BJMS
Borneo Journal of Medical Sciences

CASE REPORT

Unilateral Hemihyperhidrosis in a Stroke Patient and Literature Review on Its Clinicoanatomical Correlation

May Honey Ohn[1*], Khin Maung Ohn[2], Khin Nyein Yin[2], Teh Yong Guang[3], Ng Pey Luen[4], Chin Li Bing[3]

[1] Department of Accident and Emergency, Sandwell and West Birmingham Hospital, Birmingham, West Midlands, United Kingdom
[2] Department of Surgery, Faculty of Medicine and Health Sciences, Universiti Malaysia Sabah, Kota Kinabalu, Sabah, Malaysia
[3] Department of Medicine Based Discipline, Faculty of Medicine and Health Sciences, Universiti Malaysia Sabah, Kota Kinabalu, Sabah, Malaysia
[4] Department of Radiology, Hospital Queen Elizabeth II, Kota Kinabalu, Sabah, Malaysia

*Corresponding author's email: mayhoney.ohn@gmail.com

Received: 3 July 2019

Accepted: 28 January 2020

Keywords: hyperhidrosis, stroke, sweating disorder, cardiovascular accident

ABSTRACT

Contralateral hyperhidrosis from the cardiovascular accident has been infrequently described in the stroke literature. The clinical significance and pathogenesis are yet well understood. This is a case of a 60-year-old man who developed excessive sweating of the paralyzed side with a pure division along the midline of the body secondary to a subcortical stroke, which region is supplied by branches of middle cerebral artery territory known as deep perforating lenticulostriate. To the best of our knowledge, a precise clinicoanatomic correlation between hyperhidrosis and subcortical stroke has not been widely reported. In this review, we summarize the existing literature of post-stroke hyperhidrosis to evaluate the correlation between clinical manifestation and its neuroanatomical location. According to the location of the infarction and clinical features, it can be concluded that both tracks en route through the ipsilateral internal capsule, after originating in the opercular cortex before crossing the brain stem and terminal connections with the contralateral thoracic spinal cord. Therefore, the phenomenon of hyperhidrosis in anterior circulation stroke might be postulated as due to the disruption of the sympathoinhibitory pathway that controls sweat glands, similarly like posterior circulation stroke.

INTRODUCTION

Sweating is physiologically controlled by two centres in the cerebral hemisphere: the hypothalamus and limbic systems, which regulate thermoregulation and emotional

sweating respectively[1]. Sweating dysfunction are ordinarily seen in neurological disease patients as autonomic disturbance[2]. Hypohidrosis or anhidrosis is a common form of the sweating disorder, seen after brain stem stroke[3] and cervical spinal cord injury[4] patients. It is ideally a presentation of Horner's syndrome which is go along with ptosis and ipsilateral miosis evidently linked to the lesions of sudomotor fibres in the uncrossed excitatory hypothalamospinal sympathetic path that supply the sweat glands[3]. On the other hand, an excessive sweating disorder called bilateral hyperhidrosis is seen in quadriplegics and high-level paraplegics after spinal cord injury[4, 5]. It is one of the clinical signs of autonomic dysreflexia and associated with other features of autonomic dysfunction such as pounding headache, flushing, goose flesh, bradycardia and high blood pressure[6]. Conversely, hyperhidrosis over half of the body is nowadays increasingly reported in posterior circulation stroke literature[3, 7 – 11]. The pathogenesis of this phenomenon was conjectured to the disruption of sudomotor fibres in the intersected inhibitory hypothalamospinal sympathetic path that supply the sweat glands. Unilateral hyperhidrosis secondary to anterior circulation cerebral infarcts is a rare clinical finding[12] and only limited studies have been found in patients with large cortical stroke involving the opercular cortex, cortical and subcortical structures[10, 12, 13]. Hyperhidrosis itself interferes with quality of life[14]. However, the clinical significance of post-stroke hyperhidrosis, involved structural lesion and pathogenesis are yet well understood[15]. Here, we present a case of a 60-year-old man who developed contralateral hemihyperhidrosis secondary to an anterior circulation stroke which involves the subcortical region supplied by deep perforating lenticulostriate branches of middle cerebral artery territory. To the best of our knowledge, a precise clinicoanatomic correlation between hyperhidrosis and anterior circulation subcortical stroke has not been widely reported. In this review, we summarize the existing literature of post-stroke hyperhidrosis to evaluate the correlation between clinical manifestation and its neuroanatomical location.

