

Spontaneous regeneration of the large femoral defect in patient with diffuse osteomyelitis after intramedullary nailing

İntramedüller çivileme sonrasında yaygın osteomiyelit gelişen bir hastada, geniş femoral defektin spontan iyileşmesi

Esat Kıter, M.D.,¹ Semih Akkaya, M.D.,¹ Murat Oto, M.D.,¹ İzge Günal, M.D.²

¹Department of Orthopedics and Traumatology, Medicine Faculty of Pamukkale University, Denizli, Turkey; ²Department of Orthopedics and Traumatology, Medicine Faculty of Dokuz Eylül University, İzmir, Turkey

Infection of the long bones after intramedullary nailing is a troublesome condition and management of the infection remains challenging to orthopedic surgeons. Associated infection can be more problematic and more diffuse in intramedullary bone fixation, since it may spread along the nail. Surgical treatment choices are also difficult especially in cases with large bone defects after debridement. In this article, we present a 75-year-old woman that had been treated only with in-situ external fixation, antibiotic therapy and observation. Despite diffuse femoral osteomyelitis, a 10 cm femoral defect healed with unexpected bone regeneration which couldn't be explained reasonably.

Key words: Bone defect; femoral fracture; osteomyelitis; spontaneous bone regeneration.

İntramedüller çivileme sonrası uzun kemiklerde gelişen enfeksiyon sorunlu bir durumdur, ortopedi cerrahları için enfeksiyonun yönetimi ise diğer bir zorluktur. Eşlik eden enfeksiyon, inramedüller kemik tespitinde çivi nedeniyle daha sorunlu ve yaygın bir hale gelebilir. Özellikle büyük kemik defektleri olan olgularda debridman sonrası tedavi seçimi de zordur. Bu yazıda, sadece in-situ eksternal tespit, antibiyotik tedavisi ve gözlem ile tedavi edilen 75 yaşında kadın olgu sunuldu. Yaygın femoral osteomiyelite rağmen 10 cm'lik femoral defekt beklenmedik bir kemik rejenerasyonu ile mantıken açıklanamayacak şekilde iyileşti.

Anahtar sözcükler: Kemik defekti; femur kırığı; osteomiyelit; spontan kemik rejenerasyonu.

CASE REPORT

In March 2003, a 75-year-old female patient was admitted to our clinic with right femoral bowing and thigh pain. Six months ago, she had a femoral fracture because of trauma, and had been treated with locking intramedullar (IM) nail in another clinic. Five months after this operation she fell a second time and deformity and thigh pain developed again. The patient did not get any medical help for a month up to admission to our clinic. She had hypertension and diabetes mellitus history for over 10 years. Laboratory parameters, including C-reactive protein (CRP) level, erythrocyte

Treatment of massive bone defect is a complicated and annoying problem to solve for orthopedic surgeons. Many surgical treatment options exist but deciding to choose the right one is difficult. In the literature, several reports have been shown that until the late adolescent period, large bone defects could be repaired by the active, immature skeleton.^[1-3] But with additional risk factors such as advanced age or the presence of systemic disorders like infection and diabetes mellitus, the condition can be more problematic. We present such a case that had been treated with external fixation, antibiotic therapy and observation.

[•] Received: March 22, 2009 Accepted: October 13, 2009

Correspondence: Esat Kıter, M.D. Pamukkale Ünivesitesi Tıp Fakültesi Ortopedi ve Travmatoloji Anabilim Dalı, 20070 Kınıklı, Denizli, Turkey. Tel: +90 258 - 444 07 28 / 2154 Fax: +90 258 - 213 49 22 e-mail: ekiter@pau.edu.tr

sedimentation rate (ESR) and level of peripheral leukocytes (WBC; white blood cells) were 0.8, 52 and 10900 respectively.

Physical and radiologic examination revealed non-union and bowing of the IM nail as well (Figure 1a). A thorough plan was made for revision of the previous surgery according to possible peroperative situations. After exploring the fracture site, we observed non-union with clinical signs of infection and drainage. The intramedullary nail was removed after apical cutting with a high-speed burr. Infected bone and soft tissues were debrided. An approximately 10 cm defect resulted after debridement of sequestral bone. A temporary monolateral external fixator was applied. Appropriate antibiotics (ceftazidime and amikacin) were administered to the patient according to bacteria (Escherichia coli and Acinetobacter baumani) cultured from the bone and surrounding tissue taken after intraoperative debridement.

After the operation, an immediate drainage was seen at the incision site. Hemorrhagic leakage changed to purulent material after the 4th day. At the 10th day, the wound was debrided and irrigated with large amounts of saline. Leukocyte labeled bone scintigraphy revealed that infection had spread to the whole femur (Figure 2). Antibiotic therapy was changed to ciprofloxacin and rifampicin combination at the 4th week of the postoperative period, but infection did not regress. A fistulous drainage with mild signs of soft tissue inflammation was seen. The same infective agents were grown in the cultures of repeated drainages. Although all the surgical treatment options were



Figure 1. Filling the bone defect with unexplained bone regeneration. (Mo: months).

discussed with the patient after the 7th week, she did not accept any of them.

