Case Report

Repeat Radiofrequency Ablation for Painful Sacrococcygeal Chordoma Refractory to Proton Therapy: A Case Report

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Abstract

A 60-year-old woman underwent radiofrequency (RF) ablation to relieve uncontrollable pain from a recurrent sacrococcygeal chordoma, which had been treated 6 years prior using proton therapy. Although the pain had disappeared completely after the first RF ablation, it recurred 4 months after ablation. Moreover, a residual tumor was observed. A second RF ablation was performed 9 months after the first RF ablation for the residual tumor, again alleviating all pain. Patient symptoms were well controlled 7 months after the second RF ablation.

Key words: Radiofrequency ablation, Sacrococcygeal chordoma, proton therapy

INTERRODUCTION

Chordomas, which are uncommon tumors, are clinically regarded as malignant tumors because of their potential to invade other tissues and metastasize [1-3]. They account for 1-4% of all primary malignant bone tumors. Data show that 50-66% of chordomas originate in the sacrococcygeal region, particularly in the fourth or fifth sacral segment, which causes lower back pain and symptoms of mass effect [1-3]. Although surgical resection and radiation therapy, including proton therapy, are standard treatments for the management of sacrococcygeal chordoma, local recurrences frequently occur after those treatments with respective local recurrence rates of 50-65% and 12-28% [1]. No second-line treatment has been established for the treatment of recurrent chordomas after proton therapy.

Accumulating evidence suggests that radiofrequency (RF) ablation is a useful palliative treatment for painful bone and soft tissue tumors [4]. Nevertheless, few reports describe the utility of RF ablation in the management of recurrent sacrococcygeal chordomas after proton therapy. Herein, we describe a case of a painful sacrococcygeal chordoma that was refractory to radiation therapy and was managed successfully by repeated RF ablations.

CASE REPORT

Our institutional review board requires no approval for the publication of retrospective case reports. A 60-year-old woman was referred to our hospital for the treatment of a recurrent sacrococcygeal chordoma after proton therapy. The initial diagnosis of sacrococcygeal chordoma was confirmed via open biopsy during a visit to our hospital 6 years prior. Then, proton therapy (70.4 Gy, 32 fr) was administered. Pelvic lymph node metastasis developed 3 years after proton therapy. Therefore, a second course of proton therapy (70.4 Gy, 32 fr) was administered after the surgical insertion of a

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spacer (Gore-Tex sheet, Applicant; W.L. Gore & Associates, Inc., Tokyo, Japan) between the tumor and intestine to avoid radiation injury [5]. However, follow-up magnetic resonance (MR) imaging revealed a recurrent tumor in the sacrococcygeal region 4 years after the initial proton therapy. Additional proton therapy was regarded as difficult because of duplication of the radiation field. Therefore, conservative treatment was chosen. However, the recurrent tumor gradually became larger. The patient’s hip pain also worsened. The visual analog scale (VAS; 0, no pain; 10, worst pain imaginable) score of her hip pain was 8 at the time our hospital visit. Subsequently, she was unable to lie on her back despite receiving aggressive opioid therapy with 100 mg of oxycodone (OxyContin®; Shionogi & Co. Ltd., Tokyo Japan) per day. Contrast-enhanced MR imaging performed immediately before her visit to our hospital revealed a lobulated hypervascular tumor, 5.7 cm in size, at the sacrococcygeal level. It was regarded as the cause of her hip pain (Fig. 1a).

After obtaining informed consent, RF ablation was performed for the recurrent sacrococcygeal tumor under local anesthesia with 1% lidocaine (Xylocaine; AstraZeneca, Tokyo, Japan) and moderate sedation using fentanyl citrate (Fentanyl; Daiichi Sankyo Co. Ltd., Tokyo, Japan). Two 3-cm exposed cool-tip RF electrodes (Cool-tip; Valleylab/Covidien, Boulder, CO, USA) were placed into the ventral and dorsal parts of the tumor under real-time computed tomography (CT) fluoroscopic guidance (Fig. 1b). Then, RF energy was applied for 10 min using a multiple-electrode switching system (Switching Controller; Valleylab/Covidien, Boulder, CO, USA). RF ablation was performed with impedance control mode. In this mode, RF energy delivery was automatically stopped when the tissue impedance increased 30 Ω from the baseline, which was defined as break down. In this case, the endpoint of the RF ablation was decided as the impedance reaching 30 Ω above the baseline was detected two times, resulting in the tissue temperature increasing to 70°C. To reduce the risk of skin burns, an ice pad was placed on the treatment site surface. The planned ablation protocol was completed without complications. On the following day, her VAS score dropped to 0-1, and she could lie on her back. Follow-up contrast-enhanced MR imaging obtained at 1 month after RF ablation revealed the disappearance of tumor enhancement.

Nevertheless, her hip pain recurred at 4 months after the first RF ablation. Her VAS score increased to 4-5. Contrast-enhanced MR imaging revealed the residual tumor within the spinal canal at the level of S2 and S3. Therefore, a second RF ablation was performed via a trans-sacral approach. Briefly, a 13-gauge bone biopsy needle (Bone marrow harvest needle; MD Tech Inc., Gainesville, Florida) was placed into the sacrum, which was adjacent to the residual tumor. Then, an RF electrode was placed into the residual tumor through the bone biopsy needle and ablation was performed. The second RF ablation was completed without complications. After the second ablation, her VAS score dropped to 0. Her symptoms were well controlled for more than 7 months after the second ablation. MR imaging at 7 months...
after the second ablation (16 months after the first ablation) indicated no signs of chordoma regrowth.

**DISCUSSION**

The results of our case demonstrate that RF ablation is a useful therapeutic option for the management of pain caused by recurrent sacrococcygeal chordomas. Accumulating evidence suggests that RF ablation is a useful therapeutic option for the management of painful bone and soft tissue tumors. Nakatsuka et al. prospectively evaluated the utility of RF ablation for painful bone tumors. Results of their study indicated that RF ablation decreased the VAS score by 2 points or more in 11 of 13 patients (84.6%) with painful bone tumors [4]. Regarding the treatment of painful sacrococcygeal chordomas, a few case reports with a total of 5 patients have been reported to date. Pain relief was obtained in all cases (100%, 5/5) during the short-term follow-up of 4-6 months [2, 3, 6-8]. Although the mechanism of pain relief by RF ablation is not understood completely, decreased tumor pressure on adjacent structures, necrosis of pain fibers, decreases in local or intratumoral interstitial pressure, and/or a decrease in cytotoxic substances released from the tumor itself are suggested as possible mechanisms of pain relief [7].

In this case, RF ablation was also effective for tumor control. Nakatsuka et al. demonstrated that complete (100%) and partial (50-99%) response can be achieved in 10% and 70% of patients with large bone tumors of 5.1-10.0 cm after RF ablation, respectively [4]. Easy repeatability is an important benefit of percutaneous RF ablation [9]. This benefit allowed for a second RF ablation for recurrence after the first ablation, resulting in complete response with disappearance of pain in this case.

Cryoablation is another therapeutic option for the management of recurrent sacrococcygeal chordomas. Kurup et al. reported immediate pain reduction and complete local control after cryoablation in patients with recurrent sacrococcygeal chordomas after surgery [10]. Therefore, ablation therapy, such as RF ablation and cryoablation, can be regarded as a salvage therapy for recurrent sacrococcygeal chordomas after failure of the current standard treatment, including surgery and radiation therapy, including proton therapy.

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**References**