The Use of a Cell-free Chondroinductive Implant in a Child With Massive Cartilage Loss of the Talus After an Open Fracture Dislocation of the Ankle: A Case Report

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Background: We present a case report of a 3-year-old girl who sustained a severe open fracture dislocation of her talus with complete loss of full-thickness articular cartilage and subchondral bone over 80% of the talar dome. At presentation there was an extensive soft tissue defect including absent anterior joint capsule. She required a free anterolateral thigh flap to reconstruct this defect. The talar dome defect was treated with a cell-free chondroinductive implant made of resorbable polyglycolic acid felt and hyaluronic acid. This was the first use of such an implant in the United Kingdom and the first use in a child anywhere in the world.

Methods: The case has been followed prospectively for 3 years.

Results: At 3 years postoperative, the patient underwent thinning of the anterolateral thigh flap and trimming of an anterior tibial overgrowth, which was causing impingement. At surgery the talar dome cartilage looked pristine, with a line representing the tidemark between the original cartilage and the new formed. Biopsies were taken and histopathology performed.

Conclusions: This was a rare and difficult case that has achieved an excellent outcome at this follow-up stage.

Level of Evidence: Level V.

Key Words: chondroinductive, osteochondral injury, open fracture, pediatric, talar fracture

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In the adult population osteochondral injuries of the talus account for 0.09% of all fractures and 1% of all talus fractures.[1] In the pediatric population, injuries to the body of the talus are very rare. They usually present in the adolescent age group (average age of 13)[2] with very few occurring before the age of 10. Children with talar osteochondral injury report a traumatic episode in up to 63% of cases.[3,4] Generally, these patients can present following acute trauma or with chronic ankle pain. Massive osteochondral injury is very rare, although Sneppen et al[5] included osteochondral injury in his talar body classification. He described it as the result of compression. In contrast, osteochondral injury associated with talar dislocation is said to occur due to shear.[6] Although open talar extrusions have been rarely reported in the literature,[6–8] we could find no reports of open osteochondral talar injury in the adult or pediatric population.

We present a unique case of an open fracture dislocation of the ankle with massive osteochondral injury to the talus caused by friction in a young child. As far as we are aware it was also the first cell-free chondroinductive implant to be used in a child anywhere in the world.

CASE REPORT

A 3-year-old girl was transferred into our Regional Plastic Surgical Unit 1 day after a road traffic collision. It was reported that the child had been dragged for several feet by a car. The child was taken to a district general hospital emergency department where advanced trauma life support protocols were adhered to, although the only apparent injury was to the left lower limb. A very deep friction burn had been sustained over the left ankle, with an underlying open ankle fracture dislocation. Routine British Orthopaedic Association/British Association of Plastic, Reconstructive and Aesthetic Surgeons protocol for treatment of open fractures was undertaken, including intravenous cephalosporin antibiotic infusion, removal of gross contamination in the emergency department, and saline-soaked gauze coverage of injury and splintage. The fibular had sustained a displaced Salter Harris I fracture and the talus had sustained a massive osteochondral injury. The distal sensation was present apart from a small patch distal to the wound. The capillary refill time of the limb was 2 seconds.

The patient was transferred as per local guidelines, and initial wound debridement was performed within 24 hours of injury at our department. The talar dome had been denuded of 80% of its cartilage with loss of a degree of subchondral bone, with only the medial chondral shoulder and part of the talar neck cartilage remaining (Figs. 1, 2). The Salter Harris I fracture of the distal fibular was unstable. On examination of the overlying soft tissue envelope, the anterior ankle capsule was absent

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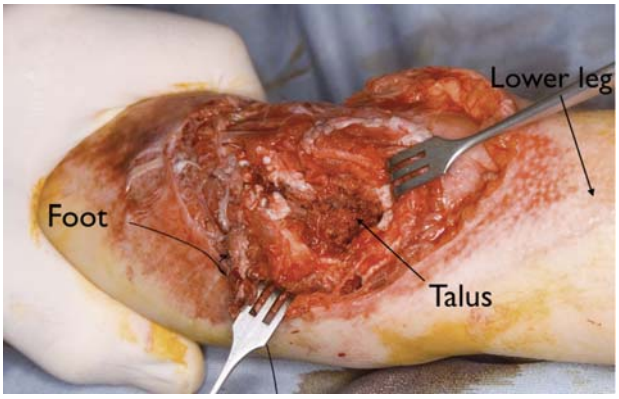

