

Article

Functional Impact of Early Prosthetic Implantation in Children with Upper Limb Agenesis or Amputation

Nicola Manocchio ¹, Marco Gaudenzi ¹, Marco Tofani ^{2,3}, Concetta Ljoka ¹, Anila Imeshtari ¹,
Laura Giordani ¹, Gessica Della Bella ⁴ and Calogero Foti ^{1,*}

¹ Physical and Rehabilitation Medicine, Department of Clinical Sciences and Translational Medicine, University of Tor Vergata, 00133 Rome, Italy; nicola.manocchio@uniroma2.it (N.M.); gaudenzi.ma@gmail.com (M.G.); cljoka@gmail.com (C.L.); anila.imeshtari@gmail.com (A.I.); laura.giordani@uniroma2.it (L.G.)

² Department of Life Sciences, Health and Allied Healthcare Professions Università degli Studi “Link Campus University”, Via del Casale di San Pio V, 44, 00165 Rome, Italy; marco.tofani@opbg.net

³ Management and Diagnostic Innovations & Clinical Pathways Research Area, Professional Development, Continuous Education and Research Service, Bambino Gesù Children’s Hospital, IRCCS, 00165 Rome, Italy

⁴ Neurorehabilitation and Adapted Physical Activity Day Hospital, Bambino Gesù Children’s Hospital, IRCCS, 00165 Rome, Italy; gessica.dellabella@opbg.net

* Correspondence: foti@med.uniroma2.it; Tel.: +39-0620900594

Abstract: This study investigated the impact of early prosthetic intervention on children with upper limb agenesis or amputation. The aim was to assess both how early prosthetics improve a child’s movement and coordination skills, as well as how satisfied both the child and their parents are with the prosthesis. Twenty-nine children were evaluated using the Unilateral Below Elbow Test (UBET) and Child Amputee Prosthetics Project—Prosthesis Satisfaction Inventory (CAPP-PSI). Results suggest that while children performed tasks faster and more fluidly without a prosthesis, they were satisfied with the prostheses and the service provided. This may be due to ongoing adaptation to the new device. The single child re-evaluated at 12 months showed improvement in prosthesis use, highlighting the potential benefits of early intervention coupled with motor re-education. Further research is needed to optimize prosthetic features and address initial challenges associated with prosthesis use.

Keywords: upper limb agenesis; amputation; children; prosthesis; motor function; quality of life



Citation: Manocchio, N.; Gaudenzi, M.; Tofani, M.; Ljoka, C.; Imeshtari, A.; Giordani, L.; Della Bella, G.; Foti, C. Functional Impact of Early Prosthetic Implantation in Children with Upper Limb Agenesis or Amputation. *Appl. Sci.* **2024**, *14*, 7259. <https://doi.org/10.3390/app14167259>

Academic Editor: Arkady Voloshin

Received: 27 June 2024

Revised: 14 August 2024

Accepted: 15 August 2024

Published: 18 August 2024



Copyright: © 2024 by the authors. Licensee MDPI, Basel, Switzerland. This article is an open access article distributed under the terms and conditions of the Creative Commons Attribution (CC BY) license (<https://creativecommons.org/licenses/by/4.0/>).

1. Introduction

In today’s clinical and rehabilitation landscape, the management of children with agenesis or amputation of the upper limb has become an area that increasingly requires the definition of shared protocols for treatment and care. This pediatric population, characterized by unique needs at both motor and psychosocial levels, requires special attention in order to ensure an optimal quality of life (QoL) and healthy development. One of the key interventions in this context is prothesization, a solution that, if introduced in a timely manner, promises to redefine the developmental trajectory of these young patients [1]. However, despite expectations and clinical evidence, the scientific literature on the evaluation of the outcomes of such interventions still remains largely limited.

The fundamental premise of this work stems from the need to fill this information gap. Considering that the upper limb plays a key role in numerous daily activities, such as interacting with objects, playing, and learning, it is essential to determine to what extent early prothesization can improve these abilities or, possibly, whether there are inherent limitations to the device that require further therapeutic intervention [2].

However, it should be noted that although prothesization is a fundamental step in the Individual Rehabilitation Project (IRP), it does not operate in isolation. The integration

of prostheses goes hand in hand with a set of interventions, including physical therapies, motor re-education, and psychological support, which together contribute to the success of the therapeutic approach. Therefore, it is essential to consider prosthesization as part of a broader system of care delivered by a rehabilitation team that works together to achieve the ultimate goal of rehabilitation and the improvement of the child's autonomy and QoL [3].

Amputations, or congenital malformations of the hand and upper limb, represent an eventuality that is not particularly frequent in clinical practice, and the literature is relatively poor in epidemiological studies on the subject [4,5].

Agenesis of the upper limb involves the congenital partial or total absence of either upper limb and can occur in various forms, ranging from complete absence of the limb to absence of parts of it. Most limb malformation abnormalities occur during the embryonic stage of differentiation. The malformation is understood as an abnormality during formation and/or differentiation, may be associated with a specific part of the developing limb, and may result in an abnormality affecting the entire limb or only part of it [6–8]. Not to be overlooked is the fact that these pathologies of the upper limb are not infrequently associated with other independent malformations or resulting from genetic syndromes such as Holt–Oram, radial aplasia with thrombocytopenia, De Lange, and others [9–14]. These defects may go unrecognized or unrecorded. In addition, minor abnormalities may never be detected or recorded. The incidence of agenesis of the upper limb, therefore, varies depending on the populations and study methodologies adopted, as well as the geographical area studied, but is generally estimated to affect 1–2 per 4000 live births [1,15].