The patient had stayed in hospital for eight weeks, and after we noticed some new bone formation at the medial side of the defect and concurrently diminished drainage, she was discharged. In her postoperative 3rd month control, new bone formation at the medial side was observed in increasing amount (Figure 1b) and wound drainage had ceased. At the 4th month, antibiotic therapy was stopped and one month later, the distal pins were revised because of loosening. During pin revision, there was no motion at the fracture site with manual examination. Monthly radiographs showed that the bone defect was filling with unexpected bone regeneration (Figure 1c). The external fixator was removed at the 7th month. Partial weight bearing with a walker was permitted at the 10th month. At one-year followup, X-rays had revealed complete bony union in the whole femoral defect and the patient had no pain and no evidence of infection (Figure 1d) and full weight bearing was allowed (Figure 3). The patient



Figure 2. Technetium-99m hexamethyl propylene amine oxime leukocyte labeled bone scintigraphy.

had been followed for three years. For the last two years she was able to walk with full weight bearing, but she died because of cardiopulmonary problems in April of 2006.

DISCUSSION

In the literature, several studies have described spontaneous regeneration of bone in patients with large bone defects. In maxillofacial surgery, it is possible to find many papers about mandible regeneration after wide resection.^[4,5] On the other hand in orthopedic surgery there are specific case reports about spontaneous regeneration of long bones in children and adolescents. Bosworth et al.^[1] reported six patients treated with large tibial resection due to osteomyelitis. The authors observed bone regeneration in all cases and they concluded that although regeneration may take place after excision of the tibial shaft for massive osteomyelitis in childhood, regeneration could be incomplete after eight years of age. Other, relatively new reports had presented similar cases but the localizations were different and the reason for the defect was trauma. In these reports, regeneration of a large segment of bone in an eight-year-old girl with a proximal femoral defect^[2] and a 16-year-old boy with a distal humeral defect^[3] were documented.



Figure 3. Full weight bearing was allowed at the one year control. (arrow: affected, right side).

In both cases, the defects occurred after the trauma and healed without evidence of infection. In all these examples, the authors focused on two basic topics; the intact periosteum and immaturity of the skeleton. Intramembranous ossification caused by an intact periosteal sleeve seems to be the main mechanism of spontaneous regeneration in such cases.^[6,7] Klein et al.^[8] presented a 18-year-old case with a large, infected tibial defect after a gunshot wound. Authors have observed spontaneous healing after the soleus muscle flap and external fixator application.

On the other hand the induced membrane concept in the treatment of adult patients who suffered from large bone defects has gained popularity during the last decade.^[9,10] This new concept is based on two classical applications. In the first step, a methyl methacrylate spacer is inserted into the defect. This spacer is responsible from the formation of a pseudo-synovial membrane between cement, bone and soft tissue. Two months later, in the second step, cancellous autografts are placed into the defect after spacer removal.^[9] Pseudo-synovial membrane is considered as an induced membrane and it is believed to provide a good blood supply, and even some growth factors to the autograft.^[10]

In comparison to other reports, our 75-year-old patient was rather old at the onset of symptoms. In this age group a good periosteal reaction and bone healing is unexpected. To our knowledge, there are no other reports about such a late manifestation of iatrogenic osteomyelitis resolved by new bone formation.

In conclusion, the extraordinary morphologic feature in this case is the new bone formation in a 10 cm femoral defect with diffuse femoral osteomyelitis. Although the certain factors responsible for that result couldn't be defined at the first intervention and other surgical options in that 75-years-old patient could be argued, the main point of the current presentation might be accepted as sharing this case with dramatic improvement beyond the optimistic expectations related to modern medicine.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Spontaneous regeneration of the large femoral defect in patient with diffuse osteomyelitis after intramedullary nailing 181

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

- Bosworth DM, Liebler WA, Nastasi AA, Hamada K. Resection of the tibial shaft for osteomyelitis in children. A thirty-two-year follow-up study. J Bone Joint Surg [Am] 1966;48:1328-39.
- 2. Pazzaglia UE, Finardi E, Pedrotti L, Zatti G. Fracture with loss of the proximal femur in a child. A case report. Int Orthop 1991;15:143-4.
- 3. Ulkü O, Karatosun V. Regeneration of bone after loss of the distal half of the humerus. Case report with a 20-year follow-up. J Bone Joint Surg [Br] 1997;79:746-7.
- 4. Adekeye EO. Rapid bone regeneration subsequent to subtotal mandibulectomy. Report of an unusual case. Oral Surg Oral Med Oral Pathol 1977;44:521-6.
- 5. Ogunlewe MO, Akinwande JA, Ladeinde AL, Adeyemo WL. Spontaneous regeneration of whole

mandible after total mandibulectomy in a sickle cell patient. J Oral Maxillofac Surg 2006;64:981-4.

- 6. Song HR, Puri A, Lee JH, Park HB, Ra DK, Kim GS, et al. Spontaneous bone regeneration in surgically induced bone defects in young rabbits. J Pediatr Orthop B 2002;11:343-9.
- 7. Korkusuz F, Korkusuz P, Özkul A. Identification of substance P receptors on fibroblast-like cells derived from the periosteum: an in vitro cell culture study. Eklem Hastalik Cerrahisi 2006;17:94-100
- Klein DM, Caligiuri DA, Riina J, Katzman BM. Spontaneous healing of a massive tibial cortical defect. J Orthop Trauma 1997;11:133-5.
- 9. Pelissier P, Martin D, Baudet J, Lepreux S, Masquelet AC. Behaviour of cancellous bone graft placed in induced membranes. Br J Plast Surg 2002;55:596-8.
- Pelissier P, Masquelet AC, Bareille R, Pelissier SM, Amedee J. Induced membranes secrete growth factors including vascular and osteoinductive factors and could stimulate bone regeneration. J Orthop Res 2004;22:73-9.