INTRODUCTION
Lower limb amputation in children is relatively uncommon and there is scarce knowledge regarding the rehabilitation process for this population. In fact, most of the available literature focuses on adults. Some key information about rehabilitation may be transferable to children, but there are significant differences in the rehabilitation processes of pediatric and adult patients after amputation. These differences are influenced by the etiology of limb loss, motor development evaluation, bone growth, appositional overgrowth of the residual limb bone and functional prosthetic supply [1–3].

The most frequent indications for major lower limb amputations are traumatic injury, congenital limb deficiencies, severe infections, tumors, and vascular abnormalities (vasculitis and vascular malformation) [4]. The etiology of thrombosis in children is not fully understood. However, the factors causing or increasing the risk of thrombosis are clearly identified. The available research has shown a connection between the occurrence of thrombosis and amputation in infancy is not common. Therefore, the existing literature lacks sufficient evidence regarding this subject. Available research primarily focuses on treatment and causes of thrombosis in children. The paper describes a case of preterm infant after transtibial amputation due to common iliac artery thrombosis during the perinatal period. The girl began neurodevelopmental therapy based on normal development patterns of infants, which was provided three times a week for 45 minutes. The rehabilitation process also included prosthetics and surgical interventions. The assessment of motor development of the child was carried out at the age of 6, 9, 12, 18 and 20 months, using the motor scale of the Albert Infants Motor Scale (AIMS). Case analysis allows to state that infant rehabilitation after transtibial amputation should be individualized, and focus on supporting normal motor development, based on the correct movement sequences of infants. Additionally, the AIMS scale can be used to assess the gross motor development in infants after amputation and is useful in formulating early therapeutic intervention. A prosthetist should perform prosthetic revisions as often as needed, i.e., in infants that is usually every 4 months. What is more, successful outcomes can be achieved by implementing proper surgical procedures regarding the appositional overgrowth of the residual limb in growing children.

KEY WORDS: congenital thrombosis, amputation in infants, rehabilitation

OPIS PRZYPADKU
CASE REPORT
REHABILITATION OF INFANTS AFTER TRANSTIBIAL AMPUTATION DUE TO THROMBOSIS IN THE PERINATAL PERIOD. CASE REPORT

REHABILITACJA NIEMOWLĄT PO AMPUTACJI PODUDZIA Z POWODU ZAKRZEPICY W OKRESIE OKOŁOPOPORODOWYM. OPIS PRZYPADKU

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ABSTRACT
Congenital thrombosis and amputation in infancy is not common. Therefore, the existing literature lacks sufficient evidence regarding this subject. Available research primarily focuses on treatment and causes of thrombosis in children. The paper describes a case of preterm infant after transtibial amputation due to common iliac artery thrombosis during the perinatal period. The girl began neurodevelopmental therapy based on normal development patterns of infants, which was provided three times a week for 45 minutes. The rehabilitation process also included prosthetics and surgical interventions. The assessment of motor development of the child was carried out at the age of 6, 9, 12, 18 and 20 months, using the motor scale of the Albert Infants Motor Scale (AIMS). Case analysis allows to state that infant rehabilitation after transtibial amputation should be individualized, and focus on supporting normal motor development, based on the correct movement sequences of infants. Additionally, the AIMS scale can be used to assess the gross motor development in infants after amputation and is useful in formulating early therapeutic intervention. A prosthetist should perform prosthetic revisions as often as needed, i.e., in infants that is usually every 4 months. What is more, successful outcomes can be achieved by implementing proper surgical procedures regarding the appositional overgrowth of the residual limb in growing children.