With regard to upper limb amputation in children, on the other hand, the causes are mainly trauma, tumors (such as Ewing sarcoma and osteosarcoma), infections, or congenital malformations [16–21].

Children with agenesis or amputation of the upper limb are confronted with four main challenges: (1) difficulties in the development of normal motor skills, (2) the need for assistance in daily activities, (3) limitations in certain activities of daily living (ADLs) and in sport, and (4) potential emotional and social problems due to physical appearance [22].

The Neuronal Group Selection Theory (NGST) comes into play to support the thesis that early prosthesization can be beneficial for the child. The NGST emphasizes the hypothesis that the brain is organized in neuronal networks that are influenced by the child's development and behavior and that, in particular, motor development appears to be characterized by two phases of variation: primary variability, in which motor activity does not depend on the environment, and secondary variability when the child learns to select the most efficient motor strategies from a repertoire of variable movements, based on active practice [23]. This current of thought suggests that the first prosthesis should be prescribed as early as possible, giving the child the opportunity to develop an expanded motor repertoire with and without a prosthesis [1,2,24].

In this descriptive observational study, we aim to examine the impact of early prosthetic intervention on motor function and QoL on a population of children with upper limb agenesis or amputation. The aim is twofold: on the one hand, to assess possible benefits in terms of motor strategies and dexterity, and on the other hand, to evaluate the influence of such interventions on child and parent satisfaction with the prosthesis.

2. Materials and Methods

The protocol was carried out at the Upper Limb Day Hospital (DH), Department of Neurorehabilitation of the Bambino Gesù Children's Hospital, Rome, Italy, EU.

The clinical protocol was conducted, recorded, and reported in accordance with Good Clinical Practice guidelines and the Declaration of Helsinki. As the children were underage, all participants' caregivers were asked to sign an informed consent form before taking part in the study; all children were, however, asked to agree to participate in the study [25]. Ethical review and approval were waived for this study due to the explorative nature of the study and the non-intervention.

All children with agenesis or upper limb amputation, referred to the DH, were evaluated for this study by a specialist in Physical and Rehabilitation Medicine (PRM) together with an orthopedic technician. In case the child did not already have a prosthesis, the prosthesis was chosen from two options: aesthetic passive prosthesis and myoelectric prosthesis [26,27].

Following prosthesis delivery, the first clinical evaluation by PRM, a hand surgeon, together with both the physiotherapist (PT) and occupational therapist (OT), was conducted. All children already had an ongoing IRP at other centers, mainly in the form of motor re-education. These interventions were continued following the recommendations of the referring physicians, with the addition of a prosthesis-based re-education program established at the DH. Children were then re-evaluated after six months of motor re-education with and without the prosthesis. Only one child received a second follow-up evaluation at 12 months.

During the multidisciplinary examinations conducted by PRM, a hand surgeon, and a PT or OT, the children underwent the following assessments:

UBET (The Unilateral Below Elbow Test) [28]. The UBET was developed to assess the function of bimanual tasks for both prosthesis wearers and non-wearers. UBET consists of nine tasks for each of the four age-specific categories defined by the developmental stages of hand function (2–4 years, 5–7 years, 8–10 years, and 11–21 years) (Table 1).

Table 1. UBET scale-tasks.

| UBET-Tasks | | | | |
|------------|----------------------------------------------|------------------------------------------|-----------------------------------------------|-------------------------------------------|
| | 2–4 Years | 5–7 Years | 8–10 Years | 11–21 Years |
| Task 1 | Take Play-Doh out of a plastic bag | Cut paper circle from construction paper | Swing a bat | Cut paper from a roll to wrap a videotape |
| Task 2 | Bang cymbals together | Remove cap from felt tip marker | Wind string onto yo-yo | Tear a piece of tape |
| Task 3 | Put sock on foot | Sharpen pencil | Open a Band-Aid | Secure wrap on videotape with tape |
| Task 4 | Thread beads | Do up buttons on vest | Tie shoelaces in a bow | Cut putty on plate with knife and fork |
| Task 5 | Open a jar of bubbles | Tie shoelaces in a knot | Do up buttons on a shirt | Open a three-ring binder |
| Task 6 | Ride on a Rolling Racer | Turn kaleidoscope | Make a telescope with paper and a rubber band | Start zipper on vest |
| Task 7 | Open drawstring bag and dump LEGO DUPLOs out | Separate LEGOs | Place glove on unaffected hand | Tie shoelaces in a bow |
| Task 8 | Separate LEGO DUPLOS | Use bow and arrow | Draw a line with a ruler | Do up buttons on a shirt |
| Task 9 | Open a box of crayons and remove one | Ride on Rolling Racer | Start zipper on vest | Use dust pan and small broom |

UBET is divided into two subscales: Task completion and Method of use.

Task completion assesses the level of function on a 5-point interval scale designed to distinguish the ease of completing the task and the quality of movement displayed by a child. Task completion is the main score to be used to assess function, as the essential assessment is whether the child is able to perform a specific task. The scale for Task completion ranges from 0 to 4, with 4 representing completion of the task without difficulty and 0 representing inability to complete the task.

The Method of use is the manner in which the child uses the prosthesis to achieve the goal. It is calculated on a 4-point scale that is denoted by categorical variables described as follows:

| Code | With the Prosthesis | Without the Prosthesis |
|------|-----------------------------------------------------------------------------|-----------------------------------------------------|
| A | Active grip through the end of the device | Manipulation and/or stabilization via the stump end |
| P | Passive use of the prosthesis through the forearm or end part of the device | Forearm stabilization |
| E | Grip through the elbow or trunk | Grip through the elbow or trunk |
| N | No use of the affected limb | No use of the affected limb |

This score can be used to compare performance between the wearer and non-wearer of the prosthesis, performance by the same subject with and without a prosthesis, and finally, to assess performance with two different types of prosthesis. To promote spontaneity, the assessment is video-recorded, and the scores for Task completion and Method of use are assessed in a subsequent viewing.