CASE PRESENTATION

A 60-year-old Chinese Malaysian man presented with sudden onset of left-sided facial and body weakness. Otherwise, there are no other symptoms such as loss of consciousness, slurred speech, blurred vision, headache, nausea, vomiting. He had a history of diabetes mellitus and hypertension for 6 years. However, he defaulted treatment and missed medications off and on for the past 10 months. Furthermore, he also had a lacunar stroke last 10 years ago with no residual weakness and full recovery. He also has Alzheimer's disease but not under any treatment. He works as a supervisor in a logging company. He is a chronic smoker for 20 pack-year and ex-alcoholic consumer. He is currently on oral Atenolol 100 mg, Amlodipine 10 mg and Perindopril 8 mg for hypertension, Gliclazide 60 mg, Metformin 1 gram, Saxagliptin 5 mg and subcutaneous injection of Actrapid 10 unit for diabetes mellitus, oral Aspirin 150 mg and Atorvastatin 40 mg.

Upon arrival at the emergency department, he was fully conscious, and peripheries were warm with good pulse volume. Pupils were 2/2 reactive. Vital signs were blood pressure 182/112 mmHg, pulse rate 72/min, respiratory rate 16/min, temperature 36.8°C and oxygen saturation of 99% under room air. Neurological examination showed facial asymmetry with loss of left nasolabial fold and reduced muscle strength (Medical Research Council Grade III) over the left upper and lower limbs. Otherwise, the other cranial nerves were intact. There were no cortical signs present. In addition, tone and reflexes were reduced as well as extensor plantar response was elicited over the left side of the body. The sensation was intact and cerebellar sign was negative. Laboratory findings were

normal except hypercholesterolaemia and hyperglycaemia. Non-enhanced computerized tomography scan of the brain showed ill-defined hypodensities at the right internal capsule with no haemorrhagic transformation which is compatible with clinical findings of left-sided body weakness (Figure 1).

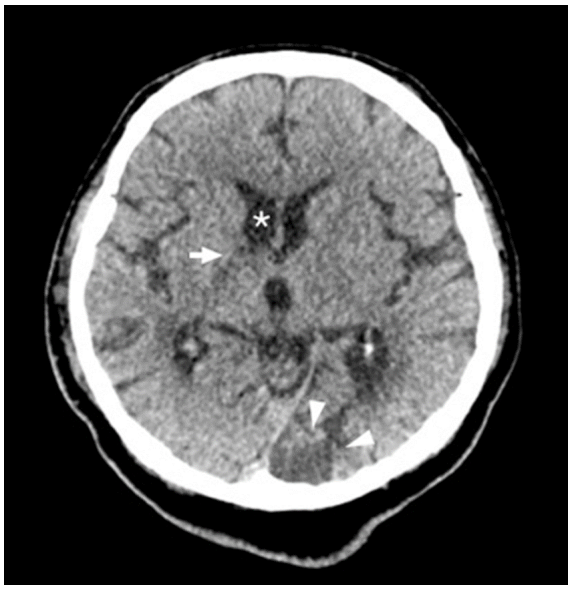

Figure 1 Axial section of non-enhanced Computerized Tomography brain scan at the level of the basal ganglia. Ill-defined hypodensities at the right internal capsule (arrow) with ex-vacuo dilatation of the anterior horn of the right lateral ventricle (Asterix) which is suggestive of right acute on chronic anterior circulation subcortical infarct. There is also a fairly well-defined hypodensity at the left occipital lobe (arrowheads) with a relatively dilated occipital horn of left lateral ventricle, suggestive of a left subacute posterior circulation artery infarct.