KEY WORDS: congenital thrombosis, amputation in infants, rehabilitation

STRESZCZENIE
Wrodzona zakrzepica i amputacje w okresie niemowlęcym nie są powszechnie spotykane. Dlatego również w istniejącej literaturze brakuje wystarczających informacji dotyczących tego tematu. Dostępne badania skupiają się przede wszystkim na leczeniu i przyczynach zakrzepicy u dzieci. W pracy opisano przypadek wcześniaka po amputacji podudzia z powodu zakrzepicy tętnicy biodrowej wspólnej w okresie okołoporodowym, który uczestniczył w terapii neurorozwojowej, trzy razy w tygodniu przez 45 minut. Proces usprawniania dotyczył również protowanie i interwencji chirurgicznych. Ocenę rozwoju motorycznego dziecka przeprowadzono w wieku 6, 9, 12, 18 i 20 miesięcy, stosując skalę motoryczną Alberta Infants Motor Scale (AIMS). Analiza przypadku pozwala stwierdzić, iż rehabilitacja niemowląt po amputacji powinna być zindywidualizowana i skoncentrowana na wspieraniu prawidłowego rozwoju motorycznego, w oparciu o prawidłowe sekwencje ruchowe niemowląt. Dodatkowo, skala AIMS może być wykorzystana do oceny ogólnego rozwoju motorycznego u niemowląt po amputacji i jest przydatna w formułowaniu wczesnej interwencji terapeutycznej. Protetyk powinien przeprowadzać rewizje przez tak często, jak jest to konieczne, u niemowląt co 4 miesiące. Co więcej, pozytywne wyniki usprawniania można osiągnąć poprzez wdrożenie odpowiednich procedur chirurgicznych dotyczących apozycyjnego przerostu kikutu u dzieci w okresie wzrastania.

SŁOWA KLUCZOWE: wrodzona zakrzepica, amputacje u niemowląt, rehabilitacja
of thrombosis and inherited thrombophilia, deficiency of proteins C and S and antithrombin, as well as factor V Leiden gene mutation [5]. Other predisposing factors are infection, surgery, congenital heart defects, injuries, nephrotic syndrome, systemic lupus erythematosus, cancer and complications from its treatment (the presence of central catheter) [6]. Symptoms of thrombosis in children may differ depending on the thrombus size, location, and cause. The changes mainly concern the lower limb vascular system, though thrombosis may also develop in other organs, such as lungs, liver, and kidneys [7]. Lower limb thrombosis causes pain near the affected extremity, in the groin or in the abdomen. Additionally, there is swelling and reddish-purple discoloration of the skin. Prolonged ischemia can lead to trophic changes in tissue, resulting in necrosis. Ischemia may be life-threatening to children; that is why treatment methods include amputation of the affected limb [8]. Statistically, amputation is rare, but it can occur even as early as in infancy [1]. Surgical amputation of lower limbs in children differs from amputation in adults. General principles of amputation surgery in children are saving growth plates, performing disarticulation rather than trans osseous amputation, and preserving the knee joint, if possible [9]. Regardless of age, a properly executed surgery and the stump shape are crucial factors which maximize the immediate and future outcomes for the patient [10].

CASE PRESENTATION

This case report documents the assessment and rehabilitation of a girl born by caesarean section at gestational week 34. Her birth weight was 2680 g and she presented with an Apgar score of 8/10. At day one, she was admitted to the Intensive Care Unit and the Neonatal Pathology Unit due to suspected thrombosis in the left lower limb and respiratory failure. Both lower limbs were swollen, and there was reddish-purple skin discoloration (Fig. 1).

A thrombosis was diagnosed in the left common external iliac artery, and subsequent changes in the left femoral artery and the umbilical artery were detected using Doppler ultrasound. Initial treatment included heparin, pentoxifylline, and topical medication. On the third day of life, limb involvement was limited to the foot and the 1/3 distal part of the left leg. After eight days of conservative treatment, and due to necrotic lesions, the risk of sepsis, and the need for higher level amputation, surgeons decided to perform a transtibial amputation. No complications arose during surgery and the entire perioperative period. The girl was discharged after a month and a half of hospitalization following birth. At the age of 7 months, the girl was readmitted to the Department of Pediatric Surgery to undergo correction surgery of the left residual limb due to bone overgrowth. The bone overgrowth has been treated using conventional practice, i.e., a resection of the overlying bursa and shortening of the bone stump. As expected, there were no complications during the intra- and perioperative periods. After a three-day hospitalization, the child was discharged. Once wound healing occurred, the girl was fitted with the first temporary prosthesis, and rehabilitation was initiated.

