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Total knee arthroplasty and patelloplasty in a patient with phocomelia caused by thalidomide

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Abstract

The treatment of osteoarthritis in patients with phocomelia with total knee arthroplasty is challenging due to the unusual anatomy and severe deformities. The authors present a case of phocomelia caused by thalidomide with end-stage osteoarthritis and grossly medialized patella. The patient was treated with a cemented constrained non-hinged prosthesis and patelloplasty. Six months later, the patient had complete relief of pain and was able to walk without walking assistance. To our knowledge, total knee replacement in a patient with phocomelia caused by thalidomide has not been described in literature.

Keywords

Total knee arthroplasty, constrained knee prosthesis, patella transfer, phocomelia, thalidomide

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Introduction

We present a unique case of a one-stage total knee arthroplasty (TKA) in a patient with phocomelia caused by thalidomide.

Due to the unusual anatomy and severe deformities, special considerations are necessary when performing TKA in patients with skeletal dysplasia. Implant selection and soft tissue balancing can be challenging, but short-term clinical results consistently reveal improvements in pain and function.[1] Many dysplastic conditions develop osteoarthritis (OA) of the knees, including, among others, hereditary multiple exostosis, achondroplasia, osteogenesis imperfecta, and phocomelia.[1]

Phocomelia is a rare congenital defect defined by the absence of intermediate segments of the extremity. Children with phocomelia present with their hands or feet directly attached to the trunk. Phocomelia famously is a teratogenic side effect of the drug thalidomide. Thalidomide was advertised to treat anxiety and morning sickness between 1957 and 1961. As a result, more than 10,000 children were born with debilitating malformations.[2] Although thalidomide affects most of the developing tissues and organs of the body, the damage to the limbs is most noticeable.

With age and skeletal deformities being major risk factors for OA, today, 60 years after thalidomide's introduction to the market, a rising number of patients may begin to need surgical treatment for OA of the knee.[3]

To the best of our knowledge, performing a TKA and a patella transfer in the challenging setting of a patient with phocomelia caused by thalidomide has not been described in literature.

Case presentation

A 59-year-old female presents with end-stage OA of the left knee and phocomelia caused by thalidomide.

In 1979, bilateral flexion contractures were treated with a soft tissue release on both knees. During rehabilitation, the patient suffered a proximal tibial fracture of the left knee. The fracture was surgically treated with open reduction internal fixation (ORIF), and the osteosynthetic material was

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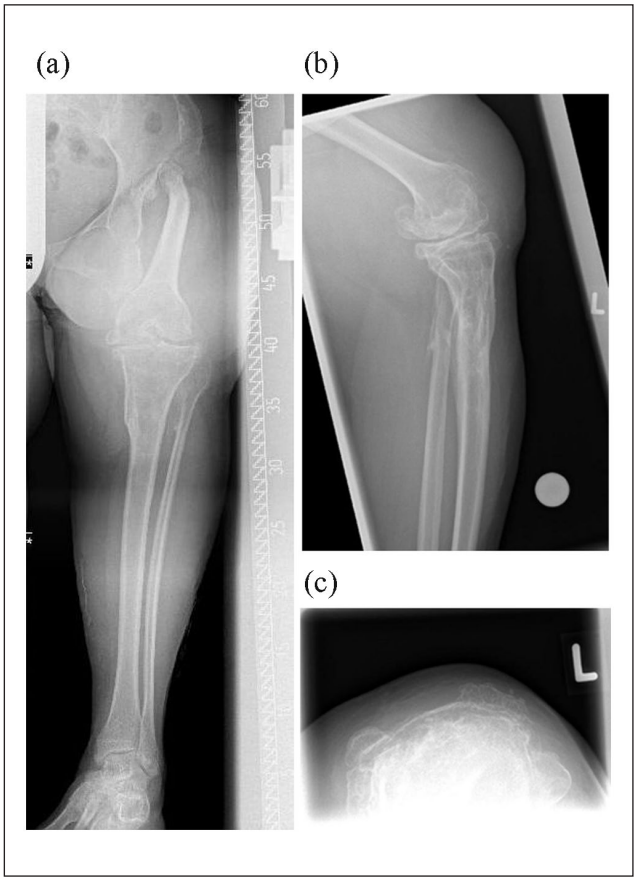

Figure 1. (a–c) Preoperative X-rays of the patient.

removed consecutively. In the further course, endoprosthetic restoration of the right knee with a non-hinged and non-constrained knee prosthesis was carried out in 1996. Because of an inlay damage, instability, and metallosis, a revision with implantation of a constrained hinged prosthesis was performed 4 years later. Afterward, the patient was symptom-free with regular mobility.

