time, how fewer children experienced this might be more likely to understand the importance of the multidisciplinary infrastructure to be in place and how fast intervention is crucial. Newer staff who are unaware of what happens when hip dislocations are not prevented might have a more difficult time to grasp this. The successful prevention of hip dislocations requires knowledge and action on many levels, including those who are in more administrative positions and make policy or decisions that affect health care.

Several measures have already been taken to halt the increasing proportion of children with CP with hip dislocation. A survey sent to orthopedic surgeons in Sweden confirm that waiting times for back surgery vary between different clinics in the country. This has been brought to the attention of the pediatric orthopedic surgeons and other health professionals in Sweden so that various measures can be taken locally to prioritize these children for surgery, or refer to hospitals that have shorter waiting lists for surgery. To prioritize children with CP may be particularly important during the COVID 19 pandemic as there is a risk that waiting times will increase even further and in more locations. Information is also planned for families with children with CP.

People with disabilities need to have well-functioning systems in place to monitor their health and wellbeing. In this report, we have used the real life example of hip dislocations of children with CP in Sweden. Nevertheless, it could have been about any of a number of topics. In the case, of CPUP, we have shown that through the hard work of the professionals involved in the health care of children with CP it is possible to reduce the number of hip dislocations. However, the importance of monitoring changes over time — in a sense surveying the surveillance — determining facilitators and barriers and acting upon them, shows the importance of keeping up the good work.

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Abbreviations

The following abbreviations are used in this manuscript:

CP	Cerebral palsy
QoL	Quality of life
WHO	World Health Organization
CPUP	Cerebral Palsy Follow-Up Program
APL	Adductor-iliopsoas lengthening
FO	Varus- derotation osteotomy of the proximal femur
GMFCS	Gross Motor Function Classification System
MP	Migration percentage
VDRO	Varus derotation osteotomy

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