Despite the subacute occipital lobe infarct appearance in CT brain, there were no posterior circulation stroke signs and symptoms. Alberta stroke program early CT score (ASPECTS) was 8. MRI brain could not proceed due to financial constraint. He was diagnosed as acute anterior circulation subcortical stroke and treated with aspirin and atorvastatin. Subsequently next day after admission, drenching sweats were documented over the left side of the body especially face, arm and upper torso. There was no previous history of similar symptoms. Furthermore, there was no evidence of autonomic or hypothalamic dysfunction. One month after physiotherapy, the patient was still hemiplegic (muscle strength over the right side of the body improved to 4/5) with the return of sweat function back to normal.

DISCUSSION

Contralateral hyperhidrosis is rarely reported in the stroke literature, with an incidence around 1% in thalamic[16], hypothalamic[17], pontine[7 – 9, 18], medulla oblongata[10, 11], and cerebral hemispheric[10, 12, 13] infarction. However, it has received scant attention in hemiplegic patients after subcortical stroke. The pathogenesis and clinical significance of hyperhidrosis in a patient with stroke are still uncertain[15, 19]at 1 month, and at 6 months after infarction. Excessive evaporation on the paretic side when compared with the nonparetic side was already found at baseline, but after the heating stimulus, this asymmetry reached statistical significance on the forehead, chest, forearm, and hand during the whole 6-month follow-up. Significant asymmetry in sweating occurred in 29 of the 40 patients (73%.

Six unilateral hyperhidrosis cases have been found primarily out of 633 consecutive strokes, particularly in the scenario of the anterior circulation occlusion by Labar[13]. Out of six, two cases were localized infarctions of the opercular cortex, and the rest four cases were large cortical infarcts. Korpelainen[15]at 1 month, and at 6 months after infarction. Excessive evaporation on the paretic side when compared with the nonparetic side was already found at baseline, but after the heating stimulus, this asymmetry reached statistical significance on the forehead, chest, forearm, and hand during the whole 6-month follow-up. Significant asymmetry in sweating occurred in 29 of the 40 patients (73% reported that it can

also be the result of lesions of the premotor cortical areas affected in anterior circulation stroke. On the grounds of studies in human[13] and animal[20, 21], the researchers conjectured the pathogenesis of hyperhidrosis as the disruption of sudomotor fibres in a putative crossed inhibitory hypothalamospinal sympathetic pathway that originates from the cerebral cortex, probably the operculum, then trespassing through the ipsilateral thalamus, hypothalamus, pons and medulla, ultimately crossed in the inferior brainstem and terminated in the contralateral thoracic spinal cord[3, 10, 16, 17, 22]. This pathway inhibits sudomotor neurons resulting in over sweating of the contralateral face and body. It has also been suggested that the pathways follow the pyramidal tract[10, 23]. Therefore, emerging evidence showed the degree of hyperhidrosis in stroke patients are correlated with the presence of significant neurological disability regardless of the size and location of ischaemic brain lesion[24]pathogenesis, and clinical correlates of sweating dysfunction in stroke. Methods: We studied sweating at baseline and after a heating stimulus in 53 patients with acute hemispheral brain infarction and in 40 healthy control subjects by using a quantitative evaporimetric method. Results: Significant hyperhidrosis on the paretic side of the body was verified in 55% of the patients at baseline, in 74% after 5 minutes of heating, and in 77% after 10 minutes of heating. Hyperhidrosis was established throughout the body and correlated with the severity of paresis, the presence of reduced muscle tone, and the extensor plantar response. Conclusions: The phenomenon of hyperhidrosis in hemiparetic patients reflecting autonomic dysfunction seems to be a common manifestation that should be listed among the expected consequences of brain infarction. This sweating disturbance might be attributed to a lesion of a putative sympathoinhibitory pathway controlling sweating. The failure of this pathway could also be related to other manifestations of sympathetic hyperfunction, e.g., cardiac complications. Therefore, assessment of sweating may provide a new, important aspect in the evaluation of stroke patients. (Stroke 1992;23:1271-1275. As such, Laber[13] affirmed that it is related to increased mortality. Contradictorily, Korpelainen et al. (1992)[24]pathogenesis, and clinical correlates of sweating dysfunction in stroke. Methods: We studied sweating at baseline and after a heating stimulus in 53 patients with acute hemispheral brain infarction and in 40 healthy control subjects by using a quantitative evaporimetric method. Results: Significant hyperhidrosis on the paretic side of the body was verified in 55% of the patients at baseline, in 74% after 5 minutes of heating, and in 77% after 10 minutes of heating. Hyperhidrosis was established throughout the body and correlated with the severity of paresis, the presence of reduced muscle tone, and the extensor plantar response. Conclusions: The phenomenon of hyperhidrosis in hemiparetic patients reflecting autonomic dysfunction seems to be a common manifestation that should be listed among the expected consequences of brain infarction. This sweating disturbance might be attributed to a lesion of a putative sympathoinhibitory pathway controlling sweating. The failure of this pathway could also be related to other manifestations of sympathetic hyperfunction, e.g., cardiac complications. Therefore, assessment of sweating may provide a new, important aspect in the evaluation of stroke patients. (Stroke 1992;23:1271-1275 and Kim et al. (1995)[10] claimed the presence of hemihyperhidrosis in stroke patient is not a sign of poor prognosis.