INITIAL EXAMINATION

At the age of 6 months, the girl was examined by a physical therapist. Due to the lack of standardized developmental scale for children after amputation, the girl’s motor development was assessed using the Alberta Infant Motor Scale (AIMS) intended to assess motor development of healthy children. This screening tool is designed to examine, discriminate, and evaluate the movement of infants from birth to independent walking, and it is useful for monitoring gross motor developmental changes in the first 18 months of life. The AIMS also provides reliable and valid measurements in preterm infants [11]. The scale identifies components of dynamic movement by observing infants as they move into and out of four positions: prone, supine, sitting, and standing. Moreover, the AIMS scale is very efficient as it takes only 20 minutes to fill out the questionnaire and assess developmental changes in a child, both in quantitative and qualitative terms [12]. The observation of spontaneous movements was carried out at 6, 9, 12, 18, and 20 months of age. The AIMS is intended to evaluate normal development of infants. Meanwhile, the authors used the scale to assess the girl’s development and to determine the main therapy goals. The therapeutic intervention was designed based on accurate descriptions of movement sequences expected at a certain month of life.

DESCRIPTION OF THE INTERVENTION

When the girl was 6 months old, rehabilitation services were provided three times a week for 45 minutes. Neurodevelopment therapy based on the NDT-Bobath concept and normal development patterns of infants was used to learn the correct movement sequences. At the age of 8 months, the girl received her first temporary prosthesis. Once the prosthesis was prepared, the parents were trained on how to use it daily as well as how to encourage the child to use it. The girl faced difficulties adjusting to the prosthesis especially during the crawling stage because it often slipped off the residual limb upon hitting a piece of furniture or another obstacle. At 14 months, when the girl could pull up to a sitting position and

Fig. 1. The image of the girl’s condition on the second day of life at the Intensive Care Unit and the Neonatal Pathology Unit.
assume weight bearing in quadruped position, her balance and equilibrium reactions were deemed appropriate, and she was finally ready to stand while wearing the temporary prosthesis. Standing practice was performed at the couch, with fully extended hips and knees (without hyperextension), and a maintained alignment between the femur and tibia. During the therapy, at 14 months of age, the girl was taught how to use the prosthesis correctly in a standing position. She was given opportunities to develop side forward and backward stepping patterns. At the age of 18 months, due to the body growth the child was fitted with a new prosthesis with an additional silicone sleeve (Fig. 2).

At 18 months of age, the girl started to learn an alternating step-through gait pattern with the use of a posterior walker. At the age of 20 months, the girl began to take first independent steps without any equipment. During this rehabilitation phase, the girl experienced a complication and had to discontinue the therapeutic intervention. Appositional bone growth of the left tibia began to bulge at the bottom of the residual limb (Fig. 3). During the whole therapy period, the girl's parents were actively involved in the rehabilitation process. Their key role was to encourage the girl to regularly wear the prosthesis or delay her with daily activities, such as getting up or using the posterior walker.

THE APPOSITIONAL BONE OVERGROWTH AND SURGICAL INTERVENTIONS

The first surgical intervention took place at the age of 7 months. At first, the girl felt stump pain when too much pressure was applied on the stump in the standing position. She suddenly began to cry during every day play. At the age of 7 months, a small skin perforation appeared on the top of the residual limb. This was caused by a bony spike which has formed at the end of the tibia. The girl's parents also reported that the child woke up at night crying and pointing to the stump. The girl was referred to an orthopedist by the physical therapist. Then the girl underwent a surgical revision (capping of the medullary canal along with resection of the bony spike) which is necessary to correct growth disorders.

