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Article Title

Ankle-foot orthosis adherence in children and adolescents with Cerebral Palsy: A scoping review

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Conflict of Interest

Alice Faux-Nightingale has received a research grant from the Orthopaedic Institute. Mihaela Kelemen and Caroline Stewart declare that they have no conflict of interest.

Abstract

Background: Ankle-foot orthoses (AFOs) are commonly supplied to children with Cerebral Palsy (CP) to support their gait. However, usage is reported to decrease through adolescence[1]. Since AFOs can allow users to engage in daily activities and develop their independence[2], a wider understanding of non-adherence is essential to determining the most appropriate ways to support orthotic prescription for children with CP in the future.

Objective*(s):* This scoping review will present the literature which investigates AFO adherence of children or adolescents with CP and identify potential avenues for future research and practice.

***Eligibility criteria*:** Papers which investigated AFO adherence in children/adolescents with CP, either through usage or factors which could affect usage, were included in this review.

***Charting methods*:** Data were synthesised using a charting form developed for this review.

Methods*:* A literature search was carried out using the EBSCO and Web of Science databases to identify literature which investigates AFO adherence by children with CP through measurement of AFO usage and exploration of factors which may influence that usage.

Results*:* In total, three papers were included in this review. Two included assessments of AFO usage and all three included at least anecdotal references to factors which could influence that usage. Variation in usage time was seen across participants in all studies, though the method used to record usage may influence reported usage values. Key factors which could affect usage were observed in four key categories: physical/AFO related factors, personal factors, social factors, and situational appropriateness.

Conclusions*:* Adherence is a complex subject, both measurement of usage and factors which influence usage are key components needed to understand how children engage with their AFO. By better understanding the motivators and barriers to adherence, it is possible to better support the provision of

AFOs in the future.

Abstract word count: 293 words

Keywords: AFO, ankle-foot orthoses, adolescence, adherence, cerebral palsy,

1. Background

Rationale

Ankle-foot orthoses (AFO) are assistive devices commonly supplied to children with cerebral palsy (CP). They support from below the knee to around the ankle and under the foot and can provide stability and support to users and facilitate more efficient walking [1]. AFOs can allow users to keep active, engage in daily activities, and develop their independence [2], reducing the chance of further physical deterioration[3]. However, such outcomes are dependent on adherence to the device.

Adherence refers to a user's engagement with the recommendations of their clinical professional regarding medical devices or prescriptions[4]. In the case of AFOs, it refers not only to wearing an AFO but also wearing it for the amount of time recommended by a clinician. An early study identified that AFO adherence of people with CP varies across age groups but is particularly varied during childhood and early adolescence. Young children (aged five-11) are reported as having some of the highest AFO usage, with 67% of five year olds reported as using them, but as children develop through adolescence (14-19 years old) this decreases, down to only 16% of 19 year olds reporting usage[1]; similar nonadherence has been observed in other medical areas[5-9]. Youth under 18 are also suggested to have the lowest satisfaction levels regarding their AFOs[10] and as being reluctant to wear their AFOs[11]. Low usage in adolescents also correlates with findings which show that children with CP often lose functional ability through adolescence and early adulthood [12 13], though no causative link has been made. Given the potential physical and social benefits of using AFOs, a wider understanding of this behaviour is essential to determining the most appropriate ways to support children with CP in the future.

This scoping review will present the literature which investigates AFO adherence of children or adolescents with CP and identify potential avenues for future research and practice. Adherence is a complex concept, influenced by a number of interconnecting factors[14] but can be broken down into two key elements: the amount of time that the AFO is used compared with the recommended usage time, (here referred to as 'usage'); and factors which influence that usage. These are often investigated separately in the literature, but both are essential for a holistic understanding of adherence.

2. Methods

Research databases EBSCO, Google Scholar, and Web of Science were used to identify relevant literature; EBSCO was used because of its ability to include searches within multiple multi-disciplinary research databases including MEDLINE, CINAHL, AgeLine, AMED, and APA PsychInfo. The most recent search was carried out during September 2021.

