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Case Report

Bilateral idiopathic club foot in baby of a rheumatoid mother: A rare case report and its management

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ABSTRACT

Congenital clubfoot has a multifaceted etiology, with several hypotheses offered in its etiopathogenesis. The clubfoot has rarely been reported in babies born to women who have rheumatoid arthritis (RA). We present a rare case of a 31-year-old lady with RA on disease-modifying anti-rheumatoid drugs who delivered a child with bilateral congenital clubfoot. She had previously been using Methotrexate, Hydroxychloroquine, and Sulfasalazine regularly, but Methotrexate was stopped seven months before pregnancy. A full-term female baby was born through the cesarean section with bilateral clubfoot deformity and a modified Pirani score of eight out of 10. The deformity correction was done with the Ponseti serial casting method. The final modified Pirani score was two out of ten. In newborns born to rheumatoid arthritis mothers, the club foot deformity was effectively treated with serial Ponseti corrective casts, as was idiopathic clubfoot in babies born to non-rheumatoid mothers. Our findings validate the Ponseti serial casting method for these kinds of patients.

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1. Introduction

Congenital talipes equinovarus or congenital clubfoot is the most common congenital orthopedic condition requiring intensive treatment. It is characterized by forefoot adduction, mid-foot cavus, heel varus, and ankle equinus.^{1,2} It is caused by congenital abnormalities of multiple musculoskeletal tissues distal to the knee (musculotendinous, ligamentous, osteoarticular, and neurovascular structures).²⁻⁴ It has a multifaceted etiology, with several hypotheses offered in its etiopathogenesis, including an in-utero mechanical block, a primary germ-plasm defect in the talus, myofibroblast contraction leading to retraction fibrosis neurogenic, myogenic, and vascular theories.⁵ The incidence of congenital clubfoot in the general population is 1 to 2 in 1000 live births.⁶ During

pregnancy, rheumatoid arthritis has a more favorable prognosis and requires fewer immunomodulatory drugs to maintain disease activity levels. The majority of women experience flare-ups during the postpartum period and the requirement of immunomodulator is increased.^{7,8} Children born to RA mothers have been reported to be healthier and have a higher mean birth weight than children born to nonrheumatic mothers if they are in remission.⁹ A higher risk for congenital abnormalities in the offspring of pregnant women with RA was not found. Thus, contrary to popular belief, RA has a favorable effect not just on pregnant mothers, but also on their fetuses.⁹ However, clubfoot has rarely been reported in babies born to women who have rheumatoid arthritis (RA). We present a rare case of a 31-year-old lady with RA on disease-modifying anti-rheumatoid drugs who delivered a child with bilateral idiopathic clubfoot.

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2. Case Report

A two-week-old girl born in a non-consanguineous marriage to a 31-year-old mother with RA presented to us with bilateral ankle and foot deformities. The mother had a history of rheumatoid arthritis diagnosed at seventeen years of age. She is being treated with disease-modifying anti-rheumatoid drugs (DMARD) since 2012. Her DMARD regimen was begun with Injection Tocilizumab 400mg administered four weeks apart for six months, followed by oral Methotrexate, Hydroxychloroquine, and Sulfasalazine. She underwent a total knee arthroplasty of the right side in September 2019. Methotrexate was discontinued seven months before the conception. However, Hydroxychloroquine and Sulfasalazine tablets were continued in all three trimesters of pregnancy. She received regular Folic acid supplementation before conception and during the pregnancy. A full-term female baby was delivered by cesarean section. APGAR score was eight on ten at one and five minutes, and baby weight at birth was 3 kilograms. On examination, the child had normal reflexes, was accepting feed normally, and there were no cyanotic spells. The vitals, oxygen saturation, urine output, and systemic examination were normal. The upper to lower segment ratio was normal. There were no signs of spinal dysraphism, and neurofibromatosis. The upper extremities were normal. Both ankles and feet were in an abnormal position, with forefoot adduction and pronation, cavus in the midfoot, heel varus in the hindfoot, and equinus in the ankle joints, indicating a bilateral clubfoot deformity. The clubfoot deformities were evaluated with the modified Pirani score, which was scored eight out of ten (Figure 2). The deformity correction was begun at two weeks of age by the Ponseti serial casting method. The Ponseti casts were changed at weekly intervals (figure 3). The forefoot adduction and heel varus deformities were corrected in five Ponseti casts. The equinus deformity was treated with Botox injections, followed by an above-knee cast with the ankle in neutral dorsiflexion for one week, then 20 degrees dorsiflexion for two weeks. Following deformity correction, the final modified Pirani score was two out of ten (Figure 3). For three months, a foot abduction brace was worn for 23 hours per day to keep the deformity corrected, after which it was only worn at night and during naps (figure 4). The child has normal developmental milestones and is thriving well. The mother has achieved sustained remission in RA.

