QUESTION 2
Do standing frames and other related physical therapies reduce the risk of fractures in children with cerebral palsy?

SCENARIO

A 9-year-old girl with non-ambulant cerebral palsy (Gross Motor Function Classification Scale (GMFCS) level 4) attends clinic. Despite adequate calcium and vitamin D supplementation, a recent dual-energy x-ray absortiometry (DEXA) bone scan shows a bone mineral density in the osteopenic range (Z score −2.6). Her mother is anxious about the risk of fractures and asks what can be done to reduce the risk.

STRUCTURED CLINICAL QUESTION

In children with osteopenia secondary to cerebral palsy, do weight-bearing activities (including standing frames, vibration platforms and physiotherapy) reduce the risk of fractures?

SEARCH STRATEGY

A PubMed search using MeSH terms bone density AND cerebral palsy AND physi* gave 26 results. A wider search of MEDLINE, Cochrane and Trip databases was searched for the terms ('cerebral palsy') AND ('physical therapy' OR 'vibration' OR 'physiotherapy' OR 'passive standing' OR 'dynamic standing') AND ('fracture' OR 'bone density'). Fifty-six results were found. No appropriate papers were found on Cochrane review. Nine papers were assessed and included.

COMMENTARY

Children and adolescents with cerebral palsy are prone to fragility fractures that can occur during normal activities such as dressing and transferring. Peripheral and vertebral fractures are associated with low BMD. Children who are non-ambulant (GMFCS level 4 or 5) are at the greatest risk; most will have osteopenia (BMD Z-score <2) and more than a quarter suffer a fracture, most commonly of the lower limb, by age 10 years.[10] Fractures impact on quality of life, causing pain and further limiting the mobility of children, leading to muscle wasting through disuse, hospitalisation and missed schooling.[11]

We chose to focus on physical therapy as there is an uncertainty regarding best practice. The recommendations frequently quoted are 60 min of mechanical loading, 4–5 times per week, which were non-evidence-based proposals made by Sturberg in 1992.[12]

There are other interventions to improve BMD, which are not considered in this report. Vitamin D and calcium supplementation are recommended given the possible effectiveness and their good safety record. Bisphosphonates increase bone mass by inhibiting osteoclastic activity and are currently considered for children who have experienced a fragility fracture.[13]

Weight-bearing stimulates osteoblastic activity, leading to new bone growth[14] with the assumption that this will decrease the risk of fractures. There is not yet evidence for this population of actual fracture risk reduction. We reviewed the evidence for static and dynamic therapy as well as for other related augmentative physical therapies. Interventions evaluated were dynamic standers, passive standing frames, whole-body vibration, physiotherapy, resistance training and treadmill training. Dynamic standers have footplates that are incorporated into the existing standing frames to provide reciprocal loading that mimics the forces applied to the lower limbs during the natural walking gait.[15] Whole-body vibration therapy uses a vibrating platform that the user stands on in a static position or moving in dynamic movements.

Overall, there is some evidence that physical activity, including standing, whether passive or dynamic, improves BMD. There is limited evidence that the improvement in bone density is in the areas most susceptible to fracture, and no evidence yet available that actual fracture rates are decreased. Findings from studies conflicted as to whether dynamic standing provided any additional benefit to BMD, and the small number of participants means no conclusion can be drawn. Vibration therapy may provide some benefit to BMD in ambulant children, but there is no evidence of benefit in those unable to stand. It is unknown whether this is valid finding or a reflection of the challenge of conducting these studies. A combination of all therapies was suggested to significantly improve BMD, but it is not possible to make generalisations of benefit outside of this case series. Therapies appear generally well tolerated, and no significant adverse events were reported in these studies.

Dynamic and combination therapies attempt to recreate the weight-bearing and muscle tension forces of ambulation that stimulate bone growth. It might be reasonable to presume that this style of therapy provides the best stimulus for bone growth, but studies to test the hypothesis are challenging because of the heterogeneity of children with cerebral palsy and the confounding factors of other therapies and conditions. Study sample sizes, particularly those that found benefit of additional therapies, are generally very small and non-blinded. Consequently, the combination and optimal dose of therapy (duration and frequency) remain unknown. Evidence from larger-scale studies of pragmatic real-life interventions, with matched control group, would be necessary before additional time-consuming therapies can be recommended.