Concerning further comorbidities, no other systemic illnesses were noted except for hypertension.

The patient contacted us in July 2021 because of persisting pain in her left knee for a couple of months. She reported using over-the-counter analgesics and nonsteroidal anti-inflammatory drugs (NSAIDs). X-rays showed an ankylosing posttraumatic knee OA in all three compartments (Figure 1).

On clinical examination, the patient (139 cm, 46 kg, body mass index 23.8 kg/m^2) showed a limping gait pattern. The scar tissue of both knees was irritation-free. The range of motion (ROM) of the left knee was for extension/flexion: 0/10/80°. There was no relevant gapping in valgus or varus stress test. Leg length difference was +2.5 cm on the left side. Digital preoperative planning (TraumaCAD™, Petah-Tikva, Israel) was performed based on long-leg weight-bearing X-rays.

The operation was performed by a senior surgeon with more than 30 years of experience in TKA. A surgical approach in a standard fashion via anterior parapatellar approach was chosen with the patient in a prone position. Ample scar tissue, due to prior ORIF, was released and removed upon preparation.

Due to the small anatomic conditions, poor ligamentous balancing, and the early revision of the contralateral side, the surgeon chose to implant a cemented constrained non-hinged prosthesis (ROKNEP; Waldemar Link GmbH, Hamburg, Germany; size extra small, with patella shield). The smallest implant size available (XS) was selected, confirming preoperative templating. To match anatomical conditions, the femoral stem needed additional trimming at 4 cm. Trimming of the stem was performed with a diamond saw (ELAN 4; Aesculap AG, Tuttlingen, Germany). Thereafter, the prosthesis was implanted in standard fashion.

Intraoperatively, the patella showed a diameter of only 1 cm and the patellar tendon presented grossly medialized. Centralization of the patella by performing a medial retinaculum release and lateral tensioning was insufficient. To obtain central patellar tracking, a lateralizing free patella transfer was performed. First, the patella was carefully dissected out of the medial subluxation position together with a small rim of the surrounding retinaculum resulting in a free patella bone graft. The donor site of the patella graft was closed with absorbable 2-0 sutures (Vicryl, Ethicon Inc., Somerville, USA) to restore an intact patellar tendon. Thereafter, the patellar tendon was able to be centralized conveniently. An interval between the synovial lining and the patella tendon was developed at the central area of the tendon. The subsynovial pouch was chosen to be slightly larger than the patella to allow easy positioning of the graft in its final position. The free patella bone graft was then slipped into the pouch and positioned into the best tracking position with respect to the femoral component. Once the best position was determined, the graft was sewn with 2-0 absorbable sutures (Vicryl, Ethicon Inc., Somerville, USA) into the pouch by passing the needle through the tendon and surrounding retinaculum of the patella. Additional lateral tensioning was performed to ensure correct patella tracking in the femoral groove. Ultimately, the patella showed a sufficiently centralized motion during the entire ROM. The intraoperative ROM was 0/10/110°.

The duration time of surgery from skin incision to closure was 133 min.

Postoperatively, the patient was immediately mobilized without weight-bearing restrictions under physiotherapeutic supervision. Passive ROM was applied to achieve sufficient extension. The patient was able to walk safely with crutches by the time of discharge. The final ROM at discharge was 0/5/80°. The patient was discharged home on the sixth postoperative day. The postoperative X-rays showed a correctly implanted prosthesis with a straight leg axis and a central patella (Figure 2). Thrombosis prophylaxis with enoxaparin