Children were also divided into two populations according to age group (preschool and school), and a comparison of the total values obtained from the UBET scales administered in the first assessment was carried out.

CAPP-PSI (Child Amputee Prothetics Project—Prosthesis Satisfaction Inventory) [29]. The CAPP-PSI is a standardized measure of prosthetic satisfaction in children with limb deficits. It is designed to assess parents' satisfaction with the prosthetic device prescribed for their child with regard to fit, function, appearance, and service. Parents rate 14 questions divided into 3 scales as follows:

1. Child satisfaction assessed by the parents with the prosthesis (e.g., "Does your child like the way his prosthesis fits?");
2. Parental satisfaction with the prosthesis (e.g., "Are you satisfied with the way your child's prosthesis helps him/her perform activities of daily living?");
3. Parental satisfaction with the service (e.g., "Are you satisfied with the time it took to repair your child's prosthesis?").

Parents select an answer for each question using the following categories with scores from 0 to 4: 0 = 'not at all'; 1 = 'slightly'; 2 = 'quite a lot'; 3 = 'very much'; 4 = 'extremely'.

All data were initially entered into an Excel spreadsheet (Microsoft, Redmond, WA, USA). Descriptive statistics are shown as mean \pm standard deviation (SD) or as a percentage.

3. Results

Twenty-nine children (19 boys, 66%, age 1–12 years) with unilateral upper limb agenesis or amputation were enrolled for the purpose of this study. Among them, 23 (79%) showed a forearm/wrist amputation and 6 (21%) a carpus/finger amputation.

At the first assessment, seven (24%) children already had a prosthesis (five had an aesthetic prosthesis, and two had a myoelectric prosthesis), while the other 22 (76%) had never worn an upper limb prosthesis. Five of the twenty-two (23%) children without a prosthesis were offered and given an aesthetic prosthesis; one of these children was then switched from an aesthetic prosthesis to a myoelectric prosthesis. Thus, a total of twelve children were fitted with prostheses; nine (75%) received aesthetic prostheses, while three (25%) were myoelectric devices.

All the young patients carried on the entire protocol. The entire population agreed to be re-evaluated without a prosthesis, and the 12 children with prostheses were additionally evaluated while wearing the device.

The UBET scale was administered to all children by dividing them into age groups (2–4 years, 5–7 years, 8–10 years, and 11–21 years) and taking into account its two components: Task completion and Method of use. Tables 2 and 3 show the efficiency of completion of each UBET scale task, respectively, without and with a prosthesis on.

Table 2. UBET Task completion scale results, without prosthesis evaluation.

| UBET-Task Completion: Evaluation without Prosthesis | | | |
|-----------------------------------------------------|----|-------|----------------|
| | N | Mean | Std. Deviation |
| Task 1 | 29 | 3.10 | 0.900 |
| Task 2 | 29 | 3.03 | 1.476 |
| Task 3 | 29 | 2.52 | 1.353 |
| Task 4 | 29 | 2.28 | 1.386 |
| Task 5 | 29 | 2.62 | 1.522 |
| Task 6 | 29 | 2.62 | 1.347 |
| Task 7 | 29 | 3.10 | 1.047 |
| Task 8 | 29 | 2.72 | 1.386 |
| Task 9 | 29 | 3.00 | 1.069 |
| Overall | 29 | 25.00 | 7.096 |

Table 3. UBET Task completion scale results, with prosthesis evaluation.

| UBET-Task Completion: Evaluation with Prosthesis | | | |
|--------------------------------------------------|----|-------|----------------|
| | N | Mean | Std. Deviation |
| Task 1 | 12 | 2.50 | 1.243 |
| Task 2 | 12 | 2.08 | 1.676 |
| Task 3 | 12 | 2.33 | 1.371 |
| Task 4 | 12 | 2.33 | 1.497 |
| Task 5 | 12 | 2.25 | 1.288 |
| Task 6 | 12 | 2.67 | 1.155 |
| Task 7 | 12 | 2.92 | 1.165 |
| Task 8 | 12 | 3.17 | 1.115 |
| Task 9 | 12 | 3.42 | 0.515 |
| Overall | 12 | 23.67 | 6.050 |

Most of the children performed and completed the various tasks with little or no difficulty, whether the tasks were performed with or without a prosthesis; none of the participants failed in completing the tasks. A difference between the two conditions emerged when comparing the overall sum of mean results (25 ± 7.096 without prosthesis— 23.67 ± 6.050 with prosthesis).

UBET Method scale results are reported in Table 4 (without prosthesis) and Table 5 (with prosthesis) as percentages of used parts.

