Femoral head avascular necrosis is a condition in which part of the femoral head undergoes necrosis due to decreased blood flow. The femoral head gradually disintegrates causing pain and even today, there are no effective rehabilitation methods other than symptomatic treatment such as decreasing the load on the hip joints with the use of a cane or walker. We herein describe our insights into this condition based on our experience with a case of femoral head avascular necrosis caused by steroid use in which KAATSU training was found to be highly effective. The patient was a 34-year-old woman (154 cm tall and weighing 50 kg, a radiologist). Since the age of 23, this patient had been receiving steroid treatment to control her refractory asthma. She later developed pain in her right hip, and gradually suffered hip joint deformation, restricted range of motion, and difficulty in walking. MRI revealed Association Research Circulation Osseous (ARCO) stage IV disease. She suffered marked pain of the right hip joint every time she walked, occasionally falling and required a cane to walk. At the patient’s own request, she received KAATSU training including KAATSU walking over a period of 3 months (total 28 sessions). Various assessments were carried out before and after training to determine the effects of KAATSU training. QOL was determined by SF–36v2, and marked improvement of role physical, body pain, general health, vitality, and social functioning were noted. Before training, the Japanese Orthopaedic Association (JOA) hip scores for pain were 10 (right), 40 (left), walking ability 16 (right), 20 (left), while 3 months after training, these scores were markedly improved in the affected side. Furthermore, not only did muscle strength on the affected side show marked improvement, but the MRI also revealed a tendency for improvement of the right femoral head avascular necrosis. DEXA showed signs of a clear increase in bone mineral density. Based on the above, these results suggest that KAATSU training is extremely useful as a rehabilitation method in patients with femoral head avascular necrosis. But, further larger scale investigations should be carried out in the future to support our findings.

**Key words:** Femoral head avascular necrosis, KAATSU training, Bone density, Pain, Rehabilitation
In addition, KAATSU training with restricted blood flow causes decreases in the intramuscular oxygen partial pressure during exercise and hypoxemia. With active refilling of vasculature in response to belt release, reactive hyperemia develops and vascular endothelial growth factor (VEGF) and nitric oxide (NO) production are enhanced (Takano et al., 2005; Horiuchi and Okita, 2012) and this may lead to improvements in both endothelial function and circulation. In this way, KAATSU training appears to be effective as a rehabilitation method in patients with femoral head avascular necrosis, but there are few reports regarding the effects of KAATSU training in these patients until now.

We experienced a case of femoral head avascular necrosis in one patient and would like to report our findings.

1. Case introduction

Case report: A 34-year-old woman (height 154 cm, weight 50 kg, profession radiologist)

Diagnosis: femoral head avascular necrosis
Chief complaint: pain of the hip joint during ambulation, gait disturbance.
Family history: no notable findings.

History of present illness: bronchial asthma with an onset during youth. Since experiencing a severe attack of aspirin-induced asthma at age 15, the patient became dependent on steroids and repeatedly required hospitalization because of poor control and had continued to take a large dose of steroids. As of age 23, HOT was introduced, and since age 25, as soon as her clinical rotations started, pain in the right hip joint began to manifest. Thereafter, deformation of the right hip joint progressed until age 33, and she developed femoral head avascular necrosis (OA). Gradually, the hip joint pain worsened with the limitations in her range of motion (ROM), and abduction external rotation contracture appeared. The patient suffered extreme pain in the right pelvis during walking and in order to reduce the joint load during walking had been using a cane. She presented at our hospital hoping to receive exercise treatment with KAATSU training. The patient had been taking prednisolone 3 mg/day, theophylline, montelukast sodium, and salmeterol/fluticasone.

This research was approved by the Institutional Review Board at the University of Tokyo and performed after receiving patient consent.