The following search terms were applied to all filters to identify papers relating to AFO adherence by children and adolescents: "(child* OR "young adult" OR adolescen*) AND (CP OR "cerebral palsy") AND (ortho* OR "ankle foot" OR AFO) AND (compliance OR usage OR adherence)". All publication years were included, however papers in languages other than English were excluded due to associated cost and time of translation. Duplicate articles were removed from the results and then papers were selected based on relevance according to title and abstract. The remaining papers were read in full and assessed for relevance, those which reported on adherence or the usage of AFOs by children or adolescents with CP, where children or adolescents were defined as any age up to 19 years, or factors which may influence the usage of AFOs by children with CP were included in this review. Relevant papers

referenced in already selected papers were also included if they had not appeared in the initial research database searches.

Data were collated using a data charting form developed for this review. Details from the publications were input into the table e.g. author information and details about the participants and findings were collected. This data charting process was the starting point of the analysis and was used to synthesise the results, describing the records of usage and factors which influence usage of AFOs by children with CP.

3. Results

Selection of sources of evidence

112 papers were initially identified in the literature search, see figure one for a flow chart of the selection process.

[Figure one: Paper selection process]

Of the identified papers, only three included information about AFO usage by children or adolescents with CP, and one additional paper was identified through key references; the remaining articles did not specifically comment on the use of AFOs by children or adolescents with CP and so were not included. A full list of papers included in this review can be found in Appendix 1. A summary of the findings can be found in Table One.

[Table One]

AFO usage

Two papers included assessments of AFO usage: one measured wear time using temperature sensors built into the AFOs[15], while one asked the child to report their usage through a questionnaire[16]. One paper reported lower wearing times than had been recommended by clinicians[15]. Variation in usage times was seen across the participants in both studies[15, 16], with some indication towards patterns of use[15].

A third paper in this review did discuss usage, however Maas et al.[17] describes patient adherence to a KAFO style device. The orthosis in this case is designed to provide a stretch during resting, rather than to enhance gait, and is prescribed to be worn overnight. This contrasts with the other studies which consider AFOs which are worn by children during the day and can be used while the child is active. The orthosis design and purpose is different from that of a standard AFO and so the study by Maas et al.[17] will be discounted from the results about AFO usage and those about factors which affect that usage. Although discounted from the results about AFO usage, the Maas et al. study provides useful insights into methods used to measure AFO usage, reporting notable variation between measurements collected using a temperature sensor and usage as measured by the user's parent[17].

Factors which affect AFO usage

All three papers (Maas et al. excluded[17], see above) included examples of varying AFO usage patterns and suggested factors or circumstances which were likely to influence individuals' usage of AFOs. Although only one study directly discussed factors which influenced usage of AFOs[16], the other two papers mentioned anecdotal cases which suggested factors that could influence usage.

Key factors identified in this review can be put into the following categories:

- Physical/AFO related factors – AFO tuning was associated with increased wear times for some participants[16]; discomfort or pain due to the AFO was viewed negatively[16] and associated with reduced wear[18]; tuned AFOs were described as supporting the user with their activities and contributed towards positive perception of gait[16].
- Personal factors – Aesthetic or cosmetic consideration of the device and associated footwear was raised as a factor of significance to children with CP but not reported as affecting the wear time[16]; AFOs were described as an object which interacts with identity, with individual perception of the device being affected by personal identity constructs[18], though this was not reported as affecting the wear time.
- Social factors – Peer perception of the device was raised by children who use AFOs but not reported as affecting wear time[16, 18]; parents and other figures of authority were described as affecting the child's level of AFO usage[18].
- Situational appropriateness – Anecdotal evidence was present to suggest that children established their own rules for AFO use according to the activity, location, and any impediment or discomfort which the AFO imposed on the user in those scenarios[18]; usage was reported as varied between the weekdays and weekends, though no further investigation was made into this observation[15].

None of these factors were identified consistently across participants in the papers.

4. Discussion

This scoping review presents a summary of the current research of the adherence of AFOs by children with CP, as investigated through usage and factors which may influence usage. The review notes that there is a lack of literature in this area with only one paper considering both the extent that children

wear their AFOs and factors which interact with that wear time, and none of them investigating adherence in depth or seeking to understand the extent that factors can influence usage; furthermore, all papers exhibited low participation numbers with one of the four a pilot study. It was apparent from these papers that while there may be similarities and patterns across the papers, usage and factors which affect usage are likely to be unique to the AFO user and will need to be considered on an individual basis.