Table 4. UBET Method of use scale results without prosthesis evaluation. A: Manipulation and/or stabilization via the stump end; P: Forearm stabilization; E: Grip through the elbow or trunk; N: No use of the affected limb.

| UBET-Method of Use: Evaluation without Prosthesis | | | | | | | | | |
|---------------------------------------------------|--------|--------|--------|--------|--------|--------|--------|--------|--------|
| | Task 1 | Task 2 | Task 3 | Task 4 | Task 5 | Task 6 | Task 7 | Task 8 | Task 9 |
| A | 41.4% | 37.9% | 41.4% | 44.8% | 37.9% | 51.7% | 41.4% | 41.4% | 41.4% |
| P | 37.9% | 27.6% | 27.6% | 27.6% | 27.6% | 27.6% | 34.5% | 34.5% | 41.4% |
| E | 13.8% | 24.1% | 20.7% | 20.7% | 27.6% | 20.7% | 20.7% | 20.7% | 10.3% |
| N | 6.9% | 10.3% | 10.3% | 6.9% | 6.9% | 0% | 3.4% | 3.4% | 6.9% |

Table 5. UBET Method of use scale results with prosthesis evaluation A: Active grip through the end of the device; P: Passive use of the prosthesis through the forearm or end part of the device; E: Grip through the elbow or trunk; N: No use of the affected limb.

| UBET-Method of Use: Evaluation with Prosthesis | | | | | | | | | |
|------------------------------------------------|--------|--------|--------|--------|--------|--------|--------|--------|--------|
| | Task 1 | Task 2 | Task 3 | Task 4 | Task 5 | Task 6 | Task 7 | Task 8 | Task 9 |
| A | 33.3% | 0.3% | 41.7% | 24.9% | 24.9% | 50% | 41.5% | 41.5% | 41.5% |
| P | 16.7% | 24.9% | 16.7% | 33.3% | 24.9% | 33.3% | 16.7% | 41.5% | 41.5% |
| E | 50% | 41.5% | 24.9% | 41.5% | 16.9% | 16.7% | 41.5% | 16.7% | 16.7% |
| N | 0% | 33.3% | 16.7% | 0.3% | 33.3% | 0% | 0.3% | 0.3% | 0.3% |

A trend of greater utilization of the distal portions of the stump in tests without the prosthesis can be appreciated; conversely, with the prosthesis on, the trend is toward greater utilization of the more proximal portions of the prosthetic limb. However, this trend is not confirmed in tasks 3-6-7-8-9, which involves the distal part of the limb (e.g., putting on a sock, riding a scooter by grasping the steering with both hands, detaching two Lego bricks, opening a zip of a case with eight crayons and extracting one of them). Notably, small percentages are scored at N value overall.

UBET scale results were also compared for age groups (Table 6) and amputation levels (Table 7).

Table 6. UBET comparison for age groups.

| | Age | N | Mean | Std. Deviation |
|--------------------------|-----------|----|-------|----------------|
| Total without prosthesis | Preschool | 13 | 22.31 | 6.486 |
| | School | 16 | 27.19 | 6.997 |
| Total with prosthesis | Preschool | 6 | 21.00 | 4.290 |
| | School | 6 | 26.33 | 6.713 |

Table 7. UBET comparison for amputation level.

| | Amputation Level | N | Mean | Std. Deviation |
|--------------------------|------------------|----|-------|----------------|
| Total without prosthesis | Forearm/Wrist | 23 | 25.00 | 7.000 |
| | Carpus/Fingers | 6 | 25.00 | 8.149 |
| Total with prosthesis | Forearm/Wrist | 9 | 23.89 | 6.698 |
| | Carpus/Fingers | 3 | 23.00 | 4.583 |

These data show a difference in motor skills without the use of the prosthesis in the school-age population compared to the preschool population, even considering age-adjusted performance as already established by the UBET.

Table 8 (overall results) and Table 9 (Difference according to the age of the child) report data from the CAPP-PSI scale for prosthesis satisfaction.

Table 8. CAPP-PSI: overall results.

| CAPP-PSI-Prosthesis Use Satisfaction Score | | | |
|--------------------------------------------|----|-------|----------------|
| | N | Mean | Std. Deviation |
| Child satisfaction (Parental judgment) | 12 | 10.42 | 4033 |
| Parental satisfaction with the prosthesis | 12 | 10.50 | 3680 |
| Parental satisfaction with the service | 12 | 17.67 | 6959 |

Table 9. CAPP-PSI: difference between preschooler and school age.

| Difference in Average Prosthesis Satisfaction Scores according to the Age of the Child | | | | |
|----------------------------------------------------------------------------------------|-----------|---|-------|----------------|
| | Age | N | Mean | Std. Deviation |
| Child satisfaction (Parental judgment) | Preschool | 6 | 10.17 | 4.535 |
| | School | 6 | 10.67 | 3.882 |
| Parental satisfaction with the prosthesis | Preschool | 6 | 9.83 | 4.167 |
| | School | 6 | 11.17 | 3.371 |
| Parental satisfaction with the service | Preschool | 6 | 18.50 | 3.834 |
| | School | 6 | 16.83 | 9.496 |

Mean results show that families and children were quite satisfied with the prescribed prosthesis and very satisfied with the service (delivery time of the prosthesis, instructions/indications given, manufacturing/repair time, rehabilitation/training activities with respect to the use of the prosthesis). Results were also comparable when preschool and school differences were analyzed.

Data from Follow-Up at 12 Months of Single Child

Table 10 (without prosthesis) and Table 11 (with prosthesis) report data on the UBET Method of use scale of the single child who received a second assessment at a 12-month time point.

