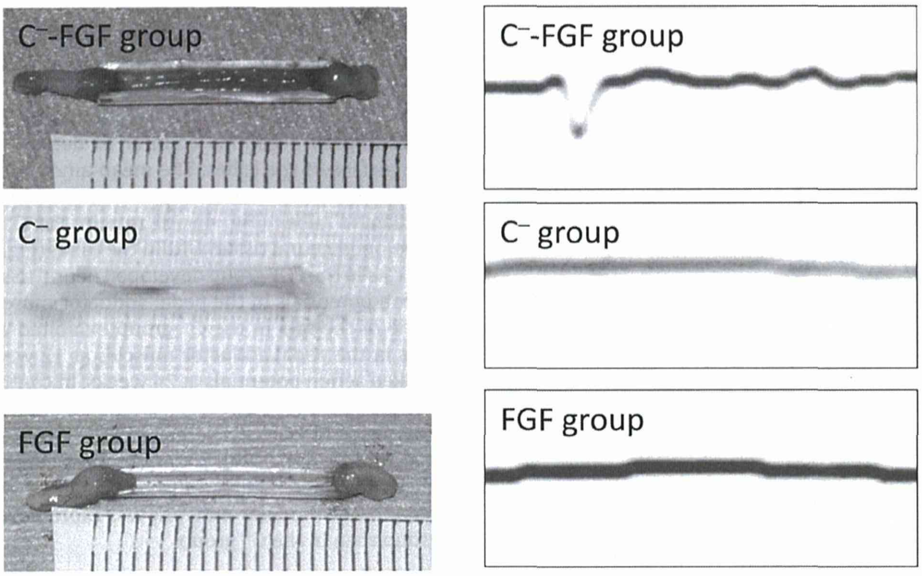

Fig. 5. Macroscopic appearance and electrophysiological study of three groups 12 weeks after tubulation. Regenerated neural structure were found in the C^--bFGF group and the C^- group. In the C^--bFGF group, evoked action potentials are found in the pedal adductor muscle at 12 weeks.

initial step of nerve regeneration in a tube follows an accumulation of exudates produced by nerve stumps in the inter-stump space of the tube [7]. These exudates contain nerve-promoting factors, such as laminin, fibronectin and nerve growth factor. Exudates produced by nerve stumps might have been stabilized in the tubes treated by C^- ions by the increased hydrophilia of the material, and this might have helped to form a 15-mm-long fibrin matrix in the exudates. The nerve-promoting factors in the exudates staying stable in the tube might have also promoted migration of neural cells through the fibrin matrix. Thus, nerve regeneration occurred over 15-mm in a rat sciatic nerve model using silicone rubber tubes implanted with C^- ions.

In Experiment 2, we observed an accelerated axonal regeneration in the C^--ion-implanted tubes treated with bFGF compared

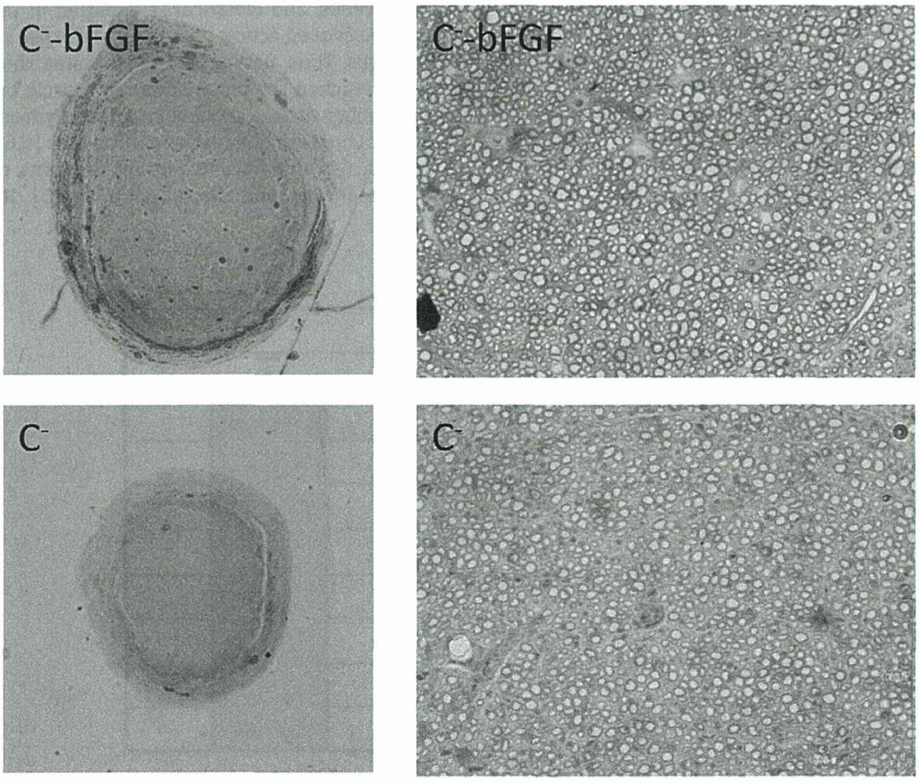

Fig. 6. Light microscopic picture of transverse section of regenerated nerves at 24 weeks. Myelinated axons were observed in all rats in the C^--bFGF group and the C^- group. In each section, vascular network was more developed in the C^--bFGF group than in the C^- group.

with the same tubes not treated with bFGF. In the C^--FGF group, nerves that regenerated through the tube extended and were functionally connected to the pedal adductor muscle by 12 weeks. This suggests that treating the tubes with bFGF significantly accelerated and improved the functional regeneration of myelinated axons. In tubulation, blood vessels and neural cells migrate from both proximal and distal nerve stumps, and the axons extend from the proximal nerve stump to form a neural structure [7]. bFGF is an angiogenic factor that stimulates vascular development across the inter-stump gap, which help promotes the spatial migration of all cellular elements [8]. We found more blood vessels in cross-sections of the regenerated nerve in the bFGF-pretreated tubes than in untreated tubes. This suggests that bFGF adhering to the chamber wall may remain bioactive and act as a promoter of angiogenesis during the process of tubulation. bFGF receptors have been demonstrated on the plasma membrane of regenerating axons, suggesting that bFGF might also work directly to promote axon regeneration [9]. Our model using bFGF attached inside the tube should enhance both vascularity and axon growth in the tube.

