Chapter 1

Introduction

"Cerebral palsy (CP) described a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems."[1] CP is the most common physical impairment in childhood.[2-5] In developed countries over the last three decades ago, the probability of survival has increased even in children with severe disabilities. Due to the widely use of electronic fetal heart rate monitoring there are 5-fold increase in the cesarean delivery rate over the same period of time. In contrast, the prevalence of CP has not decreased but remained constant for about 2 to 3 per 1,000 live births for several decades.[6, 7] However, recent data shows that the overall prevalence of CP declined both in the European[8] and Australian[9] Registers. Additionally, CP in term infants differs from that in very premature babies in prevalence and trends over time. Prevalence rate of CP increases 40 to 100 per 1,000 live births among infants born prematurely or low birth weight, and also tends to rise in recent years for this group.[4, 10] Many children with CP have accompanying impairments such as seizure disorders, visual impairments, hearing impairments, communication problems, cognitive and attention deficit. Most children with CP need help specifically for their education. As adults, half of them are faced with the difficulty of the job and living independently.[2, 11, 12] Because of the high incidence and multiplicity of symptoms, so CP is a medical and social importance contribute to the economic burden.

Worldwide attention to the issue of establishing a network of CP surveys and registers to monitor trends in CP rate. Such registers exist in several places around the world: The Surveillance of Cerebral Palsy in Europe (SCPE) is a collaborative network of CP registers and survey in 14 centres in eight countries across Europe. Some European centres have been established for a long time, e.g. Ireland, Denmark, and Sweden, as well as in Norway, and the United Kingdom. In other places, such as France and Italy, the registers started more recently.[13] Population based CP registries can be easy for many countries with universal health care system, and there are not many people.[14] Despite of the survey research can find prevalence rate of CP but it is difficult. It is difficult to do the survey research on prevalence rate of CP because it depends on the definition of prevalence. The study of prevalence should make it clear how numerator and denominator were determined.[15] Prevalence of children with CP surveys may be

lower than the actual prevalence.[11, 16] A retrospective cohort study[17] of all Asians infants who were born in California were categorized as East Asian (Chinese, Japanese, Koreans), Filipino, Indian, Pacific Islander (Guamanians, Hawaiians, and Pacific Islanders), Samoan, or Southeast Asian (Cambodian, Laotian, Thai, Vietnamese) found that prevalence of CP was lower in Asians than whites (Asians=1.09, whites=1.36 per 1,000), but still cannot explain why. Although, this study showed that the prevalence rate is lowest 0.61 per 1,000 in Thai children, there has never been a true study on the prevalence of CP in Thailand because there is no a database or the Cerebral Palsy Registry yet. Besides, the survey report of the disability of the National Statistical Office in 2007 found that there were 29,841 people with diagnosis of CP who were found the most in the northeast (12,019), followed by the northern (8,944) part of Thailand.[18]

Prior to 1980, the medical information that is the most common cause of CP is hypoxic-ischemic encephalopathy at birth,[19] but at present, the most common cause of CP is premature labor and problems in the prenatal period.[20] There is not the best way to treat CP, as well as age to start treatment, and duration of treatment.[21] However, one of the treatment goal is to ensure that patients can ambulate and take care of themselves independently.[5] For decades, the direct goal of medical care for motor disabilities was to reduce the motor related-impairments such as spasticity and muscle contracture in order to enhance functional capacity.[22] However, among several conceptual models of disability, the World Health Organization (WHO)'s International Classification of Functioning, Disability, and Health (ICF),[23] have shifted the primary focus of treatment to the level of activity and participation of the individual patient. Most parents of children with CP always want to know its severity and whether their children will ever walk. The prognosis of ambulatory outcome in children with CP has difficulty because several factors can influence ambulatory status during child's growth. However, an identification of predictors for ambulation is the most important in order to assist appropriate plan of intervention.[24, 25] Especially when the prognosis capacity on the walking tends to poor, appropriate treatment planning is the most effective way to prevent the loss of ambulatory capacity.[26]