Table 10. UBET Method of use scale results without prosthesis evaluation of a single child A: Manipulation and/or stabilization via the abutment end; P: Forearm stabilization; E: Grip through the elbow or trunk; N: No use of the affected limb.

| Evaluation without Prosthesis—UBET Method of Use | | | | | | | | | |
|--------------------------------------------------|--------|--------|--------|--------|--------|--------|--------|--------|--------|
| Time | Task 1 | Task 2 | Task 3 | Task 4 | Task 5 | Task 6 | Task 7 | Task 8 | Task 9 |
| 6 months | A | E | E | A | A | A | E | A | A |
| 12 months | A | E | E | A | A | A | E | A | A |

Table 11. UBET Method of use scale results with prosthesis evaluation of a single child. A: Active grip through the end of the device; P: Passive use of the prosthesis through the forearm or end part of the device; E: Grip through the elbow or trunk; N: No use of the affected limb.

| Evaluation with Prosthesis—UBET Method of Use | | | | | | | | | |
|-----------------------------------------------|--------|--------|--------|--------|--------|--------|--------|--------|--------|
| Time | Task 1 | Task 2 | Task 3 | Task 4 | Task 5 | Task 6 | Task 7 | Task 8 | Task 9 |
| 6 months | E | E | E | P | P | P | E | P | P |
| 12 months | A | E | E | A | A | A | E | A | A |

Results completely overlap for the without prosthesis evaluation. On the other hand, an improvement in the performance of tasks with the aid of the prosthesis is evident in almost all tasks.

Table 12 (with prosthesis) and Table 13 (without prosthesis) report data of the UBET Task Completion scale.

Table 12. UBET Task completion scale results with prosthesis evaluation of a single child.

| Evaluation with Prosthesis—UBET Task Completion | | | | | | | | | |
|-------------------------------------------------|--------|--------|--------|--------|--------|--------|--------|--------|--------|
| Time | Task 1 | Task 2 | Task 3 | Task 4 | Task 5 | Task 6 | Task 7 | Task 8 | Task 9 |
| 6 months | 1 | 1 | 1 | 3 | 1 | 1 | 3 | 3 | 4 |
| 9 months | 3 | 4 | 3 | 0 | 0 | 3 | 4 | 3 | 4 |

Table 13. UBET Task completion scale results without prosthesis evaluation of a single child.

| Evaluation without Prosthesis—UBET Task Completion | | | | | | | | | |
|----------------------------------------------------|--------|--------|--------|--------|--------|--------|--------|--------|--------|
| Time | Task 1 | Task 2 | Task 3 | Task 4 | Task 5 | Task 6 | Task 7 | Task 8 | Task 9 |
| 6 months | 3 | 4 | 3 | 0 | 0 | 3 | 4 | 3 | 4 |
| 12 months | 3 | 4 | 3 | 0 | 0 | 3 | 4 | 3 | 4 |

The higher scores in Table 12 at the 9-month time point show a progressive adaptation to the prosthesis, resulting in a reduction in the difficulty of performing tasks with the aid. Again, as before, the results without a prosthesis are overlapping.

4. Discussion

This descriptive observational study investigated the efficacy and impact of early prosthetic intervention on motor function and quality of life (QoL) in children with upper limb agenesis or amputation.

Our findings based on UBET scores suggest that upper limb movements appeared faster and more fluid without the prosthesis compared to movements with the prosthesis, which were perceived as slower and more awkward. This may indicate a stronger ingrained motor pattern with the stump than with the prosthesis. Given that this was a first-time introduction to the prosthesis, adaptations to the new motor schema were likely still ongoing. Disruption of the child's established "normal" movement pattern could explain the observed slower and less fluid movements with the prosthesis. As a result, the child might initially perceive the prosthesis as an impediment rather than a facilitating tool. Addressing movement disruptions, improving prosthetic features, and considering the impact of motor impairments are crucial factors in enhancing the effectiveness and acceptance of prosthetic devices in children. Literature reports suggest that interventions aimed at improving movement skills in children could involve educating parents on recognizing good and poor skill performance in their children [30]. Further support for a link between movement disruptions and prosthesis use can be found in research on skilled tool use

and prosthetic hand function. These studies have shown disruptions to typical eye-hand coordination [31]. To address these challenges and improve user experience, researchers have emphasized the importance of advancements in prosthetic features. Key areas of focus include improved gloving materials, extended battery life, enhanced hand reliability, and more natural finger movement [32].

A second peculiar finding stems from the UBET Method of use scale. During the evaluation, children exhibited a more distal execution mode (greater use of the terminal stump) for various activities without the prosthesis. However, this pattern changed with prosthesis use. Excluding the N parameter (complete non-use), children displayed a more proximal utilization of the prosthesis, likely attempting to manipulate it as if it were his/her own stump. This dynamic tends to generate hostile reactions, helping to establish a negative perception in the child that may detract from his involvement in the use of the prosthesis. Interestingly, the percentage of distal usage for both the stump and prosthesis increased during tasks requiring peculiar actions (items 3, 6, 7, 8, and 9). Examples include tasks like putting on socks, grasping scooter handlebars with both hands, detaching Lego bricks, opening a crayon case, and extracting a crayon. This suggests that children can overcome established compensatory strategies and adapt to utilize the prosthesis distally for specific tasks. This adaptation to using the prosthesis distally for specific tasks could be influenced by factors such as patient satisfaction, functional outcomes, and the perceived value of prosthetic use. Studies have shown that the age at the first prosthetic fitting can impact later functional outcomes in children with limb deficiencies, highlighting the importance of early intervention [33]. Furthermore, the effectiveness of prosthetic devices in children is influenced by factors such as the prescription of the first prosthesis, with studies indicating lower rejection rates in children fitted with their first prosthesis at a younger age [24]. Additionally, advancements in pediatric prostheses and considerations for future developments underscore the importance of designing prosthetic devices that facilitate the effective performance of daily tasks and promote healthy social interactions [34]. Another critical aspect concerns postural balance, both in static and dynamic conditions. The child, without a prosthesis, has autonomously developed its own balance strategy, resorting to different forms of adaptation and compensation that take into account the amputated/agenetic limb. The introduction of the prosthesis represents, in the initial stages, an element of disharmony that can generate feelings of discomfort. In particular, the weight of the artificial limb may negatively affect postural stability. However, it is important to emphasize that time, together with adequate training and adaptation, can play a crucial role in facilitating the acquisition of greater stability and optimal posture. Consequently, it can be inferred that the prescription of a prosthesis should not be intended as a passive act but should necessarily be integrated into a motor re-educational program, thus making the prosthetic intervention fully effective [35]. Our data also suggest a potential link between age and motor skills without a prosthesis, with school-aged children exhibiting higher performance compared to preschoolers. This difference may be attributed to the extended adaptation time afforded to school-aged children, allowing for the consolidation of compensatory strategies that enhance motor skills and functional performance during activities.

