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Case Report

Understanding The Risk Factor of CTEV and Its Management at Primary Health Care

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ABSTRACT

Congenital Talipes Equinovarus (CTEV) is a congenital deformity affecting the lower extremities. CTEV is classified into four components: Cavus, Adductus, Varus, and Equinus. Risk factors for CTEV are most commonly associated with twin pregnancies, with a higher incidence reported in monozygotic twins compared to dizygotic twins (32% vs. 2.9%). Other contributing factors include maternal smoking during pregnancy, which has been reported to increase the risk of CTEV by approximately 1.3–2 times. In addition, exposure to teratogenic drugs during pregnancy may disrupt fetal musculoskeletal and neuromuscular development, increasing the risk of CTEV by 1.5–3 times, particularly during the first trimester, thereby increasing the likelihood of congenital deformities of the lower extremities. The Ponsetti method has a high success rate, is minimally invasive, and is effective in improving foot function and deformity when performed early in suspected CTEV patients. A 12-month-old twin infant presented to the outpatient clinic with deformities in both lower extremities. History taking revealed oligohydramnios, tobacco exposure, and medication consumption during pregnancy. Management was performed using the Ponseti method, including serial casting correction and bracing with Dennis Brown splint shoes which implemented in primary health care. Early recognition of risk factors may reduce the risk and severity of CTEV.

Keywords: Case Report, CTEV, Management, Risk Factors

INTRODUCTION

Malformation of the foot known as CTEV is categorized as varus in hindfoot, adductus in metatarsus, and cavus in arch of midfoot. This congenital malformation is predominantly found in pediatric patients while visiting the outpatient department, or it can be seen towards syndromic problems which are associated with any other congenital phenomenon (15-20% cases). The etiology is still unknown, secondary CTEV can be found as the manifestation of meningomyelocele and myotonic dystrophy. Signs of CTEV are the same, but the secondary type is specifically obtained from neuromuscular disorder.[1] Fetal malformation is more likely to occur in the second trimester.[2] Secondary CTEV and idiopathic forms are rather different in presentation, management.[2,3,4] Idiopathic CTEV (ICTEV) is the most common case with prevalence estimated 10-40 cases/year, depending on birth frequency. Incidence of ICTEV increased from 0.6-2.0 cases/year and mostly associated with multiple factors in the gestational period.[5] The incidence increased mostly in middle-income countries, possibly due to limited knowledge, genetics, and exposure during pregnancy.[5] Kruse et al. studied the genetic factor of gender in 2009, but phenotypic variability remains unclear.[6] Ponseti method was chosen as our management option; This study aims to identify the risk factors and their prevention of CTEV.

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CASE REPORT

12-month-old twin girls were brought to the Orthopedic center at Arifin Achmad Hospital by their parents with complaints of club feet since birth, with 2300 grams and 2449 grams birth weight, full term, born spontaneously. During pregnancy history, amniotic fluid was lacking, ANC is about 2-3 times in 9 months, once a history of miscarriages, and family members are smokers. Pregnancy supplement intake, such as drug consumption during pregnancy, deficiency of folic acid, and iron supplement tablets, is unclear. The physical examination was performed and resulted in *cavus, equinus, varus, and adductus* foot on both feet with a Pirani score of 6 **(Figure 1).** Both of them were diagnosed as bilateral CTEV and treated using the Ponsetti method **(Figure 2).**

Figure 1. Both babies presented with Bilateral Congenital CTEV

Figure 2. Post Initial Casting on both babies on their bilateral feet

The patient was fitted with a boot cast on both lower extremities. The first step is to smear the extremities using tincture benzoyl before the cast is installed. This procedure can prevent the baby's foot from disrupting the cast while moving their lower limbs. Boot cast was observed 6 weeks after initial installation and should go back to the polyclinic every week to evaluate the cast condition. The patient was advised by the orthopedic surgeon to remove the cast while bathing. After 6 weeks, the patients are continuing for about 8 weeks to use Dennis Brown splint shoes to adjust foot position specifically while walking. The parents were informed by the orthopedic surgeon to use the boot cast every time either sleep or during activities. To make a greater position of the foot during the patient's development, the boot cast has to be installed during sleep at night for about 1 year. After a year, the patient came back to the polyclinic to evaluate the outcome after using a boot cast and Dennis Brown splint shoes. The result revealed that the patient is now able to walk with slight unstable, there was no sign of abnormality during standing and walking. The foot structure is anatomically improved, and the patient also performs certain physical rehabilitation by the orthopedic surgeon. After using boot cast and Dennis Brown splint shoes, there's an improvement in the patient's foot **(Figure 3).**

Figure 3. An outcome after 6 weeks installation of boot cast

DISCUSSION

Clubfoot is defined as a congenital malformation characterized by deformities of the foot, specifically varus, adductus, cavus, and equinus. This condition may affect the unilateral or bilateral foot. Among musculoskeletal system disabilities, clubfoot is the

most common congenital deformity. The incidence in Indonesia is estimated to be around 0.76 – 3.60 cases per 100 births and is expected to increase worldwide.[7] If the previous children of the family had clubfoot, 10-20% of the other children would get the same condition. The rates of children with clubfoot are doubled in males compared with females. The abnormality found in clubfoot patients is the malposition of tarsal bones that affects the shape of the tarsal joint, so the metatarsal bone becomes flexed, causing calf muscle atrophy and leg shortening.[7]