Table 1

Citation	Study group	Study type	Outcome	Key result	Comments
Caulton *et al*, 2004[1]	26 non-ambulant children with cerebral palsy; randomised to standing programme (50% extra standing) for 9 months	Randomised controlled trial (level 2b)	Vertebral and proximal tibial BMD measured using CT at onset and 9 months	6% greater vertebral BMD in treatment group (p=0.01) No increase in proximal tibial BMD	Lower limb long bones are the commonest site of fracture, and this study suggests that extended standing does not influence BMD in those bones
Chad *et al*, 1999[2]	18 children with spastic cerebral palsy; randomised to control or weight bearing activity 1 h 2–3 times/week for 8 months Intervention and control groups were matched and included ambulant and non-ambulant children	Randomised controlled trial (level 2b)	Femoral neck BMD and BMC at 8 months and BMC at 8 months	Intervention group showed increases in Femoral neck BMD of 5.6% (p=0.02), femoral neck BMC of i 9.6% (p=0.03) and Proximal femur BMC 11.5% (p=0.08)	The percentage changes in bone density are only statistically significant when compared with reduction in density of the control group. The intervention was not clearly described, ('normal movement with emphasis on weight bearing') with controls not offered any 'standard' physiotherapy
Ward K *et al*, 2004[3]	20 disabled ambulant children; randomised to standing on active or placebo vibration devices for 10 min/day, 5 days/week for 6 months	Randomised controlled trial (level 2b)	Proximal tibial BMD and Spine BMD at 6 months	Proximal tibial BMD: active average increase 6.27 mg/mL (+6.3%) Placebo decrease 9.45 mg/mL (−11.9%) No change in diaphyseal bone, spine and muscle	There is only a small increase in BMD; however, a large net difference is reported due to a decrease in BMD on the placebo devices Compliance was only 44% and 3 children withdrew
Eisenberg *et al*, 2009[4]	22 non-ambulant children with spastic quadriplegic cerebral palsy; randomised to gait training walking device or a passive standing programme	Randomised controlled trial (level 2b)	Bone quantitative ultrasound was performed for the tibia Outcome at 6 months	No added quantitative benefit	Well tolerated, but no effects on bone identified. Bone density was just one of a number of outcomes
Gudjonsdottir *et al*, 2002[5]	4 non-ambulant children; randomised to static or dynamic stander for 30 min a day, 5 days a week for 8 weeks	Randomised trial (level 2b)	BMD in lumbar spine, proximal femur and distal femur At onset and after 2 months	BMD increased in both children in dynamic stander, and one of the two children in the static stander	The standers were equally well tolerated, but no conclusions can be drawn from this study of only 4 children
Wren *et al*, 2010[6]	31 independently standing children with cerebral palsy; randomised to standing, or a vibrating platform for 10 min/day for 6 months, then groups swapped intervention	Randomised cross-over trial (level 2b)	CT scan measurements of vertebral and tibial cancellous and cortical bone density At onset, and after 6 and 12 months	Increase in cortical bone area in the tibia (p<0.03) No difference in cancellous bone or muscle	Participants were ambulant with or without aids. Promising study, but unproven clinical significance. Findings cannot be assumed applicable to non-ambulant children
Ruck *et al*, 2010[7]	20 children with cerebral palsy (GMFCS levels 2–4); randomised to 9 min of side-alternating whole-body vibration per school day in addition to their school physiotherapy programme	Randomised controlled trial (level 2b)	BMD at 6 months	No significant differences between groups	Well tolerated, although non-ambulant children were less able to manage standing therapy
Stark *et al*, 2010[8]	78 children with all levels of bilateral cerebral palsy were given a novel therapy concept that included whole-body vibration, physiotherapy, resistance training and treadmill training	Case series (level 4)	BMD Bone mineral content Muscle mass Muscle force Gross motor function At onset and after 6 months of therapy	Percentage changes were highly significant for BMD, content and muscle mass	Retrospective data analysis of combination therapy, with no control group Unable to identify the impact of each individual therapy on BMD
Kilebrant *et al*, 2015[9]	19 non-ambulant children with motor disability. Self-controlled whole-body vibration platform 5–15 min twice a week for 6 months	Case series (level 4)	Total body BMD. Biochemical markers of bone metabolism At 6 and 12 months	Increase in BMD, but no change in Z-score	No evidence of benefit as Z-scores unchanged Well tolerated intervention, and fracture history well described

BMC, bone mineral content; BMD, bone mineral density; DEXA, dual-energy X-ray absorptiometry scan; GMFCS, Gross Motor Function Classification Scale.

Clinical bottom line

- There is some evidence that standing therapies increase bone mineral density in non-ambulant children, which it is hoped might therefore reduce fracture risk (grade c).
- Vibration and dynamic physical therapy programmes may have additional potential for improvement in bone mineral density, but evidence of clinical value has not been established (grade c).
- Combining different activities and therapies may seem a reasonable approach, but there is a lack of evidence for the optimum combination and duration of therapy (grade d).

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