Some studies concluded that ischaemic brain infarction almost inevitably damages the autonomic nervous system. It is because central autonomic network (CAN) is located around the insular cortex, amygdala, hypothalamus, medulla, periaqueductal grey matter, parabrachial complex and nucleus of tractus solitarius[25]. Damage to these regions due to brain injury could cause loss of cortical inhibition of the hypothalamus resulting in

increased contralateral sympathetic outflow. The failure of this inhibition can be related to manifestations of paroxysmal sympathetic hyperfunction, e.g. tachycardia, hyperthermia, vasodilation in addition to hyperhidrosis[24]. However, we believe none of those CAN regions have been affected by stroke according to the imaging finding which in accord with the absence of other associated autonomic dysfunction features.

According to one of the case reports of a pure hypothalamic stroke patient, hyperhidrosis phenomenon was possibly explained that due to the fact of hypothalamus receiving its blood supply from the posterior cerebral artery, hypothalamus being a central thermoregulatory centre is responsible for the hyperhidrosis in this posterior circulation stroke patient[3, 17, 22]. Bassetti (1995)[3] reported the combination of both contralateral hemihyperhidrosis and ipsilateral anhidrosis happened in a posterior circulation occlusive disease patient where anterolateral midbrain, ventroposterolateral thalamic-subthalamic and temporo-occipital lobes were affected. Therefore, it was speculated that the thermoregulating fibres descend in the ventroposterolateral thalamic-subthalamic area, anterolateral midbrain, dorso-lateral part of the pontine tegmentum and the lateral reticular formation in the medulla[7]. Likewise, Kim et al. (1995)[16] reported that pure thalamic infarction is also associated with persistent contralateral hyperhidrosis due to sharing of the same crossed inhibitory sweating pathway.

In spinal cord disorder patients, the mechanism of hyperhidrosis has been due to the activity of isolated disinhibited spinal cord. A specific stimulus such as the bladder or rectum distension triggered episodic and transient hyperhidrosis above the level of spinal cord lesion mostly associated with cutaneous flushing, headache, hypertension and reflex bradycardia. However, unlike spinal cord injury, hyperhidrosis seen in stroke patients are not commonly found to be associated with such autonomic dysfunction[10, 13, 19]. Nevertheless, the literature review shows post-stroke hyperhidrosis can be spontaneous or provoked by stress, exercise, infection, heat exposure or effort[9, 11, 17]. In our patient, it was noted to have no instigating factor for hyperhidrosis.