Although small amount of bFGF attached to the surface of untreated silicone rubber tubes in the FGF group, nerve axons did not regenerate over the 15-mm gap in the tube. The bFGF attached to the untreated silicone tube might have been swept away in the process of nerve regeneration in tubulation because the bFGF did not have a strong affinity for the surface of the untreated silicone tube. Thus, the cellular affinity and biocompatibility provided by the C^--ion-implanted silicone surface might also play an important role in nerve regeneration.

The surface implanted with C^- ions had a higher affinity for cells and bFGF than the silicone rubber surface alone [5,6]. These observations suggest that silicone rubber tubes implanted with C^- ions can serve as both a guide to nerve growth and as a provider of bFGF to stimulate peripheral nerve regeneration. The C^- ion-implantation technique might be used to create nerve guides to provide various neurochemical factors and cells to promote nerve regeneration.

5. Conclusions

Silicone tube treated with C^- ions showed increased hydrophilic properties and cellular affinity, and axon regeneration was promoted with this increased biocompatibility and this effect was enhanced by basic fibroblast growth factor.

References

[1] G. Lundborg, L. Dahlin, N. Danielsen, Ulnar nerve repair with the chamber technique, Scand. J. Plast. Reconstr. Surg. 25 (1990) 79–82.

[2] S.E. Mackinnon, A.L. Dellon, Clinical nerve reconstruction with a bioabsorbable polyglycolic acid tube, Plast. Reconstr. Surg. 85 (1990) 419–424.

[3] T. Glück, Über neoplastik auf dem wege der transplantation, Arch. Klin. Chir. 26 (1880) 606–616.

[4] G. Lundborg, L.B. Dahlin, N. Danielsen, H. Gelberman, F.M. Longo, H.C. Powell, Nerve regeneration in silicone chambers, influence of gap length and of distal stump component, Exp. Neurol. 76 (1982) 361–375.

[5] H. Sato, H. Tsuji, S. Ikeda, N. Ikemoto, J. Ishikawa, S. Nishimoto, Enhanced growth of human vascular endothelial cells on negative ion implanted hydrophobic surfaces, J. Biomed. Mater. Res. 14 (1999) 22–30.

[6] H. Tsuji, H. Sato, S. Ikeda, N. Ikemoto, Y. Gotoh, J. Ishikawa, Surface modification by silver-negative-ion implantation for controlling cell-adhesion properties of polystyrene, Surf. Coat. Technol. 103/104 (1998) 124–128.

[7] Q. Zhao, L.B. Dahlin, M. Kanje, G. Lundborg, Repair of the transected rat sciatic nerve: matrix formation within silicone tubes, Restor. Neurol. Neurosci. 5 (1993) 197–204.

[8] N. Danielsen, B. Pettmann, H.L. Vahlsing, M. Manthrope, S. Varon, Fibroblast growth factor effects on peripheral nerve regeneration in a silicone chamber model, J. Neurosci. Res. 20 (1988) 320–330.

[9] E. Fujimoto, A. Mizoguchi, K. Hanada, M. Yajima, C. Ide, Basic fibroblast growth factor promotes extension of regenerating axons of peripheral nerve. In vivo experiments using a Schwann cell basal lamina tube model, J. Neurocytol. 26 (1997) 511–528.

Spine

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CASE REPORT

Spontaneous Reduction in Ossification of the Posterior Longitudinal Ligament of the Thoracic Spine After Posterior Spinal Fusion Without Decompression

A Case Report

Hiroaki Kimura, MD, PhD, Shunsuke Fujibayashi, MD, PhD, Mitsuru Takemoto, MD, PhD, Bungo Otsuki, MD, PhD, and Shuichi Matsuda, MD, PhD

Study Design. Case report.
Objective. We report on a patient with thoracic myelopathy caused by ossification of the posterior longitudinal ligament (OPLL) of the spine.
Summary of Background Data. Surgery for thoracic OPLL (T-OPLL) is associated with a high rate of complications. Posterior spinal instrumentation (PSI) with decompression is a relatively safe procedure with satisfactory results. However, the mechanisms by which PSI with decompression produces neurological recovery in patients who undergo T-OPLL have not been elucidated fully, and no reports have described the radiographical progress after PSI.
Methods. The patient was a 78-year-old female with a 4-month history of gait disturbance and paresthesia of the lower extremities who had continuous OPLL at T1–T4. The spinal cord was impinged by beak-type OPLL with a nonossified area at T3–T4. PSI without decompression of C7–T6 was performed.
Results. After surgery, the patient showed clear improvement in neurological symptoms. She recovered fully after 2 years, by which time, the point of the beak-type OPLL at T3–T4 had become a mild protuberance and the nonossified area was fused through the remodeling process. This reduced the size of the ossified lesion that had caused severe compression of the spinal cord before surgery.
Conclusion. The remaining mobility at the nonossified lesion, where the point of the beak-type OPLL compressed the spinal cord, might have caused the myelopathy in this patient. We propose that stabilization with PSI stimulates bone remodeling of nonossified lesions, leading to a reduction of OPLL. This finding provides a clue about the mechanisms responsible for neurological improvement after PSI for T-OPLL.
Key words: thoracic, posterior longitudinal ligament, OPLL, posterior spinal instrumentation, spinal fusion, remodeling, ossification, myelopathy, decompression, beak-type.
Level of Evidence: N/A
Spine 2014;39:E417–E419