At the age of 20 months, the scar shifted to the middle of the stump and the surrounding soft tissues were flattened due to bone growth. The first signs of residual limb issues were manifested as the girl's reluctance to put on and wear the prosthesis. Most likely weight bearing on the left side of the body resulted in pain, which the girl communicated with crying. These changes in the residual limb disrupted correct weight bearing, proper prosthesis use, and the girl's tolerance of the prosthesis. Surgical resection was repeated after an orthopedic consultation. The girl will certainly receive a new prosthesis and once the wound is healed, she will return to the standing position and continue learning the correct gait.

DESCRIPTION OF THE NEURODEVELOPMENTAL OUTCOME

Gross motor function was evaluated six times for corrected age (CA) using the AIMS. The outcome measurements are presented in Table I.

For comparative purposes, during the last functional exam using the AIMS scale, the child was provided with a below-the-knee prosthesis. Since the girl was born at 34-week gestation, her scores on the AIMS are for her corrected age. Her corrected age was adjusted by 6 weeks or 1.5 months (the child's gestational age of 34 weeks was subtracted from a 40-week full-term). At the 6-month measurement, the girl obtained a total score of 23 and a percentile rank of 50%, which indicates that the patient demonstrated typical motor development for her adjusted age. At 4.5 months CA, the girl was able to roll supine to prone, lift, and also maintain head past 45° and controlled reach with free arms in prone position. The girl also reached hands to knees due to active abdominals and proper pelvic mobility in the supine position. In subsequent gross motor performance measurements, the girl's total score increased, but her skills were not age-appropriate. Therefore, the percentile range diminished with age in subsequent studies. At 7.5 months CA, additionally, she learned to pivot in the prone position and to sit with arm support. At 10.5 months CA, the girl used half-sitting and was

Fig. 2. The second temporary prosthesis with a stocking for the residual limb

Fig. 3. The postoperative scar on the left residual limb
able to roll prone to supine, crawl, and sit without arm support. At 16.5 months CA, she was able to creep, pull to stand, stand with arm support and without prosthesis, and move to the side by jumping on one leg with support. At the age of 20 months (18.5 months CA), the girl reached maximum results in the prone, supine, sitting and standing subscales. At 20 months, the girl’s percentile rank dropped significantly due to the fact that the child was not able to walk. During the 20-month evaluation, the child obtained higher scores when using the below-the-knee prosthesis. The device encouraged the patient to shift weight from side to side, cruise sideways, control lowering through standing, and stand alone. But most importantly, the girl was able to take several independent steps using a posterior walker.

Usually, the AIMS is intended to evaluate normal development of infants. However, in case of this study the scale was used not only to assess the girl’s development but also constituted the basis for determining the main therapy goals.

### TABLE I. Alberta Infant Motor Scale (AIMS) subscale and total scores.

<table>
<thead>
<tr>
<th>AIMS subscales and total score</th>
<th>6 months old (4.5 months CA)</th>
<th>9 months old (7.5 months CA)</th>
<th>12 months old (10.5 months CA)</th>
<th>18 months old (16.5 months CA)</th>
<th>20 months old (18.5 months CA)</th>
<th>20 months old with a prosthesis (18.5 months CA)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Prone</td>
<td>11</td>
<td>12</td>
<td>16</td>
<td>18</td>
<td>21</td>
<td>21</td>
</tr>
<tr>
<td>Supine</td>
<td>8</td>
<td>9</td>
<td>9</td>
<td>9</td>
<td>9</td>
<td>9</td>
</tr>
<tr>
<td>Sitting</td>
<td>3</td>
<td>5</td>
<td>12</td>
<td>12</td>
<td>12</td>
<td>12</td>
</tr>
<tr>
<td>Standing</td>
<td>1</td>
<td>1</td>
<td>2</td>
<td>6</td>
<td>9</td>
<td>14</td>
</tr>
<tr>
<td>Total score</td>
<td>23</td>
<td>27</td>
<td>39</td>
<td>45</td>
<td>51</td>
<td>56</td>
</tr>
<tr>
<td>Percentile</td>
<td>50%</td>
<td>25%</td>
<td>5%</td>
<td>&lt;5%</td>
<td>&lt;5%</td>
<td>&lt;5%</td>
</tr>
</tbody>
</table>