The only child who received a 12-month follow-up revealed a progressive optimization in the use of the prosthesis. The child went from a passive use of the prosthesis to an active mode of use of the prosthesis, which is expressed by an increased use of the end part of the aid. The performance of the same activities without the prosthesis did not show any variation. Over time, the adaptation to the use of the stump had probably already taken place and been consolidated. On the contrary, the child still had to adapt to the use of the prosthesis. Motor re-education aims to bridge this gap by promoting the active integration of the prosthesis and fostering the development of motor skills that enhance dexterity and fluidity for daily tasks. It is also possible that initially, wearing the prosthesis disrupted the postural balance learned by the child for dynamic and static compensations with the agenetic/amputated limb. In the subsequent evaluations, the adaptation to the

prosthesis may have led to an improvement of the postural balance, with a better use of the prosthesis at different times. This is in agreement with the literature data, which confirm the postural improvement that may result from the combination of an early prosthesis and a correct motor re-education program, prompting the child to understand at an early stage the possible benefits that may result from the use of the prosthesis, so as to motivate him to overcome the initial phases in which the aid will certainly be perceived as an agent disrupting the postural and motor scheme acquired by the child over time with his amputation [32,36].

Early prosthesis cannot be understood solely in relation to the time factor. The earliness of prosthetising a child is certainly a fundamental parameter of great importance. Citing NGST again, motor development seems to be characterized by two phases of variation: primary variability, when the motor activity does not depend on the environment, and secondary variability when the child learns to select the most efficient motor strategies from a repertoire of variable movements, based on active practice. According to this theory, experience-related information induces changes in synaptic connections within and between neuronal groups, giving rise to the variable secondary repertoire [23]. The altered connectivity within the secondary repertoire enables a situation-dependent selection of neuronal groups. The analysis of the data shows how the early adoption of a prosthetic implant, accompanied by an appropriate proprioceptive and motor re-educational program, can facilitate the process of development of the neural pathways necessary for the effective integration of the prosthesis by the child. This results in a more rapid acceptance of the artificial limb and in its perception as a functional tool for the realization of optimal motor patterns rather than as an obstacle, which is how the child tends to perceive it in the initial stages of approach to the aid. In this initial phase, in fact, it is natural for the child to consider the execution of the proposed tasks easier using the stump alone [2].

It is well known that the first contact with the prosthesis is experienced by the child with a certain strangeness and sometimes even annoyance or frustration. The latter stems from the awareness that control of the artificial limb is not immediate, which generates difficulties in performing the desired actions and reinforces the belief that the same actions would have been more easily achievable without the aid of the prosthesis [36]. The data we collected further confirms these considerations.

Limitations

This study has several limitations.

First of all, since this is a descriptive observational study, no statistical analysis could be carried out. Thus, no statistical significance could be extrapolated from our data.

Moreover, since the children were already undergoing an IRP, motor re-education was not standardized, which could be a confounding factor.

Lastly, no evaluation of the psychological well-being of children has been made.

Future research should try to solve these issues and cover the gap.

5. Conclusions

Our study provides preliminary evidence regarding the evaluation of early prosthetic use in children with agenesis or amputation of the upper limb.

Early prosthesization could be useful in ensuring a fast adaptation of the child to the aid, considering the disruption of movement in the early phase of prosthesis use.

A multidisciplinary approach involving medical specialists and PT is essential. This professional synergy is crucial in order to guarantee comprehensive support to both children and their families, identify the most appropriate prosthesis, propose appropriate motor treatments, and outline an optimal psychological pathway that favors adaptation and learning to use the prosthesis. Such an intervention accelerates and facilitates the development of motor strategies necessary to allow the child to accept and make the most of the prosthesis, resulting in an improved quality of life, increased autonomy in activities of daily living, and promotion of social integration with peers.

Author Contributions: Conceptualization, M.T., G.D.B. and C.F.; methodology, G.D.B. and N.M.; investigation, N.M., M.T., A.I. and M.G.; writing—original draft preparation, C.L., M.G. and L.G.; writing—review and editing, N.M., G.D.B. and C.F.; supervision, C.F. All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Institutional Review Board Statement: The study was conducted in accordance with the Declaration of Helsinki. Ethical review and approval were waived for this study due to the explorative nature of the study and the non-intervention.

Informed Consent Statement: Informed consent was obtained from all caregivers of the subjects involved in the study; subjects were asked to agree to take part in the study.

Data Availability Statement: Data are available upon request to the corresponding author.

Conflicts of Interest: The authors declare no conflicts of interest.