Decompressive laminectomy for ossification of the posterior longitudinal ligament of the thoracic spine (T-OPLL) is not effective and leads to exacerbation in some cases. Therefore, spinal instrumentation is recommended for T-OPLL. Among various procedures, posterior spinal instrumentation (PSI) with decompression is less technically demanding, is associated with a lower risk of neurological complications, and produces favorable surgical results.[1–3] Spinal instrumentation is effective for treating T-OPLL, whereas the effect of decompression with PSI is unclear because the backward shift of the spinal cord is restricted because of physiological kyphosis in the thoracic spine.[1–4] We report on a patient with thoracic myelopathy caused by beak-type T-OPLL, in which the symptoms improved markedly after PSI without decompression. Computed tomography (CT) revealed that the size of the ossified lesion was reduced through remodeling. This is the first report on the radiographical progress of T-OPLL after PSI and provides information about the efficacy of instrumented fusion for T-OPLL and a clue about the mechanism responsible for the neurological improvement.

CASE REPORT

A 78-year-old female with T-OPLL presented with a 4-month history of gait disturbance. On admission, she had paresthesia of the lower extremities and was unable to walk without a

From the Department of Orthopedic Surgery, Graduate School of Medicine, Kyoto University, Kyoto, Japan.

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Address correspondence and reprint requests to Hiroaki Kimura, MD, PhD, Department of Orthopedic Surgery, Graduate School of Medicine, Kyoto University, 54 Shogoin Kawahara-cho, Sakyo-ku, Kyoto 606-8507, Japan; E-mail: hkimura@kuhp.kyoto-u.ac.jp

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cane. Muscle strength in the lower extremities was normal, but she had a distal sensory loss bilaterally below the inguinal region. The heel-knee test result was abnormal in the left extremity. Bladder function was disturbed slightly. A CT scan showed OPLL at T1–T4, including beak-type OPLL with a nonossified area (NOA) at T3–T4. T2-weighted magnetic resonance (MR) images showed impingement of the spinal cord anteriorly by beak-type OPLL at T3–T4 with abnormal intensity in the spinal cord, although the subarachnoid space posterior to the spinal cord was detectable (Figure 1).

We planned a 2-step surgery. The first step was PSI without decompression because the subarachnoid space posterior to the spinal cord was maintained. The second step was OPLL extirpation *via* a thoracotomy if neurological recovery was insufficient after the first step.

We performed C7–T6 PSI without decompression and with correction of kyphosis using pedicle screws (Figure 2). We inserted a short pedicle screw into the left side of T4 as a landmark for the width for drilling the vertebral body in the second-step surgery. Posterior bone grafting using local bone harvested from spinous processes was performed between the laminae. Immediately after the surgery, the patient showed improvement in neurological symptoms, and she was able to walk without a cane 6 weeks after surgery. The Japanese Orthopedic Association score (full score = 11 points) improved markedly from 6.5 points preoperatively to 11 points 2 years after surgery, and her recovery was noted as 100%. MR images and CT scans 2 years after surgery showed that the NOA at T3–T4 was fused and the point of the ossified mass had become a mild protuberance (Figure 3). Axial CT scans of T3–T4 showed that the heterogeneous ossified mass comprising both low- and high-intensity areas with a rough surface preoperatively had become a homogeneous mass comprising a high-intensity area with a smooth surface at 2 years after surgery. The occupying ratio at T3–T4 had decreased slightly from 39.7% preoperatively to 33.9% at 2 years after surgery.

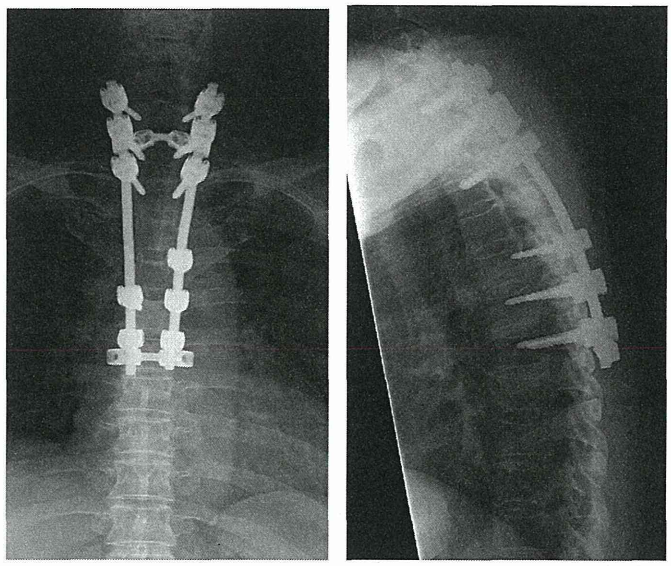

Figure 2. Anteroposterior view (left) and lateral view (right) of radiographs 2 weeks after posterior spinal instrumentation without decompression (C7–T6).

