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Effect of Pelvic Floor Muscle Stimulation in Paediatric Paraspinal Ewing's Sarcoma with Cauda Equina Syndrome - A Case Study

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ABSTRACT

Background: Ewing's sarcoma causing cauda equina syndrome is a rare entity. Cauda equina syndrome causes cord compression due to which there may be loss of bowel –bladder control

Clinical Description: We report a case study of a 4-year-old female, case of paraspinal Ewing's Sarcoma with cauda equina syndrome having bowel-bladder incontinence. The patient while undergoing chemo-radiotherapy was referred to physiotherapy department for management of bowel-bladder incontinence.

Management & Outcome: Pelvic floor muscles stimulation (PFMS) i.e. faradic stimulation was given for 6 weeks to the vaginal and anal area. Pelvic floor muscle exercises were started along with PFMS. Assessment was done at baseline and post treatment completion using the Modified Oxford Scale (MOS) for pelvic floor muscle strength, Manual Muscle testing (MMT) for Lower limb muscle strength along with lower extremity functional scale (LEFS), Paediatric balance score (PBS) and the Childhood bladder and bowel dysfunction questionnaire (CBBD).

Conclusion: There was significant improvement in LEFS, PBS and CBBD. PFMS along with exercises is safe and feasible to improve bowel-bladder dysfunction in paediatric patient with paraspinal ES with cauda equina syndrome.

Keywords: Pelvic floor muscle stimulation, Paraspinal Ewing's sarcoma, Cauda equina syndrome

INTRODUCTION

Ewing's sarcoma (ES) is a poorly differentiated, highly malignant, round cell tumour without cellular or structural differentiation. (1) First described by James Ewing in 1921, ES of bone is the most common malignant tumour in the first decade of life. (2) It can affect any part of the skeleton but most commonly affected areas are the ilium and the diaphysis of the femur and tibia. Extra-skeletal ES was first described by tefft et. al in 1969, arising in various locations like the chest wall, paravertebral muscles, extremities, pelvis and retroperitoneal space. (3) Patients may present with neurological deficit when the tumor extends into the spinal canal, causing compression of the spinal cord. (1)

Cauda equina syndrome is a severe neurological condition and surgical emergency caused by compression of the nerve roots at the lower end of the spine, leading to symptoms like lower back pain, numbness, lower limb weakness and loss of bowel-bladder control, requiring immediate intervention. (2)

Conservative treatment like faradic stimulation is used for pelvic floor muscle stimulation (PFMS). Faradic current is a short duration interrupted direct current with a pulse duration 0.1 to 1 millisecond (ms) at a frequency of 50-100 Herts (Hz), which helps in strengthening weak muscles by increasing their muscle tone and blood flow which also aids in muscle re-education. [(4)] It also provides biofeedback creating awareness around the pelvic floor aiding in improving muscle control. [(5)]

Few studies which have administered PFMS along with exercises has shown improvement in pelvic floor muscle strength in pediatric population. [(5)]

Here, we present a case of a 4-year-old female child a case of paraspinal ES with cauda equina syndrome referred to physiotherapy with complaints of lower limb weakness, bowel and bladder dysfunction, with no sensory deficit in the lower limb and pelvic floor region.

MATERIALS & METHODS

Clinical Description:

A 4-year-old female diagnosed as a non-metastatic ES of paraspinal region (D12-L3) with cauda equina syndrome treated with chemotherapy 24 cycle of VIDE (Vincristine, etoposide, adriamycin, Ifosfamide) and 50.4 Gy /28 of radiotherapy presented at physiotherapy department, Tata memorial hospital with complaints of bowel - bladder dysfunction and lower limb weakness. The patient had significant bladder dysfunction presenting with urinary incontinence, for which the patient was catheterized. The bowel dysfunction presented as fecal incontinence for which the patient used diapers throughout the day. She had developed ulcerative rash in the perineum which was treated with regular dressing. The patient also had weakness in bilateral lower limb with foot drop. She was using ankle foot orthosis and was able to walk with a walker. Sensory examination revealed normal sensation in the lower limb and in the pelvic floor muscles. Dynamic balance was affected in standing.

Assessment:

General assessment included the patient's treatment history (radiation, chemotherapy), physical examination (pelvic floor muscles strength, muscle charting for lower limb muscles), Functional assessment and symptom severity of bowel -bladder. Although there are no standard guidelines for the assessment of Pelvic floor muscle strength for the pediatric population, assessment tools recommended for the adult population were used in our case report.

Detailed assessment of muscle strength was done by using Manual Muscle Testing (MMT) for lower limb and Modified Oxford Scale (MOS) for pelvic floor muscles. Paediatric balance score (PBS) was used to assess the balance and Lower limb Functional assessment was done using the Lower Extremity Functional Scale (LEFS). Childhood bladder and bowel dysfunction questionnaire (CBBD) was administered to assess the severity of bowel and bladder symptoms. All the assessments were done at the baseline and 8 weeks after completion of intervention.

