Shapiro Syndrome: Recurrent Hyperhidrosis and Hypothermia With Corpus Callosum Agenesis

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A 46-year-old man suffered recurrent episodes of diffuse hyperhidrosis and hypothermia (Video 1) lasting approximately 20 minutes for 20 days. His temp was 35.4°C, BP was 159/101 mmHg, HR was 120/min and diffuse hyperhidrosis was the only neurologic sign during an episode. Seizures were considered, but the EEG during an episode was normal. MRI showed corpus callosum agenesis (Figure 1). He was diagnosed with Shapiro syndrome which may be due to hypothalamic dysfunction and is responsive to clonidine and carbamazepine. However, due to lack of clonidine and hyponatremia, he was treated with levetiracetam and the episodes disappeared after 5 days.

Video 1 --- http://links.lww.com/WNL/B768

References

Video 1: During these episodes he sweated profusely in the face and upper body and remained alert.

Figure 1: MRI of the brain

(A) Axial and (B) sagittal T2WI showing corpus callosum agenesis. The third ventricle is dilated and the posterior horns of the lateral ventricles are enlargement.
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