2. KAATSU training protocol

A KAATSU belt is wrapped around the base of the thigh and a KAATSU Master device (KAATSU Japan, Co., Ltd) was used. KAATSU training was provided for a total of 28 sessions over 3 months. Base pressure was 45 – 50 SKU. The optimal pressure started at 100 – 140 SKU and while repeatedly pressurizing and depressurizing, it gradually increased in 20 SKU increments, until an optimal pressure was reached. The training menu was as follows. 1) 3 point set: Toe curls, ankle dorsiflexion, and ankle plantar flexion. 2) KAATSU walk: gradually increased distance to approximately 150 to 300m during each session until a comfortable walking speed was achieved. 3) Non-KAATSU walk: belts were removed and the patient was allowed to walk for 75m to check for symptoms. In late stage intervention, calf-raises and squats were added, and the patient extended her comfortable walking distance to approximately 300m. Calf raises and squats were performed standing and bearing one’s own weight. The KAATSU side in the early stage of intervention was the affected leg only, starting with a KAATSU pressure of 300-320 SKU. In the latter stages, in addition to the affected right thigh, the healthy left thigh (160-300 SKU) exercise therapy under KAATSU condition was also conducted.

3. Evaluation

The following evaluations were conducted before and after training.
1) Magnetic resonance imaging (MRI)
The effect of training on life functioning was investigated using the SF-36v2 evaluation survey. Role physical, body pain, general health, vitality, social functioning, role emotional, and mental health were assessed.
3) Muscle strength evaluation using Cybex Measurements were taken with the subject seated in a chair with the trunk and thighs immobilized. Exercise involved bending the knee joint 90°, from a flexed position to extension and flexion. Bilateral lower limb muscle strength (isomericknee extension strength: knee joint 75°, isokinetic knee extension and flexion muscle strength: 30°/sec, 90°/sec, 180°/sec) was determined.
4) Functional analysis of the hip joint by Japanese Orthopaedic Association (JOA) hip scores as shown in Table 1 (Takatori et al., 2010)
5) Dual energy x-ray absorbed absorptiometry (DEXA) bone mineral density measurement device.

4. Results and clinical course

MRI findings before training are shown in Fig. 1A (left). T1 and T2 weighted images of the right femoral head weight-bearing areas revealed areas of low intensity signals. The surrounding areas showed high signals in the T2 weighted image, suggesting bone necrosis and an edematous change of the surrounding marrow. Her right hip joint showed joint space narrowing and formation of bone spurs, while the femoral head showed signs of flattening leading to
Table 1. JOA scores for the hip joint (From Takatori et al., 2010)

<table>
<thead>
<tr>
<th>I. Pain</th>
<th></th>
<th></th>
<th></th>
<th>point</th>
</tr>
</thead>
<tbody>
<tr>
<td>No complaints regarding the hip joint</td>
<td></td>
<td></td>
<td></td>
<td>40</td>
</tr>
<tr>
<td>Indefinite complaints (feeling strange, fatigue) present but no pain</td>
<td></td>
<td></td>
<td></td>
<td>35</td>
</tr>
<tr>
<td>No pain during walking (however at the start of walking or after walking for long distances there is sometimes pain)</td>
<td></td>
<td></td>
<td></td>
<td>30</td>
</tr>
<tr>
<td>No spontaneous pain. Pain during walking, but disappears with short rests</td>
<td></td>
<td></td>
<td></td>
<td>20</td>
</tr>
<tr>
<td>Spontaneous pain is sometimes present. Pain during walking but alleviates with rest</td>
<td></td>
<td></td>
<td></td>
<td>10</td>
</tr>
<tr>
<td>Continuous spontaneous pain or nocturnal pain</td>
<td></td>
<td></td>
<td></td>
<td>0</td>
</tr>
</tbody>
</table>

II. Range of motion assessment

Flexion: Joint angle counted in 10° intervals, one point per 10°. However, all points beyond 120° are counted as 12 points (if joint contracture is present this is subtracted and evaluations based on movable range).