DISCUSSION

In this case, the point of the beak-type OPLL at T3–T4 impinged to the spinal cord, causing thoracic myelopathy. We performed PSI without decompression as the first-step surgery for several reasons. First, decompression did not seem to be effective in the present case, in which the subarachnoid space posterior to the spinal cord was maintained. The backward shift of the spinal cord was not expected because

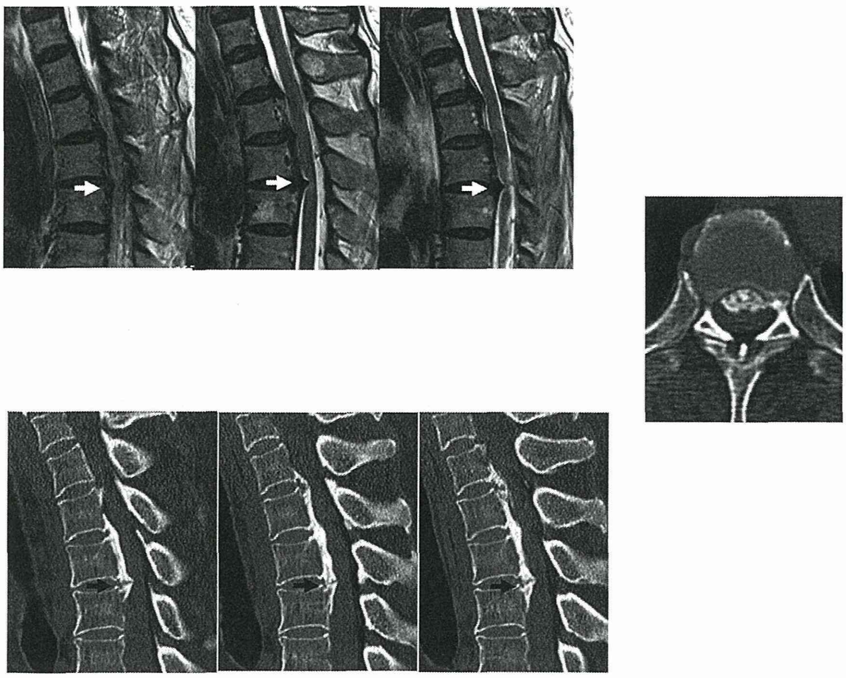

Figure 1. Preoperative radiographical images of a 78-year-old female with thoracic myelopathy. Sagittal views of the T2-weighted magnetic resonance image (left upper) show that the spinal cord was compressed sharply by the beak-type OPLL (white arrows). Sagittal views on CT (left lower) show the beak-type OPLL with a nonossified area at T3–T4 (black arrows). The axial view on CT scan at T3–T4 (right) shows the heterogeneous ossified mass with a rough surface. OPLL indicates ossification of the posterior longitudinal ligament; CT, computed tomography.

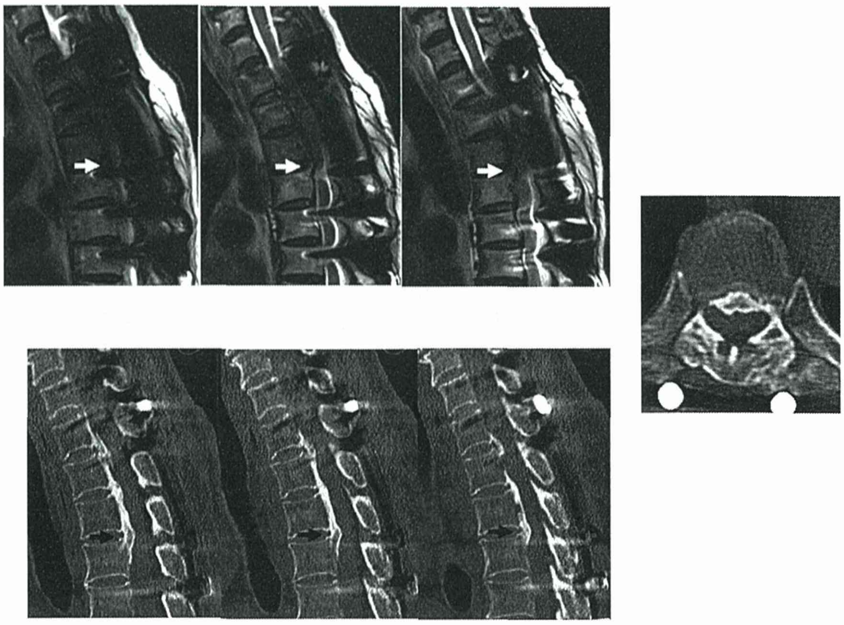

Figure 3. Radiographical images 2 years after surgery. Sagittal views of the T2-weighted magnetic resonance images (left upper) and CT images (left lower) show that the nonossified area at T3–T4 was fused and that the point of the ossified mass had become a mild protuberance (white and black arrows). The axial view at T3–T4 on CT scan (right) shows that the heterogeneous ossified mass observed preoperatively had changed into a homogeneous mass comprising a high-intensity area with a smooth surface. CT indicates computed tomography.

of physiological kyphosis in the thoracic spine.[1–4] Second, the mobility in the NOA associated with OPLL was considered to be related to the development of myelopathy. Miyashita *et al*[5] reported that the clinical results of posterior stabilization without decompression for thoracolumbar burst fractures with neurological deficits were comparable with those of posterior stabilization with decompression, suggesting that stabilization without decompression may improve myelopathy related to spinal instability. Third, Matsuyama *et al*[4] demonstrated that the spinal cord can become impaired during the laminectomy using intraoperative spinal cord monitoring of compound muscle action potentials. Their study indicated that the instability of the spinal column and the slight alteration in spinal alignment caused by a laminectomy can cause neurological deterioration intraoperatively because the spinal cord is fragile at the site. PSI without decompression decreases the risk of intraoperative neurological deterioration during laminectomy. Fourth, PSI without decompression is less invasive and more favorable for achieving fusion than is PSI with decompression because a laminectomy decreases spinal stability and the size of the bed for bone grafts.[5]

In our patient, the neurological recovery after PSI without decompression was excellent. Surprisingly, the ossified lesion decreased in size with NOA fusion, and the point of the ossified lesion had become a mild protuberance 2 years after surgery. Yamazaki *et al*[3] reported that some mobility remains in the thoracic spine despite the presence of OPLL. Thus, the mobility remaining at T3–T4 might have been related to the development of myelopathy in the present case. An NOA associated with OPLL may be similar to pseudarthrosis, a failure of bone healing, which comprises fibrous, fibrocartilaginous, and bony tissues. Stabilization using instrumentation may stimulate normal bone remodeling at the NOA, and the area may then be replaced by mature bone tissue, resulting in spontaneous reduction of the OPLL. This is the first report to show that an NOA at a T-OPLL became fused and reduced, and that the shape changed from beak type to flat type after PSI. This demonstrates the efficacy of instrumented fusion for T-OPLL.