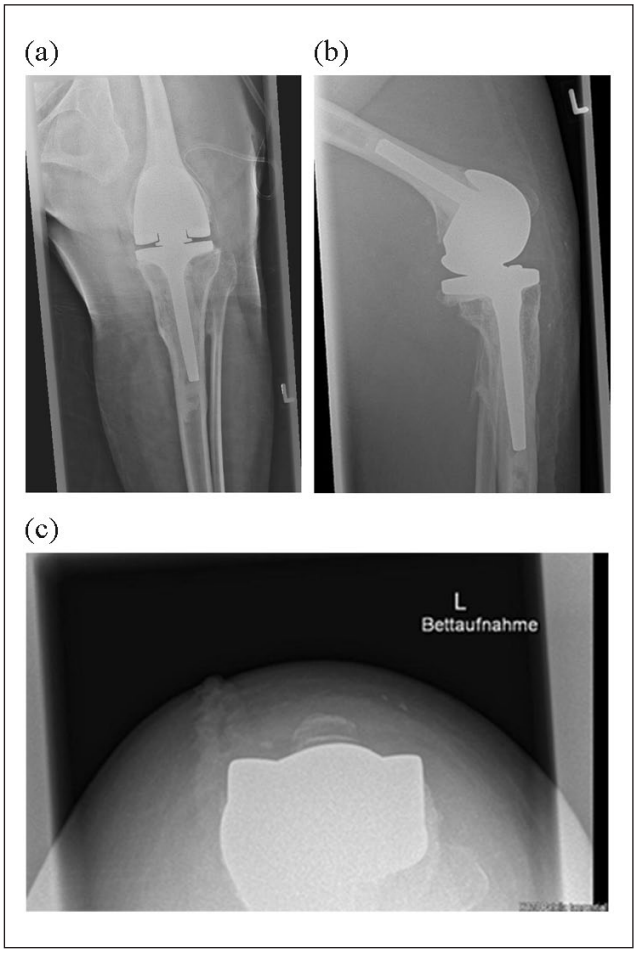

Figure 2. (a–c) Postoperative X-rays of the patient.

40 mg was continued until the patient was fully stressed. However, 6 months postoperatively, the patient was able to walk without crutches. Extension of the leg was possible against force and the ROM was 0/0/110°. The patient was very satisfied with the result.

Discussion

Because of the low prevalence of TKAs in patients with skeletal dysplasia, especially phocomelia, no treatment has been described yet. However, 60 years after thalidomide's introduction to the market, a rising number of patients may begin to need surgical treatment for OA of the knee.[3] In patients with phocomelia, severe shortening and/or loss of the proximal long bones while retaining parts of the distal structures is common.[4] The severe femoral deformity, inadequate soft tissue balancing, and tiny medialized patella in this patient required special considerations.

Preoperative evaluation of the bony dimensions is critical to ensure the appropriate fit of the keels and stems of the implants. If preoperative templating reveals particularly small bony anatomy, customized implants may need to be considered. Customized implants can be produced based on preoperative radiographs or computed tomography (CT) scans and are often used in patients with unusual anatomy.[1] In this case, the tiny anatomy of the femur was limiting the stem size. Femoral stems can help bypass distributing the increased stress of a constrained articular fixation.[5] The optimal stem length is achieving a mechanically sound construct while preserving as much native bone as possible.[6] However, in literature, an ideal length of femoral length is not described. In this case, a simple and cost-efficient customization of the femoral implant was performed by shortening the femoral stem during the operation.

To obtain adequate balancing and achieve satisfactory stability, we decided to use a constrained non-hinged prosthesis in this primary TKA; as in our view, sufficient stabilization could not be achieved by a non-constrained prosthesis and led to early revision surgery on the contralateral knee. A highly constrained hinge prosthesis should also be made available in cases of severe ligamentous compromise.

We faced a tiny medialized patella comparable to the small patella syndrome.[7] Because a medial retinacular release did not obtain satisfactory central patella tracking and outcomes are poor after patellectomy, a patella transfer was performed.[8] An intact patella displaces the tendon away from the femoral surface producing a mechanical advantage and therefore improving the quadriceps leverage. In our case, however, due to the small size of the patella, the restrain of the patellar complex for posterior shifting can be neglected. The use of patellar bone autograft has been described previously by Buechel[9] who used iliac crest for bone grafting and showed good results in the early stages. Lakshmanan and Wilson[10] and Tirveilliot et al.[11] proposed bone grafting of the patellar tendon using the tibial plateau obtained from the routine tibial cut. Transosseous sutures are proposed to fix the graft to the extensor system to avoid secondary migration but were not feasible in this case due to the tiny conditions and small bone stock.[11] It is important to note that the free patella transfer is a salvage procedure and should only be considered as a last resort if adequate centralization of the patella cannot otherwise be achieved. In the long-term patella, autografts can suffer from resorption and osteonecrosis.[12] However, alleged radiographic changes can also be a consequence of the patella conforming to the femoral articulation.

Conclusion

The unusual anatomy and severe deformities that accompany phocomelia require thoughtful implant selection and careful soft tissue balancing. Constrained and customized cemented implants should be considered for primary TKA in these patients. Patella transfer proved to be a simple and viable technique for balancing a tiny grossly medialized patella. Short-term results show promising clinical outcomes; however, future studies need to be performed for long-term results.

Authors' note

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Author contributions

H.F. and M.K. contributed to conceptualization; H.F., T.B., and M.O. participated in methodology; all authors validated the article; H.F., M.K., M.O., and H.M. performed formal analysis; H.F., M.K., and M.O. participated in writing—original draft preparation; all authors reviewed and edited the article; H.F. contributed to visualization; T.G., M.C., M.O., M.K., and H.M. participated in supervision; H.F. and M.K. participated in project administration. All authors have read and agreed to the final version of this article.

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

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