References

1. Shaperman, J.; Landsberger, S.E.; Setoguchi, Y. Early Upper Limb Prosthesis Fitting: When and What Do We Fit. *JPO J. Prosthet. Orthot.* **2003**, *15*, 11–17. [CrossRef]
2. Peterson, J.K.; Prigge, P. Early Upper-Limb Prosthetic Fitting and Brain Development: Considerations for Success. *J. Prosthet. Orthot.* **2020**, *32*, 229–235. [CrossRef]
3. Mano, H.; Fujiwara, S.; Haga, N. Effect of Prostheses on Children with Congenital Upper Limb Deficiencies. *Pediatr. Int.* **2020**, *62*, 1039–1043. [CrossRef]
4. Dillingham, T.R.; Pezzin, L.E.; Mackenzie, E.J. Limb Amputation and Limb Deficiency: Epidemiology and Recent Trends in the United States. *South. Med. J.* **2002**, *95*, 875–883. [CrossRef] [PubMed]
5. Bermejo-Sánchez, E.; Cuevas, L.; Amar, E.; Bakker, M.K.; Bianca, S.; Bianchi, F.; Canfield, M.A.; Castilla, E.E.; Clementi, M.; Cocchi, G.; et al. Amelia: A Multi-center Descriptive Epidemiologic Study in a Large Dataset from the International Clearinghouse for Birth Defects Surveillance and Research, and Overview of the Literature. *Am. J. Med. Genet. Part C* **2011**, *157*, 288–304. [CrossRef]
6. Swanson, A.B.; Barsky, A.J.; Entin, M.A. Classification of Limb Malformations on the Basis of Embryological Failures. *Surg. Clin. N. Am.* **1968**, *48*, 1169–1179. [CrossRef] [PubMed]
7. Tonkin, M.A. Classification of Congenital Anomalies of the Hand and Upper Limb. *J. Hand Surg. Eur. Vol.* **2017**, *42*, 448–456. [CrossRef]
8. Wilcox, W.R.; Coulter, C.P.; Schmitz, M.L. Congenital Limb Deficiency Disorders. *Clin. Perinatol.* **2015**, *42*, 281–300. [CrossRef]
9. McDermott, D.A.; Bressan, M.C.; He, J.; Lee, J.S.; Aftimos, S.; Brueckner, M.; Gilbert, F.; Graham, G.E.; Hannibal, M.C.; Innis, J.W.; et al. TBX5 Genetic Testing Validates Strict Clinical Criteria for Holt-Oram Syndrome. *Pediatr. Res.* **2005**, *58*, 981–986. [CrossRef] [PubMed]
10. Basson, C.T.; Cowley, G.S.; Solomon, S.D.; Weissman, B.; Poznanski, A.K.; Traill, T.A.; Seidman, J.G.; Seidman, C.E. The Clinical and Genetic Spectrum of the Holt-Oram Syndrome (Heart-Hand Syndrome). *N. Engl. J. Med.* **1994**, *330*, 885–891. [CrossRef]
11. Johnston, J.J.; Olivos-Glander, I.; Killoran, C.; Elson, E.; Turner, J.T.; Peters, K.F.; Abbott, M.H.; Aughton, D.J.; Aylsworth, A.S.; Bamshad, M.J.; et al. Molecular and Clinical Analyses of Greig Cephalopolysyndactyly and Pallister-Hall Syndromes: Robust Phenotype Prediction from the Type and Position of GLI3 Mutations. *Am. J. Hum. Genet.* **2005**, *76*, 609–622. [CrossRef] [PubMed]
12. Biesecker, L.G. The Greig Cephalopolysyndactyly Syndrome. *Orphanet J. Rare Dis.* **2008**, *3*, 10. [CrossRef] [PubMed]
13. Wagner, T.; Wirth, J.; Meyer, J.; Zabel, B.; Held, M.; Zimmer, J.; Pasantés, J.; Bricarelli, F.D.; Keutel, J.; Hustert, E.; et al. Autosomal Sex Reversal and Campomelic Dysplasia Are Caused by Mutations in and around the SRY-Related Gene SOX9. *Cell* **1994**, *79*, 1111–1120. [CrossRef] [PubMed]
14. Stoll, C.; Alembik, Y.; Dott, B.; Roth, M.-P. Associated Malformations in Patients with Limb Reduction Deficiencies. *Eur. J. Med. Genet.* **2010**, *53*, 286–290. [CrossRef] [PubMed]
15. Catena, N.; Calevo, M.; Adani, R.; Baldrighi, C.; Bassetto, F.; Corain, M.; Landi, A.; Lando, M.; Monticelli, A.; Novelli, C.; et al. Epidemiologia Della Malformazioni Della Mano e Dell'arto Superiore in Italia: Studio Multicentrico. 2017. Available online: https://www.researchgate.net/publication/321485275_Epidemiologia_della_malformazioni_della_mano_e_dell'arto_superiore_in_Italia_studio_multicentrico (accessed on 2 May 2024).
16. Borne, A.; Porter, A.; Recicar, J.; Maxson, T.; Montgomery, C. Pediatric Traumatic Amputations in the United States: A 5-Year Review. *J. Pediatr. Orthop.* **2017**, *37*, e104–e107. [CrossRef] [PubMed]
17. Conner, K.A.; McKenzie, L.B.; Xiang, H.; Smith, G.A. Pediatric Traumatic Amputations and Hospital Resource Utilization in the United States, 2003. *J. Trauma Inj. Infect. Crit. Care* **2010**, *68*, 131–137. [CrossRef] [PubMed]
18. Rodríguez-Galindo, C.; Navid, F.; Liu, T.; Billups, C.A.; Rao, B.N.; Krasin, M.J. Prognostic Factors for Local and Distant Control in Ewing Sarcoma Family of Tumors. *Ann. Oncol.* **2008**, *19*, 814–820. [CrossRef]