Usage

Adherence was notably varied across the studies with papers reporting considerable heterogeneity within the data. One of the papers which objectively measured usage using temperature sensors reported lower usage levels than had been recommended[15], although with so few papers in this review this is not a firm finding and would benefit from being investigated in greater detail in the future.

One paper in the review compared methods to measure AFO usage and identified variation in recorded wear times according to the monitoring method used[17]. The paper highlights the importance of considering methodology carefully when conducting research into AFO adherence, and potential problems with using subjective methods (i.e. questionnaires or interviews) of data collection for accurately measuring wear time where the data is produced retrospectively by invested stakeholders. Objective methods of measuring usage have been suggested to be an effective way of monitoring orthosis usage, with temperature sensors most commonly used in this review and more widely in the literature[15, 17, 19 20], though another paper has also reported success with the use of pressure sensors[19]. While there were reports of problems with data collection using temperature sensors in this review [15, 17], the continuous data collection not only produces more accurate measurements of wear time, but has the added advantage being able to use the data for a more detailed investigation of AFO usage. As

suggested in the review[15], these data could be analysed further to identify patterns of use which may provide superficial insight into factors which affect the user's behaviour, though discussion with the individual would be needed to fully understand the reasons behind the behaviour.

Factors which influence usage

All of the papers in the review, even those which did not explicitly investigate factors which influence AFO usage, included at least anecdotal elements which were suggested to contribute towards AFO wear. The most common factors raised in the review were: perception of the device, particularly regarding impact on user function; and cosmesis.

The thoughts about the device discovered in this review were similar to those seen in wider literature about AFO and similar orthotic use in other populations. Perception of the device supporting and aiding user function was a contributing factor to usage in the review[16] with participants reporting improvements in functionality when wearing their AFO. Pain and discomfort was viewed negatively [16] and associated with reduced wear [18], particularly where the device impaired activities, seen in this review as sitting on the floor or playing sport[18], and similar factors have been seen in papers which more widely examine orthotic adherence [10, 15, 18, 21-24].

As an externally worn medical device which is visible to peers, AFOs represent a medical intervention intertwined with sociological and psychological factors[25]. With the additional difficulties of developing identity and changes in social environments due to school changes etc., childhood and adolescence represent a significant age range that may be influenced by external pressures into being reluctant to wear AFOs[11], and this may complicate a patient's choice to engage with clinical recommendation. Cosmesis of device and its associations with peer perception [16, 18], and varying usage between week days

and weekends were seen in this review[15], suggesting that social environments may be a factor that can AFO usage. Interestingly, despite most participants commenting about the look of their AFOs and making comments about how they were perceived by their peers, children with tuned AFOs all reported using them regularly[16]. Although comments seen in another paper within this review ("Anna: I do not want to use it. Sister: I am sure it is because no others use orthosis. Anna: nodding)"[18]) and wider literature ("[…] now we have a 13-year-old who just wants to be like other 13-year-old girls, and she doesn't want to wear AFOs […]"[26]) suggest that cosmesis and peer perception may be more important to some children and have a greater impact on usage. This can apply to both the AFO and the footwear, and comments were seen in this review which specifically discussed the cosmesis of the footwear, independent of the AFO[16]. Further research would be needed to investigate the interactions in more detail, with recognition that this is likely to vary according to individual users.

There is an growing recognition of individuals' agency over adherence with medical direction and wearing orthoses [10]. In this review there was anecdotal evidence to suggest that even children dictate and adhere to their own rules of AFO usage around the guidance of their parents and care team[18], and this highlights the importance of working on an individual level to identify the factors which affect each child before engaging with them to improve adherence. There is already evidence to support the suggestion that if an AFO is adapted to meet the needs of the child, it can improve the adherence[27], and this was similarly seen in this review through AFO tuning[16]. By ensuring that AFOs are tailored to their user and acknowledging the agency that people with CP have over their healthcare and including them in a patient-led treatment pathways, it could be possible to ensure that they receive the support that that is most appropriate for them, physically and socially, and meaningfully contribute to their quality of life. This is particularly important for children and adolescents who are at a period of development

where they establish life-long behaviours. Encouraging AFO wear during these periods may not only ensure that the AFO is best able to support them and promote their independence in the short term but establishes a foundation for use in later life.