FIGURE 1. Intraoperative photograph of left ankle wound during initial debridement. The talar dome has been denuded of 80% of its cartilage with loss of a degree of subchondral bone. The anterior ankle capsule is absent. full color online

and the extensor digitorum comminis was 100% divided. Extensor hallucis longus and tibialis anterior were present. After debridement, the resulting soft tissue defect measured 8×4.5 cm.

After careful consideration and long discussions with the parents of the child, definitive surgery was undertaken 5 days after injury. The osteochondral injury was treated with a single layer cell-free chondroinductive implant (Chondrotissue; BioTissue AG, Zurich, Switzerland) with hyaluronic acid as per product specifications, and was anchored with 5.0 vicryl sutures and Tisseel fibrin sealant (Baxter, Newbury, UK) around the margins. A single Kirschner wire was placed across the medial malleolus and under the graft to prevent shear stresses and improve stability. The extensor communis was transferred to the intact extensor hallucis longus. The Salter Harris fracture was treated by suture fixation and protection with the Kirschner wire in the reduced position. The soft tissue defect was treated using a free anterolateral thigh flap anastomosed end to end to the anterior tibial vessels and limited split-skin grafting. Postoperatively the Kirschner wire was removed at 2 weeks, when passive ranges of motion exercises were commenced. A removable splint was used for the first 4 weeks postoperative and the child was kept non–weight-bearing throughout this time.

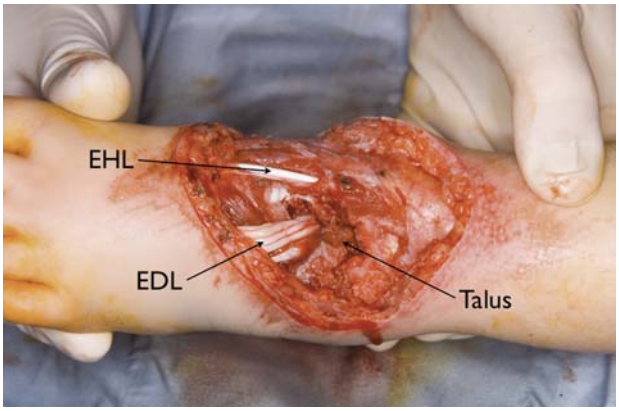

FIGURE 2. Further debridement of devascularized skin reveals the extensor digitorum comminis is 100% divided. Extensor hallucis longus and tibialis anterior are both intact. EDL indicates extensor digitorum longus; EHL, extensor hallucis longus. full color online

The patient underwent regular review under the care of both the plastic and orthopaedic departments. The wounds healed well. At 10 months postoperative an MRI scan was performed to evaluate the chondral surface of the talus. The cartilage appeared thick and intact with no underlying edema of the talus (Figs. 3A, B). At 15 months postoperative, the range of motion of the ankle declined with loss of dorsiflexion. Ankle range of motion at that time was measured as 5 degrees dorsiflexion and 45 degrees plantar flexion. An impingement lesion had developed on the anterolateral tibia margin. This loss of motion did not progress and it was decided to not intervene at this stage.

At 3 years after injury, the child had grown enough to contemplate refashioning of the anterolateral thigh flap, to cosmetically improve the appearance of the ankle. This was achieved by debulking the free flap and revising the area that had been previously split-skin grafted. We took this opportunity to treat the anterior impingement of the left ankle at the same time. On opening the joint it was noted the anterolateral impingement lesion was a consequence of overgrowth of the tibial plafond and this was excised (Figs. 4, 5). The talar surface, where the cell-free chondral graft was used, looked and felt normal. As agreed with the parents, biopsies of the child's talar cartilage were taken. These biopsies were taken from the central dome (where the chondroinductive implant had been grafted), from the talar neck (normal cartilage), and the tidemark zone between the 2. The biopsies demonstrated a cartilage matrix rich in cells with a homogenous cell distribution (Fig. 6). The columnar cell arrangement of the chondrocyte cells, in contrast to native tissue, was only a little less structured but generally with very similar appearances. Unlike fibrocartilage there were no fibroblasts, a homogenous cartilage-bone-interface, and a hyaline-like to hyaline structure. The collagen type II and proteoglycan formation in particular in the central and lower part of the biopsies was typical for hyaline-like cartilage. There were no visible scaffold residues from the chondral graft used. Currently, the child has an improved dorsiflexion of the ankle 3 months after surgery. The cosmetic appearance is satisfactory.