➢ Key Points

- ❑ Thoracic myelopathy caused by beak-type OPLL, which impinged the spinal cord, improved markedly after PSI without decompression.
- ❑ The remaining mobility at the nonossified lesion of the beak-type OPLL correlated with the development of myelopathy in this case.
- ❑ CT scans demonstrated that the nonossified lesion of OPLL was fused and that the beak-type OPLL became a flat-type OPLL with some reduction in ossified mass, 2 years after surgery.

References

1. Matsumoto M, Chiba K, Toyama Y, et al. Surgical results and related factors for ossification of posterior longitudinal ligament of the thoracic spine: a multi-institutional retrospective study. *Spine (Phila Pa 1976)* 2008;33:1034–41.
2. Matsumoto M, Toyama Y, Chikuda H, et al. Outcomes of fusion surgery for ossification of the posterior longitudinal ligament of the thoracic spine: a multicenter retrospective survey: clinical article. *J Neurosurg Spine* 2011;15:380–5.
3. Yamazaki M, Mochizuki M, Ikeda Y, et al. Clinical results of surgery for thoracic myelopathy caused by ossification of the posterior longitudinal ligament: operative indication of posterior decompression with instrumented fusion. *Spine (Phila Pa 1976)* 2006;31:1452–60.
4. Matsuyama Y, Sakai Y, Katayama Y, et al. Indirect posterior decompression with corrective fusion for ossification of the posterior longitudinal ligament of the thoracic spine: is it possible to predict the surgical results? *Eur Spine J* 2009;18:943–8.
5. Miyashita T, Ataka H, Tanno T. Clinical results of posterior stabilization without decompression for thoracolumbar burst fractures: is decompression necessary? *Neurosurg Rev* 2012;35:447–54; discussion 54–5.

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Reconstruction of active elbow flexion in patients with radial ray deficiency: report of two cases

Souichi Ohta, MD, PhD*, Ryosuke Kakinoki, MD, PhD, Takashi Noguchi, MD, Yukitoshi Kaizawa, MD, Syuichi Matsuda, MD, PhD

Department of Orthopaedic Surgery, Kyoto University, Kyoto, Japan

Radial ray deficiency is a complex congenital abnormality of the radial side of the upper extremity. Stiffness of the elbow in extension is frequently observed in association with a radial clubhand involving radial angulation and displacement of the carpus with respect to the distal ulna.[7] In such cases, corrective surgery on the carpus should not be attempted in the absence of an adequate range of elbow flexion because insufficient elbow flexion precludes bringing the hand to the face and feeding oneself. Active elbow flexion has been reported to improve spontaneously after the first year or two of life, although less than 90° of flexion is finally obtained in many patients.[6] In some patients, the elbow remained stiff in extension or gained flexion to only 40° or less.[7] The insufficient elbow flexion might be one of the causes of gradual bowing of the ulna or recurrence of radial clubhand deformity after wrist corrective surgery. To date, there have been only a few case reports of reconstruction of active elbow flexion with a good outcome, which was achieved by triceps transfer.[7,9] However, the triceps is important as an antagonist of flexion of the elbow joint and should therefore be preserved as much as possible. Here, we report two cases of radial clubhand without active elbow flexion in which transposition of the rudimentary insertion of the biceps to the ulna resulted in a good outcome.

Parent/guardian of each patient gave their agreement to the publication of the cases discussed herein.

*Reprint requests: Souichi Ohta, MD, PhD, Department of Orthopaedic Surgery, Kyoto University, 54 Kawahara-cho, Shogoin, Sakyo-ku, Kyoto City, Kyoto 606-8507, Japan.

E-mail address: sota@kuhp.kyoto-u.ac.jp (S. Ohta).

Case reports

Case 1

A boy was born with a Bayne type 4 radial clubhand associated with a Blauth type 5 hypoplastic thumb in the right upper extremity and a Bayne type 3 radial clubhand associated with a Blauth type 5 hypoplastic thumb in the left upper extremity (Fig. 1). When the patient was 10 months old, he underwent the right wrist centralization procedure and transposition of the rudimentary insertion of the ipsilateral biceps to the ulna.

Although the wrist was splinted as straight as possible before surgery, the passive range of motion (ROM) of the right elbow joint was 0° to 110° and the active ROM was 0° to 20°. The passive pronation/supination was 90°/90°. There was severe radial deviation of the right hand.

An anterior approach was used to expose the distal part of the biceps in the elbow joint. The thin fascia-like insertion of the biceps was fused with the fascia in the proximal radial forearm. A very thin fibrous band was found between the ulnar side of the distal biceps and the fascia in the proximal ulnar forearm and considered to represent a rudimentary lacertus fibrosus.

After separation of the accompanying artery from the biceps, the thin fascia-like insertion of the biceps was detached from the radial side of the forearm and held with 2-0 Ti-Cron suture (Covidien, Dublin, Ireland). Two drill holes (0.7 mm in diameter) were made anteroposteriorly a few millimeters distal to the coronoid process of the ulna.