This review has identified a clear lack of research in this area and recommends that further investigation investigate AFO adherence in greater detail, considering methodology carefully to investigate adherence on an individual level and determine not only the amount of time that the AFO is worn for but the reasons why and interactions between the two with a view to supporting clinicians and people with CP who use AFOs in the future.

Conclusions

This review investigated adherence through two components: usage and factors which influence usage, and while most of the papers in the review touched on both elements, none explicitly looked at both or discussed the interactions between the two. Adherence is a complex subject, and both components are needed to gain a true understanding of how an AFO is perceived and engaged with by children with CP and how these perceptions affect its use. A mixed methodology supports a detailed investigation of adherence, with measurement of usage favouring quantitative assessment, and factors which influence that usage being indicatively investigated using qualitative methods. By investigating using multiple approaches, it is possible to gain a more holistic view of AFO use and better understand the motivators and barriers that affect adherence. This knowledge can better support the prescription of AFOs in the future.

Limitations

The paucity of literature in this area and the low participation numbers within the papers included in this review limit the extensibility of the findings. It is also possible that this review may have missed some papers which included discussion about usage of AFOs by children with CP or factors which influence that usage as part of a broader investigation of a tangential subject. While we included one such paper in this review[18] which we found through in the references of our literature reading, it is possible that more were overlooked by the search terms. However, this review has successfully drawn attention to the paucity of research in this subject area, commented on appropriate methodologies for the investigation, and provided some suggestions for future research.

Word count: 3,194 words

Figure one: Paper selection process

5. References

1. Wingstrand M, Hägglund G and Rodby-Bousquet E. Ankle-foot orthoses in children with cerebral palsy: a cross sectional population based study of 2200 children. *BMC Musculoskeletal Disorders* 2014; 15: 327. DOI: 10.1186/1471-2474-15-327.
2. Osam JA, Opoku MP, Dogbe JA, et al. The use of assistive technologies among children with disabilities: the perception of parents of children with disabilities in Ghana. *Disability and Rehabilitation: Assistive Technology* 2019: 1-8.
3. Nooijen C, Slaman J, van der Slot W, et al. Health-related physical fitness of ambulatory adolescents and young adults with spastic cerebral palsy. *Journal of rehabilitation medicine* 2014; 46: 642-647.
4. Swinnen E and Kerckhofs E. Compliance of patients wearing an orthotic device or orthopedic shoes: a systematic review. . *Journal of bodywork and movement therapies* 2015; 19: 759-770.
5. Muscari ME. Rebels with a cause: when adolescents won't follow medical advice. *AJN The American Journal of Nursing* 1998; 98: 26-30.
6. Nevins TE. Non-compliance and its management in teenagers. *Pediatric transplantation* 2002; 6: 475-479.
7. Petrini P and Seuser A. Haemophilia care in adolescents–compliance and