The factors to predict ambulation for children with CP have been informed for decades by Sala and Grant.[27] They are divided into three main groups: (1) primitive reflexes and postural reactions, (2) gross motor skills, and (3) type of CP. In addition to these factors, other factors (e.g. epilepsy, intellectual disability, visual impairment, and hearing impairment) have been

considered in several studies.[12, 24, 25, 28-31] Bleck[32] has also established a scoring system to predict ambulatory children with CP aged one year or more, which is the problem of delay primary walk. This scoring system has seven reflexes as predictors. However, a recent study in Japan is about Bleck's scoring system to examine the difference of this score between the walking group and the non-walking group in children with spastic quadriplegia CP. This study showed that there was no significant difference in Bleck's scores between two this groups.[33] It shows that the study does not support the Bleck' study possibility that he using predictor of a clinical predictor reflexes only, which also has other clinical predictor affecting walking prognosis. However, there is no consensus that these factors may have contributed to the success of the walk independently. Only the conclusion of Sala and Grant are generally considered to be the most prognostic predictors significant of the achievement of ambulation.

Some previous studies about predictors of ambulation in CP had a relatively small number of patients recruited from single clinic,[24, 30, 31] study only subgroup of CP,[24, 31] using only a univariable analysis[12, 31] and the results were sometimes conflicting. In addition, the definition of "ambulation" varies across studies, which makes comparison of results difficult and important in the operational definitions did not provide enough information to determine whether the "ambulation" can be used to achieve the function. Therefore, using the operational definitions of the accurate of gross motor skills and ambulation in future studies is necessary. In year 1997, Palisano and colleagues[34] developed a tool for children with CP, five-level of gross motor function classification system (GMFCS), and edit it by including age band for youth 12 to 18 years old and adding the concepts of ICF in 2007.[35] The only study of Simard-Tremblay et al. (2010)[31] and Kułak et al. (2011)[36] in the past year, which GMFCS was used as a tool to classify ambulation. Many experts in clinical practice has developed its own unique measurement for predict ambulatory status in children with CP. However, these measures may offer appropriate prognostic accuracy but not transferable to other contexts.[24] Although prognostic tool for gross motor function among children with CP have been developed,[25, 32, 37] but a simple tool to predict ambulatory status for CP children and GMFCS was used to measure ambulatory status is still lacking. And the amount of research for predicting ambulatory children with CP has increased. However, there's no quantitative synthesis of the evidence, only literature review by Montgometry.[28]

Scope of the thesis

This thesis would like to know what prognostic predictors in the model of the ambulatory children with CP by using a systematic review and meta-analysis. And the under the context of Thailand which is not the CP register to note that there are prognostic predictors of walking ability among children with CP. Finally, these prognostic predictors used to create a simple tool to predict the ambulatory children with CP in Thailand.

1. Prognostic predictors for ambulation in children with cerebral palsy

The GMFCS is the functional assessment that has been widely accepted.[34] However, only two recent studies used this to classify ambulatory status.[31, 36] To determine prognostic predictors for ambulation will help in planning appropriate therapeutic and rehabilitation goals. And the prognostic predictors from routine data can used to develop prognostic score chart for predicting the future ability to walk among Thai children with CP.

The studies entitled “prognostic predictors for ambulation in Thai children with cerebral palsy aged 2 to 18 years” and “prognostic predictors for ambulation in children with cerebral palsy: a systematic review and meta-analysis of observational studies” were conducted to determine prognostic predictors for ambulation in Thai children with CP and confirm prognostic predictors for ambulation in worldwide children with CP by routine data.

2. Clinical prediction rule for ambulation in children with cerebral palsy

Most parents want to know that their children with CP will walk or not. A simple tool to predict ambulatory status and one which uses the GMFCS is still lacking. A simple prognostic scoring scheme will help planning proper treatments and rehabilitation goals, and to notify the caregivers.

The study entitled “derivation of an ambulatory prognostic score chart for Thai children with cerebral palsy aged 2-18” was conducted to develop a simple prognostic score chart for predicting ambulatory status in Thai children with CP using routine data.

In conclusion for the scope of this thesis, the first part of this dissertation descripts the epidemiology of CP presented in Chapter 2. The second part deals with diagnosis, treatment, and prevention of CP presented in Chapter 3. The prognostic predictors for ambulation in children with CP, associated with studies I and II, are presented in Chapter 4. Finally, the Clinical prediction rule for ambulation in children with CP associated with studies III presented in Chapter 5. Philosophical context of clinical epidemiology design in this thesis, including theoretical, data collection, and data analysis design are presented in Appendix A.

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