Early Recurrence of Holmes Tremor after Radiofrequency Thalamotomy

Editor,

A-25-year-old male patient admitted due to right-sided tremor which had started 8 years ago in his hand and progressed over the period spreading also to the lower limb within the last 5 years. The patient stated that the tremor disturbed seriously his daily living activities and that he could not maintain his job as a construction worker over the last 2 years due to tremor. The patient was born from consanguineous parents, and the medical or family history was unremarkable. The neurological exam at admission showed unilateral, high-amplitude rhythmic oscillatory movements, present at rest and exacerbated with posture and additionally intensified with action on his right hand and leg. However, also a jerky component in the postural and action tremor causing repetitive, abnormal postures was observed considering dystonic features [Video 1]. Besides, a right-sided dysmetria was also observed during the finger-to-nose testing. Other examinations including motor and sensory tests were within the normal limits. The cranial magnetic resonance imaging (MRI) was within normal limits without a lesion in the midbrain and thalamus. Based on the accompanying dystonia and dysarthria, primary dystonia was also considered among the differential diagnoses; however, the patient did not accept further genetic investigations. Previous medication attempts including levodopa/benserazide, baclofen, and clonazepam had not yielded any amelioration. The patient was re-evaluated and radiofrequency (RF) thalamotomy was decided after obtaining the informed consent. During the operation, target coordinates were obtained by indirect Cartesian measuring based on schaltenbrand stereotactic brain atlas. The coordinates of target for thalamotomy were defined as: 6.3 mm posterior to the midpoint of anterior commissure (AC) and posterior commissure (PC) line and 14 mm lateral and parallel to AC–PC line. The microelectrode-test-stimulation was serially performed starting from 5 mm superior (−5 mm) to the target point and serially down within 0.5 mm intervals. The RF lesioning was performed when the patient was awake and the process was required to administer three times in distinct points to provide the desired clinical response (−4 mm, −3.5 mm, −3 mm). Permanent ablations were performed with a bipolar RF lesioning probe at 60–70°C and have lasted for longest 55 s for a unique target (−4 mm: 54 s, 60°C; −3.5 mm: 55 s 60°C + 15 s 70°C; −3 mm: 55 s 60°C + 15 s 70°C). The tremor had improved dramatically soon after the operation [Video 2]. The improvement was apparent in all types of tremors that was more significant at resting tremor; however, the leg tremor improved to a lesser degree. The patient stated that he could use his hand more comfortably and he defined marked improvement in his daily activities. However, the patient re-admitted 1.5 months later after the operation due to re-emerging of tremor [Video 3]. The deterioration was apparent at resting tremor. The jerky muscle contractions in his hand and marked dystonic posturing also accompanied to the clinic and, therefore, an additional globus pallidus internus-deep brain stimulation (GPİ-DBS) was considered to treat accompanying dystonic movements. However, the phenomenology of the clinic and the neuroimaging data were re-evaluated. The recurrence had occurred rapidly, and the control MRI showed marked resolution of the lesion [Figure 1]. Taken together, the clinical recurrence was associated with acute resolution of the edema associated with the lesion and, considering that the patient was at the early period after surgery and RF associated lesion was at the dynamic phase, clinical follow-up without an intervention was decided.

Discussion

One of the main criticisms regarding the efficiency of focused ultrasound (FUS) thalamotomy is the potential recurrence of tremor at follow-up. Such that, early experiences show that tremor scores worsen by 23.2% within the first year after FUS.[1] Several factors, such as inaccuracy of targeting, insufficient lesion size, resolution of perilesional edema, and disease progression have been discussed to be responsible for symptom recurrence after FUS thalamotomy.[2]

However, RF thalamotomy is classically accepted as a very critical treatment option for medication-refractory tremor with a long-term efficiency.[3] On the other hand, we also know that initially successful RF thalamotomies can loss effectiveness in up to 15% of cases in the long-term course without any identifiable cause.[4] In another study, a 25% recurrence rate of tremor after RF thalamotomy was reported in Parkinson's disease (PD) subjects.[5] The investigation of the neuroimaging data of patients undergoing RF thalamotomy reveal tiny punctate central signal hyperintensity surrounded by concentric

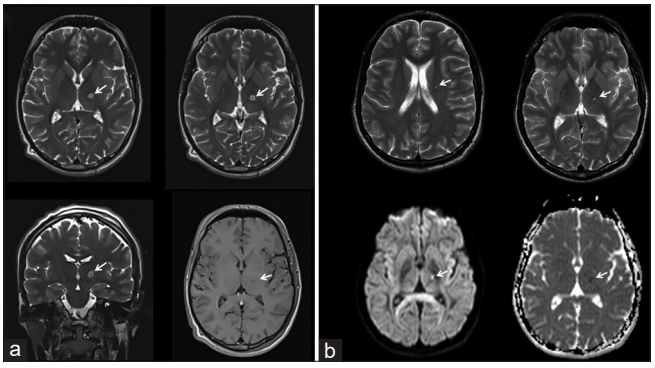

Figure 1: The images of the cranial MRI shows marked resolution of the thalamic lesion associated RF thalamotomy. (a) Images of the postoperative MRI sequences. (b) Images of the cranial MRI, performed, 1.5 months later

zones of hypointense and hyperintense signal. Concentric zone of hyperintensity is surrounded by more extensive, irregular area of mild signal hyperintensity representing the lesion-associated edema.[6] However, the more extensive edema pattern is not found to be associated with a clinically identifiable neurologic deficit.[6] Similar to the MR-guided focused ultrasound thalamotomy, the appearance of the acute lesion suggests an area of central necrosis surrounded by vasogenic edema, the size of which diminishes within the following period. On the other hand, in our patient, the clinic also deteriorated with the resolution of edematous lesion in a very short-term period. This may be associated with the specific method of our procedure. We used bipolar electrode and to achieve the optimal clinical response, the RF lesioning was performed three times in distinct points of the 'ventral intermediate nucleus (VIM) leading to disproportional edema effect. We think that the perilesional edema in our patient might have provided a functional impact on tremor in the acute period, in contrast to the milder edema effect in a routine single RF ablation procedure. According to this view, the recurrence of tremor might be associated with the resolution of perilesional edema within the early subacute period. On the other hand, there are also studies reporting rather increase in lesion size with advancing time in some patients undergoing gamma knife thalamotomy which are accompanied by progressive neurological deficits.[7] The remarkable point was that; we performed ablations with a bipolar RF lesioning probe which leads a milder lesion effect in comparison to monopolar RF lesioning probe. Therefore, we had to perform the ablation procedure to the three separate points to achieve the optimal clinical response. On the other hand, the injury to the adjacent tissues in the monopolar RF is higher leading to increased risk for potential side-effects.[8,9] We hypothesize that the use of monopolar RF lesioning might be more sensible in complex clinical manifestations of tremor that would require a larger volume of ablation. However, there is no randomized controlled study to clarify this hypothesis. We think that the clinical course and the radiological data of this patient is valuable. The presentation of this rare experiences may contribute to our understanding of the lesion effect of RF thalamotomy and provide insights to be used in the clinical practice. Besides, the future prospective reports comparing the efficiency and side effects between bipolar and monopolar lesioning probe techniques in RF thalamotomy may provide critical contributions to the clinical practice.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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