Abduction: Joint angles are measured in 10° increments and each 10° is counted as 2 points. However any angle over 30° is counted as 8 points

III. Walking ability

Absolutely no complaints regarding the joint |   |   |   | 40    |
Indefinite complaints (feeling strange, fatigue), no pain |   |   |   | 35    |
No pain during walks (however there may be pain at the start of the walk or after walking long distances) |   |   |   | 30    |
No spontaneous pain. Pain during walks disappears after short rests. |   |   |   | 20    |
Spontaneous pain is sometimes present. Pain present during walks but is relieved after rest. |   |   |   | 10    |
Continuous or spontaneous pain or nocturnal pain. |   |   |   | 0     |

IV. Activities of daily living (ADL)

<table>
<thead>
<tr>
<th>Evaluation</th>
<th>easy</th>
<th>difficult</th>
<th>impossible</th>
</tr>
</thead>
<tbody>
<tr>
<td>Knee blankets</td>
<td>4</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>Standing work (includes housework) (continues for about 30 minutes). If rests are required, it is considered &quot;difficult&quot;. In cases where exercise can only be continued for 5 minutes, it is considered &quot;impossible&quot;. Squatting and standing (those who require assistance: consider it &quot;difficult&quot;)</td>
<td>4</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>Climbing up and down stairs (those who require a railing: consider it &quot;difficult&quot;)</td>
<td>4</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>Getting on and off cars and buses</td>
<td>4</td>
<td>2</td>
<td>0</td>
</tr>
</tbody>
</table>

Figure 1. A: MRI findings before and after KAATSU training. Before training (left), after training (right) B: Dual-energy x-ray absorptiometry (DEXA) scan bone mineral density findings before and after KAATSU training. After training, bone mineral density in the right leg (affected side) was clearly increased, compared with the left leg (healthy side).
a case of secondary arthrosis. Surrounding areas included a small high-signal intensity subchondral bone cyst. The other hip joint showed no signs of abnormally high intensity areas. Based on these MRI findings, the patient was diagnosed with Association Research Circulation Osseous (ARCO) stage IV right femoral head avascular necrosis and secondary hip arthrosis.

Before and after training, several evaluations were carried out. Fig. 2A shows the effectiveness of KAATSU training on JOA hip scores. JOA (pain, b) scores were 10 (right), 40 (left); while the JOA (walk, c) scores were 16 (right), 20 (left); and JOA (score, comprehensive, a) was 55 (right), 84 (left). After training, all scores showed marked improvement in the right affected leg compared to the left healthy leg. JOA (ROM (range of motion), d) revealed no signs of improvement on the affected side. In the latter half of training, pain of the hip joint during walking dissipated so that the patient no longer required a cane to walk. In addition, there was a clear improvement in the patient’s walking style.

Fig. 2B shows the effect on SF–36v2 before and after KAATSU training. Role physical, body pain, general health, vitality, social functioning, role emotional, and mental health were evaluated. Physical function, role physical, body pain, general health, vitality, social functioning all improved after KAATSU training.

Figure 3 shows the effects of KAATSU training on lower limb muscle strength (extension and flexion). Muscle strength during extension and flexion was markedly improved in the right leg on the affected side, compared to the healthy left leg.

Fig. 1B shows the efficacy of using KAATSU training on bone mineral density measured by DEXA. There was a clear increase in right affected leg bone mineral density from 0.798 to 0.836. On the other hand, the left healthy leg went from 0.857 to 0.868 with no clear signs of improvement. Furthermore, MRI findings after KAATSU training shown in Fig. 1A (right) showed a tendency for the right femoral head necrosis to improve.
5. Discussion
We treated a patient with femoral head avascular necrosis for 3 months with KAATSU training and noted the following improvements. 1) After KAATSU training, the JOA score of the hip joint and the JOA pain score both improved markedly. 2) Together with an apparent improvement in gait, femoral muscle strength improvement was noted while bone mineral density of the lower limbs had clearly increased in the affected side. 3) MRIs showed a tendency for improvement of the right femoral head necrotic site.

