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Case Report
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Upper Limb Congenital Gangrene — 3D-Printed Prosthesis: A Case Report

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Abstract

Congenital gangrene at limb level is a rare event. More than 100 cases of perinatal gangrene have been reported in literature only 10% of them were intrauterine gangrene. Congenital or intrauterine gangrene is the onset of necrosis before birth and neonatal gangrene is acute ischemia that occurs during the birth evolving in necrosis if the cause of ischemia is not removed. The causes of perinatal gangrene include hypercoagulable states, thromboembolic disease, maternal diabetes, congenital rubella and varicella, constriction bands, umbilical artery or peripheral catheterization, septicemia, and necrotizing fasciitis. A newborn girl was referred to our attention due to an advanced state of distal ischemia at the elbow level. The ring and the fingers had a small advanced necrosis stage with several blisters and 1 cm below the elbow there was a semi-circumferential fibrotic line involving the cutaneous and subcutaneous tissue.

Keywords: Congenital gangrene; Prosthesis; Newborn; 3D-printed

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Introduction

Congenital gangrene at limb level is a rare event. The first described case of ischemic gangrene in a newborn was reported by Martini in 1828 [1]. More than 100 cases of perinatal gangrene have been reported in literature only 10% of theme were intrauterine gangrene [2].

A congenital or intrauterine gangrene is the onset of necrosis before birth and neonatal gangrene is an acute ischemia occurs during the birth evolving in necrosis if the cause of ischemia is not removed.

The causes of perinatal gangrene include: Hypercoagulable states, thromboembolic disease, maternal diabetes, congenital rubella and varicella, constriction bands, umbilical artery or peripheral catheterization, septicemia and necrotizing fasciitis. Although most of the cases are idiopathic [3-5]. In the majority of the cases small part of the limbs are involved mainly at lower level.

The surgical treatment needs to be postponed as much as possible using adequate antibiotic therapy to allow the demarcation of the necrosis and perform an amputation as minimal as possible.

Case Presentation

We report a case of intrauterine gangrene in a newborn, involving the upper extremity. That was treated with surgery and prosthesis created using a 3D-printer.

A newborn girl was referred to our attention due to an advanced state of distal ischemia at the elbow level. The ring and the fingers had a small advanced necrosis stage with several blisters and 1 cm below the elbow there was a semi-circumferential fibrotic line involving cutaneous and subcutaneous tissue (Figure 1).

Movements were preserved at shoulder and elbow, none at the hand and fingers. The baby was born at 38 weeks of gestational and the pregnancy was uneventful. Ultrasound (US) performed

Figure 1: Movements were preserved at shoulder and elbow, none at the hand and fingers.

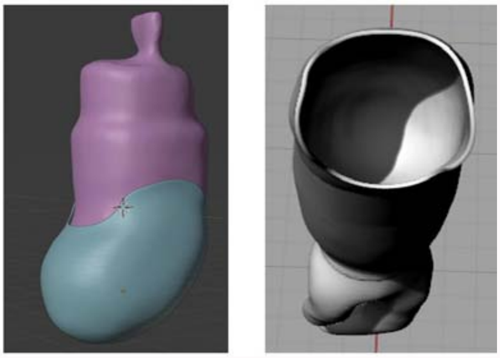

Figure 2A: 3D model precise measurements on anatomic landmark were done and finally a printing phase using 3D.

during pregnancy showed no alterations. No history of maternal hypertension, diabetes, renal disease, viral infections during pregnancy and family thrombotic or hemorrhagic diathesis was reported.

A computed tomography angiography performed few hours after birth, showed an arterial stop of both radial and ulnar vessels.

The placenta referred to the pathology department for analysis and no amniotic bands or other pathological features were detected.

Diagnosis of neonatal gangrene was confirmed. There was no sign of any revascularization attempt due to advanced saponification state of all the tissues related to amniotic liquid immersion.