Details of the various assessment tools used are provided below

MMT: It is a subjective, clinician administered assessment tool to judge muscle strength by measuring the capacity of the muscle to move against gravity when resistance is applied. Grading is from 0-5, with 0 signifying no contraction and 5 indicating normal strength

MOS: This scale is a subjective grading system used to assess the pelvic floor muscle strength by digital examination via vaginal or anal opening. The score ranges from 0-5, with higher score signifying strong squeeze pressure with inward/upward lift of the pelvic floor towards the umbilicus

LEFS: It is a valid patient-related outcome measure for the measurement of lower extremity function. It contains 20 questions about a person's ability to perform everyday tasks. The score ranges from 0 (extreme difficulty)-4 (no difficulty) for each question. Lower score correlates with greater disability. [(6)]

PBS: PBS is a modified version of the Berg Balance scale, specifically adapted to assess functional balance skills in children with mild to moderate motor impairments. The scale consists of 14 balance-related test items that mimic everyday living experiences, with each item scored from 0 (lowest function) to 4 (highest function), yielding a maximum total score of 56 points. Higher scores mean better balance and low fall risk. [7]

CBBD: It is a parent reported tool consisting of 18-item covering 10 bladder and 8 bowel symptoms with bowel and bladder dysfunction. The score ranges from 0 (never) - 4 (almost daily) for each item. Higher scores indicating greater symptom severity indicating poor quality of life. This scale was developed for pediatric population of age above 5 years but due to non-availability of scales for younger age group this scale was used. [8]

Our patients' baseline status of Hip and knee muscle power was 3 (full range of motion against gravity) and ankle power was 0 (no contraction). Likewise, Pelvic floor muscle strength was 0 (absent). Dynamic balance showed affection which reciprocates with being unable to maintain balance during walking.

Management and Outcome:

Intervention: The patient was treated with surge faradic stimulation to the pelvic floor muscles to provide reeducation, along with strengthening exercises to the pelvic floor and lower limb, to improve balance and gait training. Supervised Physiotherapy interventions were administered for a period of 6 weeks along with a home program which was taught to the parents.

Procedure:

PFMS: The child was made to lie in supine position (hook-lying). Electrical stimulation was given using diagnostic muscle stimulation (DMS-4) with two flat electrodes (our modification). Both flat Electrodes were placed over the vaginal and anal areas bilaterally (figure 1&2). The electrical stimulation was administered using faradic current, with pulse duration of 1 ms, frequency of 30 Hz with varying Intensity of the current at each session. The intensity of current was increased gradually till observable muscle contraction was seen in the form of a vaginal and anal wink. The patient was asked to do voluntary vaginal and anal contraction with each stimulus. A total of 90 contractions were given in each session, wherein 30 contractions were followed by 15 seconds of rest period. This was continued for 5 days per week for 8 weeks.

Figure 1: Vaginal stimulation

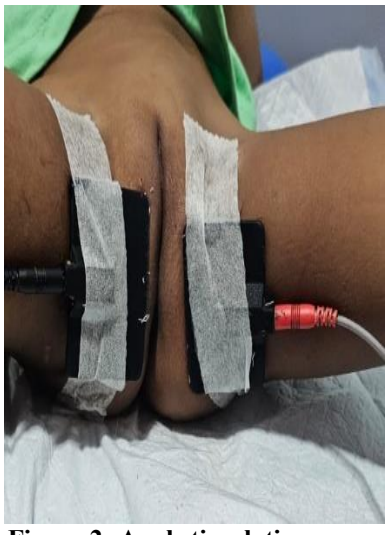

Figure 2: Anal stimulation

Exercises:

Pelvic floor muscle strengthening in the form of supervised pelvic floor muscle exercises, along with tightening of the abdominal muscles, adductors and gluteals and drawing up the pelvic floor muscles for 5 seconds and then rest. [5] Pelvic tilts, bridging, rolling, quadripods and squats with support were started.

As the Pelvic floor muscle strength improved, weaning of diapers and catheter was done along with toilet training i.e. passing the urine every 2 hours to prevent leakage and monitor fluid intakes.

Stretching exercises for bilateral Dorsiflexors were done for 3 repetitions with 10 seconds hold which was taught to the carer so as to be carried out at home. Exercises were begun as passive then progressed to doing movements in gravity eliminated plain to recruit dorsiflexors muscle were done as the strength improved.