19. Ginsberg, J.P.; Goodman, P.; Leisenring, W.; Ness, K.K.; Meyers, P.A.; Wolden, S.L.; Smith, S.M.; Stovall, M.; Hammond, S.; Robison, L.L.; et al. Long-Term Survivors of Childhood Ewing Sarcoma: Report From the Childhood Cancer Survivor Study. *JNCI J. Natl. Cancer Inst.* **2010**, *102*, 1272–1283. [[CrossRef](#)]
20. Wang, M.N.H.; Chen, W.-M.; Lee, K.-S.; Chin, L.-S.; Lo, W.-H. Tuberculous Osteomyelitis in Young Children. *J. Pediatr. Orthop.* **1999**, *19*, 151–155. [[CrossRef](#)]
21. Wall, L.B.; Ezaki, M.; Oishi, S.N. Management of Congenital Radial Longitudinal Deficiency: Controversies and Current Concepts. *Plast. Reconstr. Surg.* **2013**, *132*, 122–128. [[CrossRef](#)]
22. Mano, H.; Fujiwara, S.; Haga, N. Adaptive Behaviour and Motor Skills in Children with Upper Limb Deficiency. *Prosthet. Orthot. Int.* **2018**, *42*, 236–240. [[CrossRef](#)] [[PubMed](#)]
23. Hadders-Algra, M. The Neuronal Group Selection Theory: Promising Principles for Understanding and Treating Developmental Motor Disorders. *Dev. Med. Child Neurol.* **2000**, *42*, 707–715. [[CrossRef](#)]
24. Meurs, M.; Maathuis, C.G.B.; Lucas, C.; Hadders-Algra, M.; Van Der Sluis, C.K. Prescription of the First Prosthesis and Later Use in Children with Congenital Unilateral Upper Limb Deficiency: A Systematic Review. *Prosthet. Orthot. Int.* **2006**, *30*, 165–173. [[CrossRef](#)]
25. Mavrov, M. *The Law Institute of Patient's Informed Consent*; Publishing House Stovi Group Bulgaria: Sofia, Bulgaria, 2018; pp. 17–18.
26. Belter, J.T.; Segil, J.L.; Dollar, A.M.; Weir, R.F. Mechanical Design and Performance Specifications of Anthropomorphic Prosthetic Hands: A Review. *J. Rehabil. Res. Dev.* **2013**, *50*, 599. [[CrossRef](#)]
27. Carey, S.L.; Lura, D.J.; Highsmith, M.J. Differences in Myoelectric and Body-Powered Upper-Limb Prostheses: Systematic Literature Review. *J. Rehabil. Res. Dev.* **2015**, *52*, 247–262. [[CrossRef](#)]
28. Bagley, A.M.; Molitor, F.; Wagner, L.V.; Tomhave, W.; James, M.A. The Unilateral Below Elbow Test: A Function Test for Children with Unilateral Congenital below Elbow Deficiency. *Dev. Med. Child Neurol.* **2006**, *48*, 569. [[CrossRef](#)]
29. Pruitt, S.D.; Varni, J.W.; Seid, M.; Setoguchi, Y. Prosthesis Satisfaction Outcome Measurement in Pediatric Limb Deficiency. *Arch. Phys. Med. Rehabil.* **1997**, *78*, 750–754. [[CrossRef](#)] [[PubMed](#)]
30. Liong, G.H.E.; Ridgers, N.D.; Barnett, L.M. Associations between Skill Perceptions and Young Children's Actual Fundamental Movement Skills. *Percept. Mot. Skills* **2015**, *120*, 591–603. [[CrossRef](#)] [[PubMed](#)]
31. Parr, J.V.V.; Vine, S.J.; Harrison, N.R.; Wood, G. Examining the Spatiotemporal Disruption to Gaze When Using a Myoelectric Prosthetic Hand. *J. Mot. Behav.* **2018**, *50*, 416–425. [[CrossRef](#)]
32. Biddiss, E.A.; Chau, T.T. Upper Limb Prosthesis Use and Abandonment: A Survey of the Last 25 Years. *Prosthet. Orthot. Int.* **2007**, *31*, 236–257. [[CrossRef](#)]
33. Huizing, K.; Reinders-Messelink, H.; Maathuis, C.; Hadders-Algra, M.; Van Der Sluis, C.K. Age at First Prosthetic Fitting and Later Functional Outcome in Children and Young Adults with Unilateral Congenital Below-Elbow Deficiency: A Cross-Sectional Study. *Prosthet. Orthot. Int.* **2010**, *34*, 166–174. [[CrossRef](#)] [[PubMed](#)]
34. Battraw, M.A.; Fitzgerald, J.; Joiner, W.M.; James, M.A.; Bagley, A.M.; Schofield, J.S. A Review of Upper Limb Pediatric Prostheses and Perspectives on Future Advancements. *Prosthet. Orthot. Int.* **2022**, *46*, 267–273. [[CrossRef](#)] [[PubMed](#)]
35. Major, M.J.; Fey, N.P. Considering Passive Mechanical Properties and Patient User Motor Performance in Lower Limb Prosthesis Design Optimization to Enhance Rehabilitation Outcomes. *Phys. Ther. Rev.* **2017**, *22*, 202–216. [[CrossRef](#)]
36. Murray, C. An Interpretative Phenomenological Analysis of the Embodiment of Artificial Limbs. *Disabil. Rehabil.* **2004**, *26*, 963–973. [[CrossRef](#)] [[PubMed](#)]

Disclaimer/Publisher's Note: The statements, opinions and data contained in all publications are solely those of the individual author(s) and contributor(s) and not of MDPI and/or the editor(s). MDPI and/or the editor(s) disclaim responsibility for any injury to people or property resulting from any ideas, methods, instructions or products referred to in the content.