The baby was tested to identify associated anomalies, (i.e., ophthalmological, cardiac and abdominal). Blood tests included specific thrombophilic researches and blood cultures were performed. Cardia US found a small defect on the interventricular wall at the cardiac apex and the PT-INR was slightly elevated (1, 25, seconds range: 0.8 to 1.17).

We decided to apply a medication to the limb and to wait few days for the necrosis area establishment. At five days of life an amputation was performed under the elbow, leaving a stump as long as possible to preserve the elbow motion and to allow a possible prosthesis implantation.

One month later the baby presented a tiny radial exposure due to a wound infection. She underwent a swab with the isolation of a *Pseudomonas aeruginosa* and an *Enterococcus faecalis* treated with appropriate antibiotic therapy.

Figure 2B: The cuff was applied to ensure a comfortable stump housing inside the prosthesis socket.

A Magnetic Resonance Imaging showed no signs of osteomyelitis. A revision surgery with an accurate cleaning procedure and a direct closure of the stump was performed.

At the end of therapy, the skin healed with no signs of infections or neurological defects.

Six months after the operation, the subject was evaluated by a physiatrist and particular attention was done regarding the stump, in particular at the amputation level of the stump [7], at elbow articulation and the general development of the arms. Due to the fact that no problems were detected we decided to apply for prosthesis. In the beginning, a manual stump measurement was performed following by a 3D scanning. Using the 3D model precise measurements on anatomic landmark were done and finally a printing phase using 3D was performed (Figure 2A).

The material with Medical use certification, used for printing is a thermo-bioplastic melted and extrude. This material is flexible and resistant (27 sh/D), and simple to print. This specific material is pleasant to the touch having all requested certification by the law. After the printing, a manual phase of the model took place, in order to make the prosthesis the most comfortable for the patient and a direct test on the patient was performed. The prosthesis consists in a prosthesis anchor-age bracelet with Velcro closure, adjusting straps in order to obtain functional and comfortable bracelet position. The cuff used as a stump-prosthesis interface was made using a soft biocompatible foam that is a replication of the stump like a glove. The cuff was applied to ensure a comfortable stump housing inside the prosthesis socket (the limb residue is inserted into the empty chamber inside the prosthesis) (Figure 2B).

Discussion

We built a prosthesis with specific measurements using a 3D printer for the comfort given by the extreme customization made to measure, the lightness and breathability, modular, with the absence of ingestible minute parts, customizable in design, and relatively low cost [6].

According with previous experiences found in the literature, we decided to apply the prosthesis as earlier possible due to the fact it is known that children with unilateral upper limb deficits mounted before 2 to 3 years tend to accept their prosthesis more than those mounted after 2 years [7].

A multidisciplinary team approach having physiatrist, surgeon, pediatrician, physiotherapist and orthopedic technician is mandatory to adequate rehabilitation and specific follow-up with family involvement is fundamental [8] also to minimize the possible discomfort that is not easy to ex-plain by the young patient [6,9].

Due to these characteristics, the prosthesis should be serially redone following the patient's growth. An ongoing training program was formulated at individual level taking into account the kind of amputation and the evolutionary state of the patient [6].

In particular, we have focused the training through play of the use of the amputated arm and control of the prosthesis, with the achievement of the normal joint range and strengthening of the muscle, of the postural passages by resting on the limb with the prosthesis.

The patient is followed twice a week by physiotherapists and a medical evaluation is performed every 4 weeks.

At the first follow-up at 1 month of treatment, the patient showed an improvement in the ability to support from prone to switch to a sitting position and a good compliance of the prosthesis.

Conclusions

We presented a case of perinatal gangrene to the upper limb followed by amputation under the elbow and implantation of a prosthesis custom built with 3D printer.

We believe that timing the prosthesis has been assembled could help the patient to grow with a harmonious development of the body scheme.

The relative simplicity of the 3D printing technique could represent a unique alternative to prostheses assembled in the traditional way.

Follow-up of the patient is underway to accomplish the growth and to prepare for a future implant of a myoelectric prosthesis.

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