DISCUSSION

Traumatic osteochondral injury of the talus usually results in lesions originating on the lateral angle of the talar dome.[2] This is likely to originate from a shearing mechanism in supination external rotation injuries to the ankle, which is the most common mechanism of injury in all ankle fractures. Talar injuries are often associated with other fractures, with Jensen et al[9] reporting an incidence of 43% (of 14 patients), the most common being ankle fractures. Both these statements were true in our case as there was an additional Salter Harris I fracture of the distal fibular and the lateral talar body was denuded of cartilage. The talar cartilage loss was more likely the result of the friction of the road than the twisting motion of the talus in the mortise. We could find no previous open osteochondral injury of the talus in either the adult or the child reported in the literature.

Normal articular cartilage is a highly organized tissue with a composite make up of both organic solid matrix, saturated with water and mobile ions.[10] The regeneration of cartilage remains a very difficult challenge

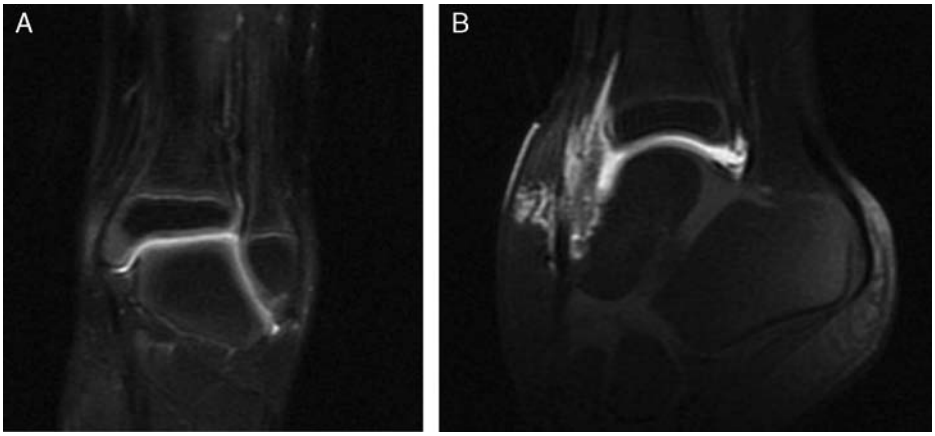

FIGURE 3. A and B, A MRI taken 10 months after injury. There appears to be thick uninjured talar cartilage with no underlying osseous edema on the coronal and sagittal sections of this T2-weighted scan.

due to the complex physiology, high mechanical loads it endures, and the poor healing potential. Although surgical regenerative procedures such as microfracture, mosaicplasty, autologous chondrocyte implantation, and osteochondral transfer are commonly performed, large cartilage defects such as presented in this study are formidable and uniformly have poor results.[11] A number of biomaterials are becoming available to augment surgical procedures, by providing a mechanically stable support and facilitate chondrocyte number expansion and organization.[10] Dinescu et al[12] found that hyaluronic acid was a potent prochondrogenic factor and human adipose-derived stem cells had high chondrogenic potential. These factors are now in common use with the available scaffolds, such as used in this case.