lifestyle issues. *Haemophilia* 2009; 15: 15-19.
8. Windebank KP and Spinetta JJ. Do as I say or die: Compliance in adolescents with cancer. *Pediatric Blood & Cancer* 2008; 50: 1099-1100. DOI: 10.1002/pbc.21460.
9. Al-Jewair TS, Suri S and Tompson BD. Predictors of adolescent compliance with oral hygiene instructions during two-arch multibracket fixed orthodontic treatment. *The Angle Orthodontist* 2011; 81: 525-531. DOI: 10.2319/092010-547.1.
10. Holtkamp FC, Wouters EJM, Van Hoof J, et al. Use of and satisfaction with ankle foot orthoses. . *Clinical Research on Foot & Ankle* 2015.
11. Hayles E, Harvey D, Plummer D, et al. Parents' experiences of health care for their children with cerebral palsy. *Qualitative health research* 2015; 25: 1139-1154.
12. Andersson C and Mattsson E. Adults with cerebral palsy: a survey describing problems, needs, and resources, with special emphasis on locomotion. *Developmental Medicine & Child Neurology* 2007; 43: 76-82. DOI: 10.1111/j.1469-8749.2001.tb00719.x.
13. Krakovsky G, Huth MM, Lin L, et al. Functional changes in children, adolescents, and young adults with cerebral palsy. *Research in Developmental Disabilities* 2007; 28: 331-340.
14. Basford J and Johnson S. Form may be as important as function in orthotic acceptance: a case report. *Archives of physical medicine and rehabilitation* 2002; 83: 433-435.
15. Schwarze M, Horoba L, Block J, et al. Wearing Time of Ankle-Foot Orthoses with Modular Shank Supply in Cerebral Palsy: A Descriptive Analysis in a Clinically Prospective Approach. *Rehabilitation Research and Practice* 2019; 2019: 1-9. DOI: 10.1155/2019/2978265.
16. Eddison N, Healy A and Chockalingam N. Does user perception affect adherence when wearing biomechanically optimised ankle foot orthosis–footwear combinations: A pilot study. *The Foot* 2020; 43: 101655.
17. Maas JC, Dallmeijer AJ, Oudshoorn BY, et al. Measuring wearing time of knee-ankle-foot orthoses in children with cerebral palsy: comparison of parent-report and objective measurement. *Disability and Rehabilitation* 2018; 40: 398-403. DOI: 10.1080/09638288.2016.1258434.
18. Øien I, Fallang B and Østensjø S. Everyday use of assistive technology devices in school settings. *Disability and Rehabilitation: Assistive Technology* 2016; 11: 630-635.
19. Hunter LN, Sison-Williamson M, Mendoza MM, et al. The validity of compliance monitors to assess wearing time of thoracic-lumbar-sacral orthoses in children with spinal cord injury. *Spine* 2008; 33: 1554-1561.
20. Rahman T, Borkhuu B, Littleton AG, et al. Electronic monitoring of scoliosis brace wear compliance. *Journal of Children's Orthopaedics* 2010; 4: 343-347. DOI: 10.1007/s11832-010-0266-6.

21. Garralda ME, Muntoni F, Cunniff A, et al. Knee–ankle–foot orthosis in children with duchenne muscular dystrophy: user views and adjustment. *European Journal of Paediatric Neurology* 2006; 10: 186-191.
22. Sugawara AT, Ramos VD, Alfieri FM, et al. Abandonment of assistive products: assessing abandonment levels and factors that impact on it. . *Disability and Rehabilitation: Assistive Technology* 2018; 13: 716-723.
23. Swinnen E, Deliens T, Dewulf E, et al. What is the opinion of patients with multiple sclerosis and their healthcare professionals about lower limb orthoses? A qualitative study using focus group discussions. *NeuroRehabilitation* 2018; 42: 81-92.
24. Ireno JM, Chen N, Zafani MD, et al. The use of orthoses in children with cerebral palsy: perception of caregivers. *Cadernos Brasileiros de Terapia Ocupacional* 2019; 27: 35-44.
25. Luger R, Geiger M and Lyner-Cleophas M. Students' voices: reflections of three young adults with cerebral palsy on factors facilitating their completion of mainstream schooling in South Africa. *International Journal of Inclusive Education* 2019: 1-17.
26. Hayles E, Harvey D, Plummer D, et al. Parents' experiences of health care for their children with cerebral palsy. *Qualitative health research* 2015; 25: 1139-1154.
27. Van Der Wilk D, Hijmans JM, Postema K, et al. A user-centered qualitative study on experiences with ankle-foot orthoses and suggestions for improved design. . *Prosthetics and orthotics international* 2018; 42: 121-128.

Appendix A – Articles included in this scoping review

Papers identified through literature search

Eddison, N., Healy, A., & Chockalingam, N. (2020). Does user perception affect adherence when wearing biomechanically optimised ankle foot orthosis–footwear combinations: A pilot study. The Foot, 43, 101655.

Maas, J. C., et al. (2018). "Measuring wearing time of knee-ankle-foot orthoses in children with cerebral palsy: comparison of parent-report and objective measurement." Disability and rehabilitation 40(4): 398-403.

Øien, I., Fallang, B., & Østensjø, S. (2016). Everyday use of assistive technology devices in school settings. Disability and Rehabilitation: Assistive Technology, 11(8), 630-635.

Schwarze, M., Horoba, L., Block, J., Putz, C., Alimusaj, M., Wolf, S. I., & Dreher, T. (2019). Wearing Time of Ankle-Foot Orthoses with Modular Shank Supply in Cerebral Palsy: A Descriptive Analysis in a Clinically Prospective Approach. Rehabilitation research and practice, 2019.