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Figure 1 (**A**) The preoperative appearance of the right upper extremity in case 1 (patient with severe radial clubhand deformity). (**B**) Radiograph of the upper arm before surgery. The radius and thumb were completely absent.

Then, the Ti-Cron suture holding the distal stump of the biceps was passed through the holes and secured in the bone under tension with the elbow in 45° of flexion. Postoperatively, a posterior splint was applied for 3 weeks, after which active flexion of the elbow joint was allowed.

Ten years postoperatively, the active ROM of the right elbow joint was 0° to 135°. Active supination of the forearm was 55° and active pronation was −20° (Fig. 2). Manual muscle testing of elbow flexion resulted in a score of grade 5.

Case 2

A girl was born with radioulnar synostosis associated with a Blauth type 5 hypoplastic thumb in the left upper extremity and a Bayne type 1 radial clubhand associated with a Blauth type 2 hypoplastic thumb in the right upper extremity (Fig. 3). When the patient was 15 months old, she underwent surgical closure of a ventricular septal defect and annuloplasty for treatment of tricuspid insufficiency. She was thought to have Holt-Oram syndrome but did not undergo genetic analysis of *TBX5*. When the patient was 22 months old, she underwent left index finger pollicization and transposition of the rudimentary insertion of the ipsilateral biceps to the ulna. Before surgery, the passive ROM of the left elbow joint was 0° to 90°, with no active flexion. The forearm was fixed in 60° of pronation without active or passive supination.

An anterior approach was used to expose the distal part of the biceps in the elbow joint. The thin fascia-like insertion of the biceps was fused with the fascia in the proximal radial forearm. The lacertus fibrosus could not be identified. The brachial artery divided into the median and ulnar arteries, but the radial artery was not found.

After separation of the accompanying artery from the biceps, the thin fascia-like insertion of the biceps was detached from the radial side of the forearm and held with two Ethibond sutures (Ethicon, Inc, Somerville, NJ, USA) (Fig. 4). Two drill holes (1.5 mm in diameter) were made anteroposteriorly a few millimeters distal to the coronoid process of the ulna. Then, the Ethibond suture holding the distal stump of the biceps was passed through the holes and secured in the bone under tension with the elbow in 90° of flexion. Postoperatively, a posterior splint was applied for 3 weeks, and then active flexion of the elbow joint was allowed.

Three years postoperatively, the active ROM of the left elbow joint was 0° to 80° (Fig. 5). The forearm was fixed in 60° of pronation. Manual muscle testing of elbow flexion resulted in a score of grade 5.

Discussion

In radial ray deficiency, most of the elbow flexors are abnormal. The long head of the biceps is almost always absent.[2] If it is present in conjunction with total aplasia of the radius, it inserts abnormally into the lacertus fibrosus. The short head is generally present but is usually fused with the coracobrachialis, brachialis, or forearm flexor muscles. The brachialis may be present but is often fused with the biceps at its origin and becomes continuous with the common flexor muscles without any specific insertion. The brachioradialis is usually absent in patients with total aplasia of the radius. If it is present, the brachioradialis can be rudimentary or fused with the extensor muscles and insert aberrantly on the carpus or ulna. Despite all this abnormality of the muscles around the elbow joint, Smith

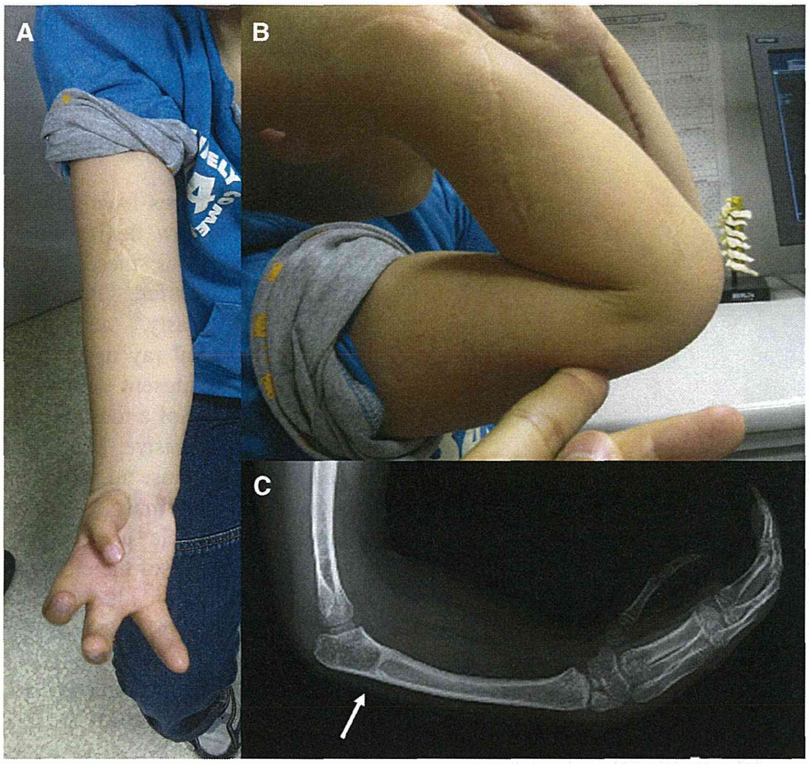

Figure 2 Case 1. (**A**) The patient's elbow extension 10 years postoperatively. (**B**) The elbow flexion. The elbow actively flexed to 135°. (**C**) Radiograph of the elbow joint 10 years postoperatively. The site of osteosclerosis was located distal to the coronoid process of the ulna. This is the remnant of the drill holes used for the attachment of the biceps insertion to the ulna with suture.