Our patient presented with a rare combination of complex injuries and reconstructive challenges. We present the clinical outcome of this exceptional case that was unique in not only its injury but also its treatment. The decision to use the cell-free chondroinductive tissue, which consists of an absorbable nonwoven polyglycolic acid textile treated with hyaluronic acid, was made after an open and frank discussion between the parents and the surgical team and following ethical approval from the hospital Trust management on a single case basis. It was decided that the benefit of attempting cartilage regeneration of the talus by using the graft outweighed the risk, even when considering the risk of infection in putting a foreign material into a wound that was likely to be colonized with bacteria. The implant had been used previously to treat osteochondral lesions of the knee[13–15] and the talus[16] in patients undergoing microfracture. As shown previously in an ovine joint cartilage defect model, covering of articular cartilage defects pretreated with microfracture with the chondroinductive implant improves the formation of cartilage repair tissue compared with microfracture treatment alone.[17] The implant may recruit subchondral stem cells that have been shown to have high chondrogenic potential. Ingrowth of stem cells may enrich the number of stem cells within the implant and thus the defect area, whereas the hyaluronic acid incorporated into the implant guides them toward cartilage formation.[18,19] Therefore, as the talar medullary

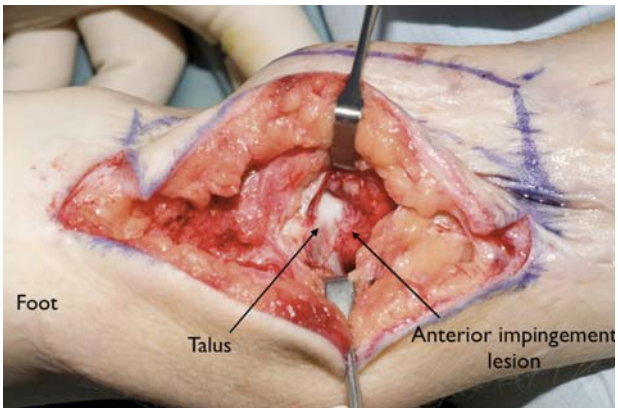

FIGURE 4. Intraoperative photograph illustrating the anterior impingement lesion originating from the tibial plafond. full color online

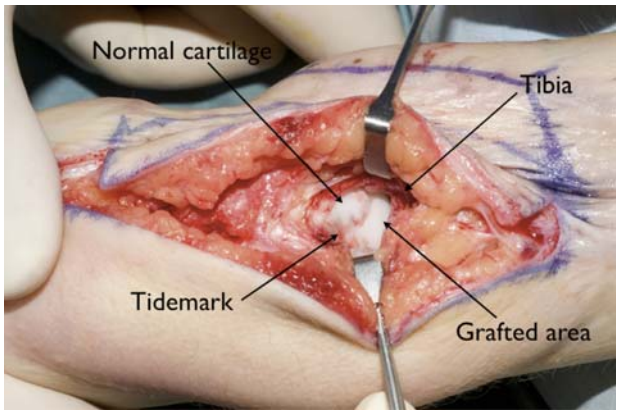

FIGURE 5. Intraoperative photograph after excision of anterior impingement lesion. The talar surface, where the cell-free chondral graft was used looks and felt no different from the normal cartilage. The tidemark between the 2 areas remains visible. full color online