CA – corrected age; TH-percentile

### DISCUSSION

Thrombosis rarely appears during the neonatal period. Unfortunately, the necessary treatment results in surgical removal of the affected tissue. The available published studies indicate that around 42% of cases of limb thrombosis in infants ended up with amputation [8,14,15]. These surgeries were life-saving. Children with amputations should receive good care before and after surgery. The patients should be fitted with a prosthesis and afterwards receive a long-term rehabilitation. It should be noted that the surgical team makes extremely important decisions about the level of amputation [16]. Another factor which plays an essential role in prosthesis selection is the scar placement on the stump. The prosthesis socket should not press against the scar. Furthermore, prosthesis fitting should occur as fast as possible and it usually occurs in 8 month old infants as its lack significantly impacts the child’s functional development [17]. Properly selected prosthesis supports maximum function, symmetry, and the development of a correct body map and a normal sensorimotor experience. The prosthesis was not equipped with a mechanical suspension mechanism proximal to the condyles, which means that it allowed typical development of quadriceps, without any reduction of muscle mass or the residual limb circumference [13]. At the age of 14 months, when the girl learned how to get up and use side walking, she received a modular prosthesis with a single axis foot. At 18 months, following another consultation with the prosthetist, the prosthesis socket was replaced by a bigger one due to increased circumference of the residual limb. Additionally, a silicone sleeve was used to control tightening over the stump. It allowed the stump to adhere and compress soft tissues into the shape required to accommodate the rigid socket. The silicone sleeve kept the prosthesis socket on the residual limb in a stable position and prevented it from slipping off during dynamic movements while learning to walk.

The protheses were designed and manufactured using a traditional cast method. The prosthetist followed up with the girl regularly every 4 months to ensure that the device was still adjusted to the child’s growing body. Changes in residual limb length and volume were monitored during the follow-up visits. The physical therapist was also in constant contact with the prosthetist, who controlled the quality of the prosthesis in use.
used, then the research would have been more time-consuming and expensive.

Furthermore, the most significant post-surgical complication described in the rehabilitation process was the appositional bone overgrowth, which caused skin perforation, and consequently made it difficult to cure. As a result, only a greater number of surgical revisions may prevent the appositional overgrowth in the residual limb, and it may be possible to change the surgical procedure during the subsequent reoperation to affect the improvement process itself [16, 24]. Appositional overgrowth of the residual limb occurs in 4% to 35% of children and is most common in below-knee amputation [16]. In every case, revision surgery may prove necessary, particularly to correct growth disorders. The child and parents should be informed of this possibility.

The purpose of physical rehabilitation of infants after amputation is to provide them with a new experience through achieving correct motor sequences expected during infancy. It is also equally important to properly educate parents on supporting the development of a small child and encouraging them to accept the prosthesis and becoming more independent in numerous real-life situations.

CONCLUSIONS

Based on this case description, the AIMS scale can be used to assess the gross motor development in infants after amputation and is useful in formulating early therapeutic intervention. The approach to infants after transtibial amputation should be individualized, and focus on supporting normal motor development, based on the correct movement sequences of infants. Prosthesis revisions should be performed as often as needed, i.e., in infants that is usually every 4 months. What is more, successful outcomes can be achieved by implementing proper surgical procedures regarding the appositional overgrowth of the residual limb in growing children.

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Conflict of interest:
The authors declare no conflict of interest.

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