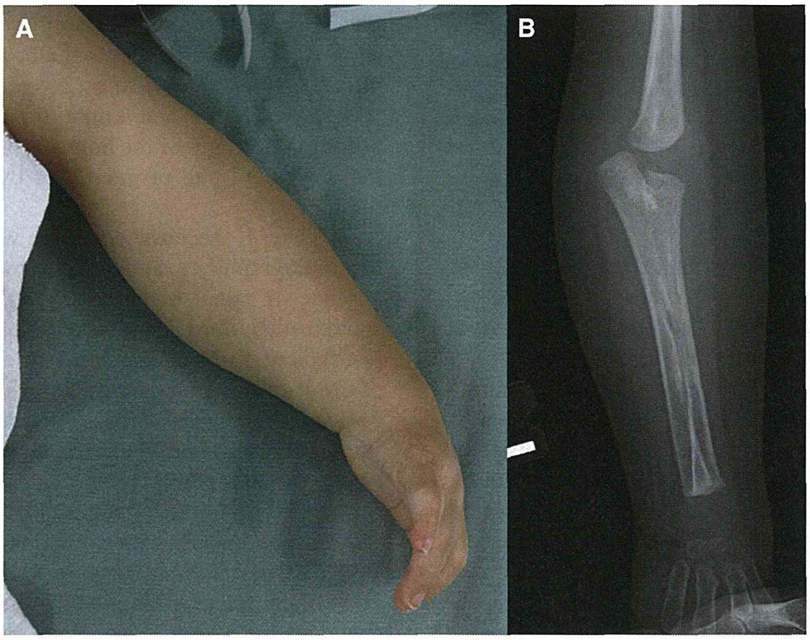

Figure 3 (**A**) The preoperative appearance of the left upper extremity in case 2. No crease was found in the antecubital fossa. (**B**) The preoperative radiograph. Radioulnar synostosis and absence of the thumb were observed.

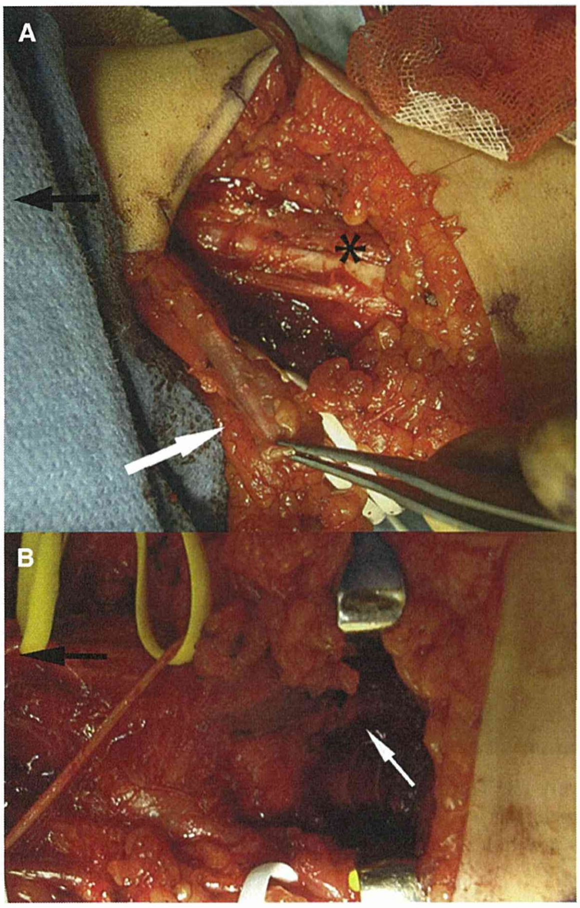

Figure 4 Case 2, intraoperative photographs. (**A**) Just after the separation of the rudimentary insertion of the biceps (white arrow) from the fascia of the common flexor muscles (asterisk). The black arrow indicates the proximal side. (**B**) The new insertion of the biceps onto the ulna (white arrow). The black arrow indicates the proximal side.

reported never observing a patient fail to regain at least 90° of elbow flexion after wrist centralization, even when the elbow was stiff preoperatively.[10] The brachialis may act as the primary flexor of the elbow joint in such patients.

The brachialis is a monarticular muscle and is considered to act as an antigravity and skill-related muscle, whereas the biceps is a biarticular muscle and is considered to act as a propulsive muscle.[8] The propulsive muscle produces gross power and accelerates reaching movements. In normal active elbow flexion, the bulging of the contracted brachialis muscle belly pushes the belly and tendon of the biceps muscle forward. This increases the biceps lever arm and thus the flexion power of the elbow. Although the biceps might be unnecessary for active flexion of the elbow joint, it would remain necessary for power flexion against resistance or maintenance of elbow flexion. In our cases, the thin fascia-like insertion of the biceps fused with the fascia in the proximal radial forearm, leading to inefficient transmission of the traction force produced by the biceps to the forearm. Direct suturing of the rudimentary insertion of the biceps to the ulnar cortex was therefore desirable to allow efficient transmission of the biceps traction force and thus improvement in the activities of daily life.

Several procedures have been reported for reconstruction of active elbow flexion in patients with congenital elbow malformations; these include triceps transfer, pectoralis major transfer,[1] latissimus dorsi transfer,[4] the Steindler flexorplasty,[5] and free muscle transfer.[3] In patients with radial ray deficiency, the short head of the biceps is usually present without a secure distal insertion. Therefore, transfer of a muscle from outside the upper arm is unnecessarily invasive. The Steindler flexorplasty would be inadequate because of the weakness of the wrist extensors. The triceps muscle should generally be preserved as much as possible because of its importance as an antagonist of flexion of the elbow joint, but triceps transfer could be used as an alternative in cases in which the insertion of the short head of the biceps is completely absent.