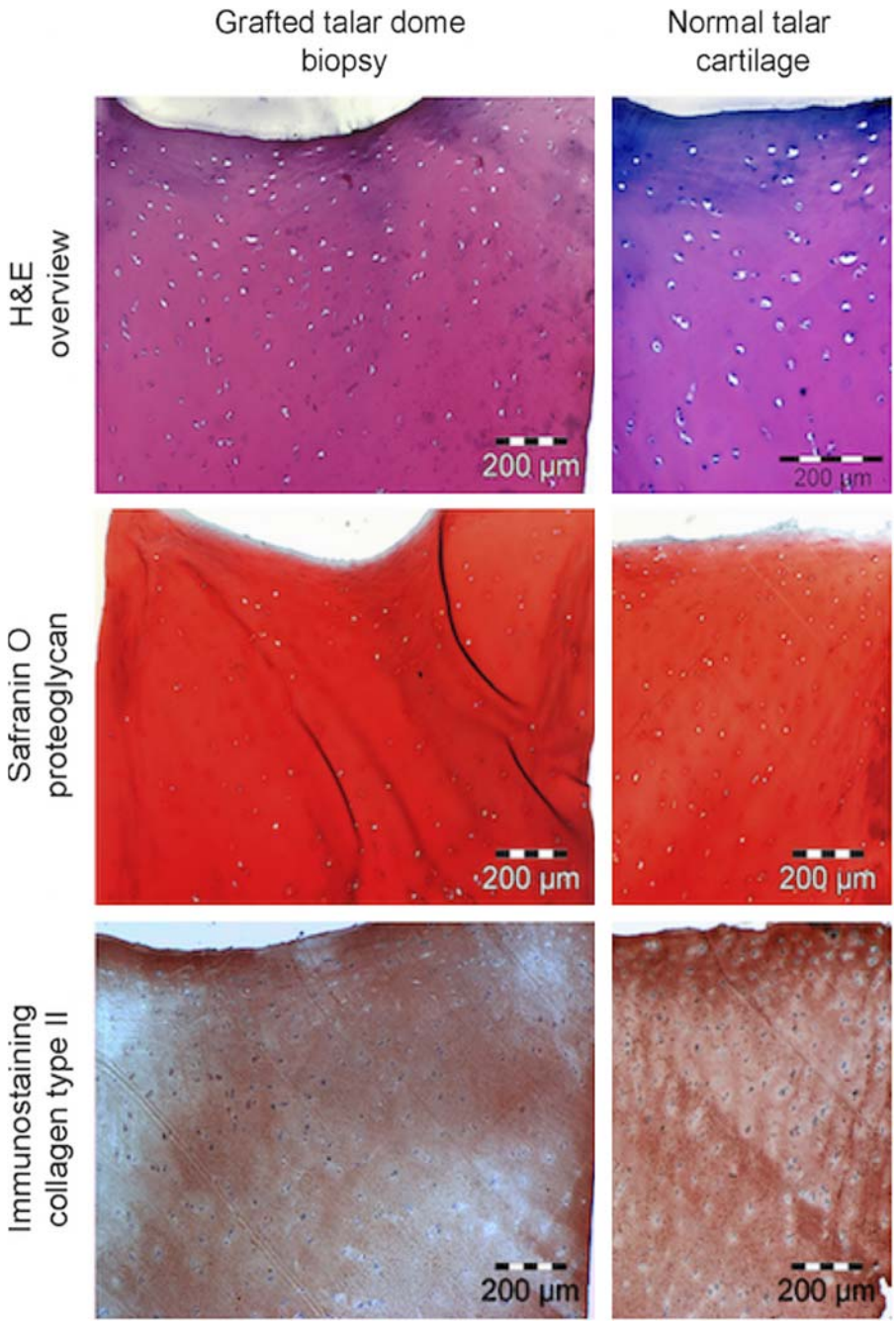

FIGURE 6. Biopsies taken of the graft talar dome and of normal talar articular cartilage stained with hematoxylin-eosin, safranin O, and anticollagen type II antibodies. The staining helps to evaluate some general aspects of the tissue-like cell arrangement, cell type analysis, tissue homogeneity, and tissue integration as well as hyaline to hyaline-like cartilage matrix formation. The biopsy of the grafted area demonstrated a matrix rich in cells with a homogenous cell distribution and presence of typical cartilage matrix components like proteoglycan and collagen type II very similar to the normal cartilage. full color online

bone was exposed, pluripotent marrow-derived stem cells were freely entering the injury site. By using the implant, we were able to contain these cells and give a scaffold to allow new chondral growth. This treatment regime was fortunately successful; the histology of the cartilage of the treated area 3 years after injury revealed almost normal chondral cartilage.

It may be argued that due to the age of the child, the healing potential is such that a satisfactory result would have been obtained without the use of a graft. In a bovine model, Liu et al[20] found that juvenile articular cartilage possessed superior chondrogenic activity and enhanced regenerative potential as compared with their adult counterpart. However, the longest outcome study performed on

talar fractures in children revealed that all displaced fractures developed talar-crural arthrosis at follow-up.[9] In addition, most authors recommend surgical treatment in regards to displaced osteochondral injury in children.[4,21,22] We feel that an excellent outcome has been achieved in a patient who had devastating injuries at presentation. A consequence of the injury has been overgrowth of the tibial plafond, which caused impingement. This may recur before completion of growth in this patient.

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