Strengthening exercises focused on core, spinal extensors and lower limb resistance exercises with weights of 250 gm. Progressive balance exercises started in standing with wide base support, weight shifting, perturbations followed by narrow based, tandem standing with and without eyes closed, single leg standing and progression to unstable surfaces like the wobble board and Frenkel's exercises for lower limb co-ordination. (figure 3) The patient was asked to do the exercises as frequently as possible with at least a minimum of two times daily.

Figure 3: Exercises

Gait training with support was initiated using the Ankle foot orthosis. Play activities like ball kicking, obstacle walking, figure of 8 walking was also included for progression. During the course of her intervention, support was gradually reduced and patient started walking with minimal to no support.

RESULT

Significant improvement was observed in the pelvic floor muscle strength which was measured using the MOS, demonstrating improvement in power from grade 0 to grade 4. Improvement in faecal incontinence was observed post 10 days of starting stimulation and exercises. This resulted in gradual weaning of diapers, which was used for faecal incontinence. Also, the urinary catheter was removed 2 weeks later as the child was able to sustain urine control for a prolonged period of time.

Likewise, overall muscle strength in bilateral lower limb measured using MMT showed significant improvement of dorsiflexors strength from grade 0 to grade 2 (table 1).

Table 1: Comparison between the strength measurement was done pre and post intervention.

Muscle Strength	Pre intervention	Post intervention
Pelvic floor muscle	0	4
HIP	3	4
KNEE	3	4
ANKLE	0	2

Functional independence of lower limb function and balance was measured using LEF scale and PBT, their scores improved from 20 to 64 scores and 0 to 41 respectively, reflecting considerable improvement. The overall CBBD scores

were remarkably reduced from 61 to 7, with considerable improvement in the bladder function score which reduced from 35 to 4 and bowel function which reduced from 26 to 3 post intervention showing substantial improvement in symptoms. (Table 2).

Table 2: Comparison between the outcome measures was done pre and post intervention.

Assessment tools	Pre intervention	Post intervention
Lower extremity functional scale	20	64
Pediatric balance test	0	41
Childhood bladder and bowel dysfunction questionnaire	61 (TOTAL) 35 (BLADDER) 26 (BOWEL)	7 (TOTAL) 4 (BLADDER) 3 (BOWEL)

DISCUSSION

The present study was designed to evaluate the effectiveness of PFMS in paediatric paraspinal ES with cauda equina syndrome.

In our case study, we found significant improvement in pelvic floor muscle strength and function which is similar to the study conducted by Ratan et.al., where in 28 paediatric patients showed significant improvement with faradic stimulation and exercises. [(9)] Faradic stimulation has been used widely in the adult population as it doesn't cause irritation when used with caution. Additionally, it produces titanic contraction and relaxation of the muscle which helps in muscle recruitment. [(9)] Similar current when used in paediatric population showed significant improvement in bowel and bladder function in rectal prolapse and numerous case studies. The population treated were above 5 years of age. In our study the child was < 5 years, and the study demonstrated that faradic current helped in improving the bladder bowel incontinence. Similar to our case report Alzayer, in a case series conducted on 5 pediatric patients with bowel and bladder incontinence secondary to myelomeningocele reported improvement in symptoms with faradic reeducation and pelvic floor strengthening from age of 4 years and above. [(5)] Pelvic floor muscle strength showed significant improvement which was similar to the study conducted by Chaharsoghi et.al. on 27 pediatric patients wherein Kegels exercises showed improved bowel health, although it was done in children with constipation. [(11)]

As the child in our study was less than 4 years old it was difficult to assess, explain and teach exercises. Also finding good subjective and objective assessment tools was difficult. Chase et.el has highlighted several gaps in pediatric research due to nonavailability of validated and reliable Standardized measurement tools for pelvic floor muscle assessment, strength and endurance estimations or specific investigations like electromyography may not be feasible due to many confounding factors e.g invasive procedure, posture, breathing, fear. Another important factor is variability in exercise protocols like type, time and repetition. [(12)]

In our study strength improved with 6 weeks of supervised exercises especially in the foot, wherein the child presented with footdrop initially and was using splint to ambulate this could be due to improvement in strength of lower limb as seen in Kafadar et al case report presenting data of 6 cases having lower limb weakness and difficulty in ambulation out of which 3 patients had paraplegia with urinary incontinence. This study specifically highlights the cause of footdrop to be nerve root compression due to lumbosacral level affection which was correlating with our case study. Kafadar emphasized on possibly good neurological recovery secondary to treatment of surgical decompression, chemotherapy and radiation. Likewise in our case report, the patient showed complete recovery in strength, footdrop and hence gait. Although rarely some deficits may remain requiring aids and assistance. [(13)]

CONCLUSION

PFMS is safe, feasible and helps to improve bowel and bladder incontinence in children below 5 years having paraspinal ES with cauda equina syndrome.

Declaration by Authors

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