In this patient, SF–36v2 was evaluated. Of the life functions, physical function, role physical, general health, vitality, social functioning, and walking ability based on the Japan Orthopedic Association hip score (JOA hip score) (Takatori et al., 2010) had all improved. Furthermore, muscle strength on the affected side had increased, proving that muscle strength improvement with KAATSU training can lead to improved walking ability and have a major impact on enhancing QOL. This meant that in the latter half of the training, the patient became able to walk without a cane. In addition, the affected limb could support weight without use of a cane, and it is believed that there was a clear increase in bone mineral density on the affected side.

The mechanism for this effect of KAATSU training includes a characteristic of KAATSU training (Takarada et al., 2000; Sato et al., 2007) which is that exercise under restricted muscle blood flow conditions leads to increased muscle strength and muscle growth after only a short period of low stress exercise. In this case, we observed a clear increase in muscle strength with only 3 months of KAATSU training. KAATSU training is just the right rehabilitation method for patients with various diseases and our aging society (Nakajima, 2010; Abe et al., 2010; Ozaki et al., 2011; Nakajima et al., 2011; Yasuda et al., 2014). Loading included non-weight bearing, elastic band (Yasuda et al., 2015), a dumbbell, walking (Abe et al., 2006), and ergometer (Abe et al., 2010) loads. In this way, KAATSU training is an extremely useful rehabilitation method for use in patients in whom excessive stress would be inappropriate. In our case, we added squats and calf raises to the KAATSU walking schedule, carrying out various forms of exercise in a way that the femoral head would not have to bear any weight. As a result, patient ADLs improved markedly. However, the ROM did not improve. Therefore, in the future we believe a longer investigation is warranted.

Furthermore, after KAATSU training, pain scores based on the SF–36v2 and JOA hip scores showed clear signs of improvement. With improvement of her pain, the patient no longer required a cane and we believe her quality of life has been greatly improved because she regained the ability to walk. The mechanism responsible for improving pain in KAATSU training is still unknown, but increased muscle strength, muscle growth and alleviation of the weight-bearing on the hip joints are believed to play a role. On the other hand, KAATSU training leads to a decrease in intramuscular partial O2 pressure and hypoxemia during exercise under restricted blood flow. Reactive hyperemia develops and vascular endothelial growth factor (VEGF), and nitric oxide (NO) production is enhanced (Takano et al., 2005; Horiuchi and Okita, 2012) in response to vascular refilling after the belts are released, leading to improvements in endothelial function and blood flow. We propose these improvements in blood flow helped to heal the necrosis in the femoral head. Animal models of femoral head avascular necrosis caused by steroids have actually shown that as blood flow into the femoral head decreases, both VEGF protein and mRNA decrease (Wang et al., 2010). However, the improvement of MRIs in the present study for 3 months was minor so the effects should be further confirmed in a long-term study. In addition,
investigations into elucidating the mechanism behind pain relief with KAATSU training are warranted.

Conservative treatment for femoral head avascular necrosis should be instituted carefully to avoid crushing the femoral head, and there are currently no effective rehabilitation methods other than symptomatic treatment such as using a cane or walker to reduce placing weight on the affected hip joint. From the present study, KAATSU training may be a useful rehabilitation method to treat femoral head avascular necrosis.

Summary

We reported our experience with a patient who developed femoral head avascular necrosis while on steroid therapy who was successfully treated with KAATSU training. We believe further clinical research will be necessary.

< Nakajima T, Yasuda T, Fukumura K, and Morita T have participated in seminars until September 2014 donated by KAATSU Japan.>

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Author’s affiliations

T. Nakajima, Heart Center, Dokkyo Medical University Hospital, Tochigi, Japan
T. Yasuda, School of Nursing, Seirei Christopher University, Shizuoka, Japan
K. Fukumura, Kanto Central Hospital of the Mutual Aid Association of Public School Teachers, Tokyo, Japan
M. Kurano, Jumonji University, Research Institute of Food and Nutrition and Health, Tokyo, Japan
T. Imanishi, Medical Affairs, Kyushiu-Kaikan, Kanagawa, Japan
T. Morita, Y. Sato, KAATSU International University, Sri Lanka
Y. Hiraiizumi, Department of Orthopaedic Surgery, Showa University School of Medicine, Tokyo, Japan

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