We secured the rudimentary insertion of the biceps to the ulna under tension with the elbow in 45° of flexion in case 1 and 90° of flexion in case 2. The optimal elbow position and degree of tension with which to secure the biceps remain unclear. In case 1, the maximum active elbow flexion was 90° after 1 year and had improved to 135° 10 years postoperatively. In case 2, because sufficient length of the rudimentary insertion could not be obtained, we secured the insertion to the ulna with 90° of flexion. The maximum active elbow extension was −40° after 2 months and 0° after 2 years postoperatively. The secure distal insertion might force the biceps itself to adapt to the length of the humerus as the patient grows, by the downward pull of gravity or by the traction force of the triceps.

In case 1, because complete absence of the radius on radiographic examination obviously indicated the absence of radial biceps insertion, we were afraid that spontaneous improvement of elbow flexion would be insufficient to bring the hand to the face. Then, transposition of the rudimentary insertion of the biceps was decided to be performed with the wrist centralization surgery and resulted in excellent active ROM and flexion power of the elbow joint. This transposition surgery should be performed on patients with radial ray deficiency who cannot gain sufficient active elbow flexion after the wrist corrective surgery. However, especially in cases of complete absence of the radius (Bayne type 4), it would be an option to combine the transposition of the rudimentary insertion of the biceps with the wrist corrective surgery.

We did not perform any other imaging examinations except radiography before the elbow surgery. Although the short head of the biceps was reported to be generally

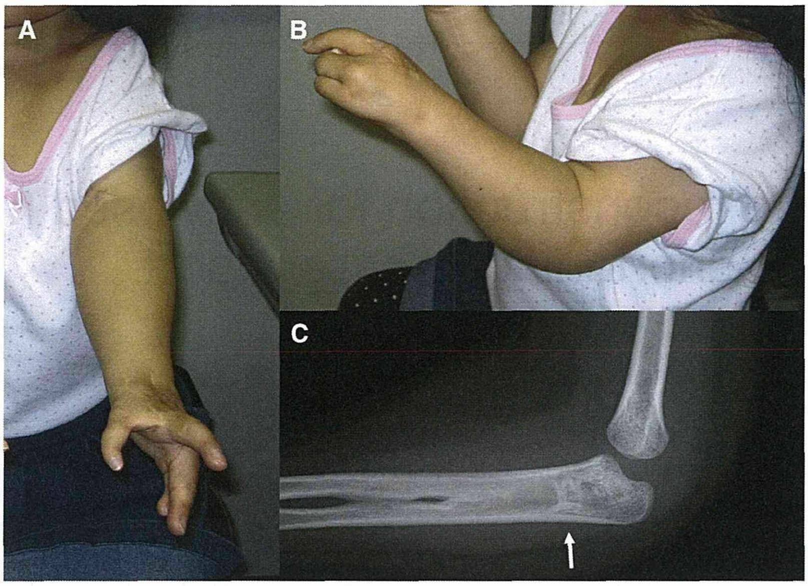

Figure 5 Case 2. (**A**) Elbow extension 3 years postoperatively. (**B**) Elbow flexion. The elbow actively flexed to 80°. (**C**) Radiograph showing the site of osteosclerosis distal to the coronoid process of the ulna (arrow).

present,[2] it might be better to perform a noninvasive imaging examination, such as magnetic resonance imaging or ultrasound, to define the presence of the biceps itself and its insertion. However, it would be difficult to find a thin fascia-like insertion of the biceps by magnetic resonance imaging or ultrasound.

Conclusions

Transposition of the rudimentary insertion of the biceps is an option for reconstruction of active elbow flexion in patients with radial ray deficiency without active elbow flexion.

Disclaimer

The authors, their immediate families, and any research foundation with which they are affiliated have not received any financial payments or other benefits from any commercial entity related to the subject of this article.

References

1. Atkins RM, Bell MJ, Sharrard WJ. Pectoralis major transfer for paralysis of elbow flexion in children. J Bone Joint Surg Br 1985;67:640-4.
2. D'Arcangelo M, Gupta A, Scheker LR. Radial club hand. In: Gupta A, Kay SPJ, Scheker LR, editors. The growing hand: diagnosis and management of the upper extremity in children. London: Mosby; 2000. p. 147-70.
3. Doi K, Arakawa Y, Hattori Y, Baliarsing AS. Restoration of elbow flexion with functioning free muscle transfer in arthrogryposis: a report of two cases. J Bone Joint Surg Am 2011;93:e105. http://dx.doi.org/10.2106/JBJS.J.01846
4. Gagnon E, Fogelson N, Seyfer AE. Use of the latissimus dorsi muscle to restore elbow flexion in arthrogryposis. Plast Reconstr Surg 2000;106:1582-5.
5. Goldfarb CA, Burke MS, Strecker WB, Manske PR. The Steindler flexorplasty for the arthrogrypotic elbow. J Hand Surg Am 2004;29:462-9. http://dx.doi.org/10.1016/j.jhsa.2003.12.011
6. Lamb DW. The treatment of radial club hand. Absent radius, aplasia of the radius, hypoplasia of the radius, radial paraxial hemimelia. Hand 1972;4:22-30.
7. Lamb DW. Radial club hand. A continuing study of sixty-eight patients with one hundred and seventeen club hands. J Bone Joint Surg Am 1977;59:1-13.
8. Matsuo T. Cerebral palsy: orthopaedic selective spasticity-control surgery. Tokyo: Soufusha; 2001.
9. Menelaus MB. Radial club hand with absence of the biceps muscle treated by centralisation of the ulna and triceps transfer. Report of two cases. J Bone Joint Surg Br 1976;58:488-91.
10. Smith PJ, Smith GD. Radial club hand. In: Hovius S, editor. The pediatric upper limb. London: Martin Dunitz; 2002. p. 153-82.