The etiology of clubfoot is still idiopathic, but some literature says that a genetic component has become one of its factors. Clubfoot is likely to be caused by multiple genetic factors based on complicated inheritance patterns. The underlying predisposition factors of clubfoot are mostly joint and bone malformation with abnormal neuromuscular development.[7]

Several risk factors of clubfoot are oligohydramnios, a history of active or passive smoking during pregnancy, low parental knowledge, multiparity, parental psychology, and alcohol usage. The patient's amniotic fluid was lacking, that mentioned as oligohydramnios. Oligohydramnios causes CTEV by reducing amniotic fluid volume, which leads to intrauterine compression and restricted fetal limb movement, resulting in abnormal fixed foot positioning and deformity.[20,21] Miedzybrodzka also found that oligohydramnios has been linked inconclusively with talipes equinovarus. We found that mothers of 11.66% cases had an obstetrical history of oligohydramnios in this study.[22]

Based on the history taking the patient was a passive smoker because there were many active smokers who lived in the same house. One study showed that maternal smoking during pregnancy affects the incidence of clubfoot. This indicates an interaction between genetic factors of clubfoot incidence and tobacco exposure.[7,8] Also, a case-control study of 100 children under 5 years old who were clinically diagnosed with congenital idiopathic clubfoot shows that maternal passive smoking significantly increased the incidence of clubfoot ($p<0.05$). This is caused by placental and fetal vascular damage by nicotine through the oxidative stress pathway. Nicotine in cigarettes inhibits the nidation of trophoblasts that are controlled by placental Nicotinic Acetylcholine Receptors (NAR). Therefore, placental vascularity is poorly developed. Also, smoking could induce the imbalance of Placental Growth Factor and Anti-Angiogenic marker, which could lead to abnormal placental circulation that will decrease the nutrient exchange between the fetomaternal circulation.[9,10]

In one of the systematic reviews made by reviewing fifty-three studies, environmental elements play an important role in the etiopathogenesis of clubfoot, increasing the risk by 20-fold. This could be affected by an alteration of numerous genes in the clubfoot clinical pathway. The most important genetic involvement is the PITX-1 gene, which was connected to human phenotypes.[11,12] Smoking could affect DNA through the oxidative damage pathway, as one of the meta-analyses shows that 15,673 individuals with clubfoot are significantly linked with maternal smoking. Smoke-derived products could affect congenital birth defects like clubfoot through alteration in N-Acetylation genes such as NAT 1 and NAT 2. The polymorphism of the NAT2 gene could decrease acetylation productivity and be associated with clubfoot. Therefore, the accumulation and biotransformation of DNA and amines may contribute to the development of clubfoot.[8]

Several drug consumptions during pregnancy influence the intrauterine fetal condition. Those drugs, including phenylephrine, diphenhydramine, albuterol, and metronidazole, might induce fetal malformation. High-dose Steroid Anti-Inflammation Drugs (SAID), such as hydrocodone and codeine, were found to be risky for having clubfoot during early pregnancy.[13] The use of anaesthesia drugs such as morphine, fentanyl, and meperidine was also reported (collectively in 2% of case mothers and about 0.3% - 0.6% found in control mothers). Medication for the common cold, prolonged cough, and asthma increased from 0.75 to 1.20 and was associated with the risk of having CTEV. Besides that, antimicrobial agents, especially macrolide group antibiotics, increase the malformation of fetuses during the middle of pregnancy.[13]

Supplementation consumption during early and middle pregnancy also plays a role in reducing the risk of having congenital malformation. In the United

States, consumption of folic acid supplements can decrease the incidence of CTEV. Similar data was also found in every pregnant woman in Denmark, and they also had a lower rate of CTEV incidence.[14] Dunn et al. assumed that oligohydramnios during pregnancy might hinder the movement of the legs while intrauterine. The volume of amniotic fluid usually ranges around 500-2000 ml, which influences the development of the lower limb. This topic is based on the association of clubfoot with the intrauterine mechanical factor guideline.[15]

The specific etiology of CTEV is still unknown, and multiple genetic factors are risk factors for the increasing incidence of CTEV. Family and smoking history are also associated with making it easier to happen. Besides that, some researchers explained that there was no connection between CTEV and the history of pregnancy.[16] Parker and Browne explained that the presence of other conditions in the uterus, such as oligohydramnios, can cause limitations in fetal movement. Some studies show that CTEV is the result of a neuromuscular defect, but other studies show no abnormalities in neuromuscular histologic findings.[17] Sætersdal et al. explained that a bilateral brace had greater outcome than a unilateral brace. There were 94 patients in this study; there were 62 patients with bilateral and 32 with unilateral feet. Children who performed bilateral bracing, which was to prevent relapse, had a greater range of motion and reduced risk for shortening and angulation. The study said that they did not recommend using with unilateral foot abduction brace in clubfoot treatment.[19]

CONCLUSION

Most literature suggests that the role of environmental factors and genetic factors may induce the evolution of CTEV during pregnancy. Due to the multi-factorial aspect, plenty of researchers are needed to investigate those factors. A physician in a primary health care unit should know how to identify clubfoot, the risk factors, and its management. The CTEV management consists of serial casting and bracing, with Dennis Brown shoes showing good results and outcomes.

ETHICS STATEMENT

My institution has exempted ethical approval from reporting this case.

COMPETING INTERESTS

The authors have no conflicts of interest to declare

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UNDERLYING DATA

Derived data supporting the findings of this study are available from the corresponding author on request.

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