3. Discussion

RA, a chronic autoimmune inflammatory disease with significant physical impairment, affects women three times as many as men, often in childbearing years.^{7,10} Pregnancy hormones, such as progesterone suppress the immune relationship between the mother and the fetus by modulating



Fig. 1: Bilateral idiopathic clubfoot at two weeks of age showing all characteristic deformities



Fig. 2: Correction done by serial ponseti casting method



Fig. 3: Complete correction of all deformities after five serial Ponseti casts



Fig. 4: Maintenance of correction by a foot abduction brace

Th1/Th2 cytokine balance, resulting in a favorable pattern of RA in pregnancy.^{11–13} Pregnancy's complex hormonal and immunological changes may have an impact on rheumatoid arthritis immunoregulation. Some studies have suggested that decreased interferon-gamma production and an imbalance in interleukin six and interleukin 12 production are to cause RA remission during pregnancy.¹⁴ The maternal immune response to fetal paternally acquired class II HLA antigens, CD8+ and CD4+ T cells, placental gamma globulins, and alterations in serum fetal DNA levels (trophoblast generated) may also play a significant role in pregnancy-induced RA remission.^{15,16} The effect of RA on fetuses in a pregnant woman is less well understood, with scant evidence on the risk of structural limb abnormalities. Although pregnancies with RA have not been linked to an increased risk of congenital anomalies in the fetus, anomalies, undescended testis, polydactyly, and syndactyly in rheumatoid arthritis fetus as in the general population. The clubfoot has rarely been reported.⁹ A higher incidence of musculoskeletal deformities in Hydroxychloroquine administered rheumatoid mothers has been shown. However, the type of anomaly has not been specified. Hence, we are reporting this rare case of bilateral clubfoot in RA's mother.¹⁷ Ventricular septal defects and coarctation of the aorta have been described in babies born to mothers who have received sulfasalazine during pregnancy. However, musculoskeletal abnormalities have not yet been reported as their teratogenicity.¹⁸ This infant was exposed to Sulfasalazine throughout pregnancy (500 mg two times a day)

The most common age group for rheumatoid arthritis in Indian women is between the ages of 35 and 45 years,¹⁹ while the median age at first pregnancy among married women in India is 21 years.²⁰ Thus, RA occurs in a later age group, which might be one of the probable explanations for the low incidence of clubfoot in babies born to RA mothers. The standard treatment of choice for idiopathic clubfoot is the Ponseti serial casting method, which consists

of two distinct stages of manipulation and maintenance. The manipulation phase includes using the talus head as a fulcrum, supinating the forefoot with pressure on the first metatarsal to eliminate the cavus deformity, and then abducting the forefoot. This is followed by the application of a plaster cast, which holds the foot in the correct position and allows enough time for soft tissue remodeling. It is repeated weekly for an average of 5-6 weeks until the abduction of the forefoot is reached by 50 degrees. The equinus deformity must be corrected by serially stretching the tendoachilles to achieve dorsiflexion of the ankle joint and lengthening the tendoachilles with a tenotomy or Botox injection in resistant cases. It is then followed by three weeks in a supporting cast to aid in healing the lengthened position. During the maintenance phase, the foot is kept in an abduction brace for 23 hours per day, reducing recurrence rates.²¹ We successfully corrected clubbed foot deformity in a baby of a rheumatoid arthritis mother using the Ponseti serial casting method. Our findings validate the Ponseti serial casting method for these kinds of patients.

4. Conclusion

Bilateral clubfoot in babies born to rheumatoid mothers is a rare presentation. It can be successfully treated with serial Ponseti corrective casts similar to idiopathic clubfoot in babies born to non-rheumatoid mothers.

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6. Conflict of Interest

The authors declare they have no conflict of interest.

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