In most of the stroke cases, unilateral hyperhidrosis was limited to the contralateral side of the body and typically involved face, arm and upper torso. The onset of the symptom varies from few days[3, 10, 17, 22] to months[11, 18] later after infarct and duration of symptoms are typically transient, lasting for few days[13], weeks[7, 18], months[10] up to years[9, 17, 22]. In our patient with pure motor stroke, unilateral sweating appeared on the second day of stroke and lasted for one month. According to a prospective study of hemihyperhidrosis patients among hemispheric brain infarction by Korpelainen et al. (1993)[15] at 1 month, and at 6 months after infarction. Excessive evaporation on the paretic side when compared with the nonparetic side was already found at baseline, but after the heating stimulus, this asymmetry reached statistical significance on the forehead, chest, forearm, and hand during the whole 6-month follow-up. Significant asymmetry in sweating occurred in 29 of the 40 patients (73%, asymmetry sweating was observed in 73% of patients in the acute phase of infarction, 56% after 1 month and 85% after 6 months. In term of management, most post-stroke hyperhidrosis cases do not require treatment as it is a benign and self-limiting disorder[12]. Awada et al. (1991)[26] inferred that the duration of hyperhidrosis may be related to the site of the nervous lesions and compensatory mechanisms are much stronger in higher lesions.

In this case, the patient presented with left facial involvement (loss of nasolabial fold, drooling of saliva and facial asymmetry) and left-sided hemiparesis. Also, the patient suffered from left-sided hyperhidrosis mainly over the face, arm and upper trunk after the

acute stroke. Noted that, in CT shows an acute right internal capsule infraction. The mapping of this lesion is to the involvement of deep subcortical structures which are both supplied by the lenticulo-striate branches of the middle cerebral artery. The presentation of contralateral hemihyperhidrosis secondary to internal capsule infarction supports the hypothesis of the existence of the putative inhibitory sympathetic pathway. Notably, there was no literature confirmed the pathway of inhibitory sympathetic pathway en routing through the internal capsule alongside with pyramidal tract as the pathogenesis of unilateral hyperhidrosis in a subcortical patient. This hypothesis can be supported by the previous study in that an association was found between hyperhidrosis and pyramidal tract lesion.

It can be also debated whether the hyperhidrosis is contributed by any medication or underlying medical illness. A study done by Akbas and Kilinç (2018) found out that there was an association between hyperhydrodrosis and diabetes mellitus (25.7%) as well as antidiabetic agents (14.2%)[27]. Furthermore, hyperhidrosis secondary to medication is more common to have generalized sweating rather unilateral. In that study, clinical types of secondary hyperhydrorosis frequently detected are palmoplantar and axillary regions, followed by forehead. However, in our case, the patient had defaulted medication and the unilateral hyperhidrosis was acute onset which occurred only after the stroke.

CONCLUSION

Profuse sweating on paresis side of the body is one of the sequels of the cerebral hemispheric infarction. The clinicians should be attentive of unilateral hyperhidrosis, although it is a transient feature affecting especially face, arm and upper torso in stroke patients. This case was reported with the hope of increasing awareness and recognition of underreported hyperhidrosis in stroke cases. To conclude, the acute internal capsule infarction damages corticopyramidal tracts as well as inhibitory sympathetic pathway resulting in contralateral hemiparesis and excessive sweating. According to the location of the infarction and clinical features, it can be concluded that both tracks en routing through the ipsilateral internal capsule after originating in the opercular cortex before crossing the brain stem and terminal connections with the contralateral thoracic spinal cord. Therefore, the phenomenon of hyperhidrosis in anterior circulation stroke might be postulated as due to the disruption of the sympathoinhibitory pathway that controls sweat glands, similarly like posterior circulation stroke.

CONFLICT OF INTEREST

The authors declare that they have no competing interests in publishing this article.

CONSENTS

Written informed consent was obtained from the patient to publish the case with its related pictures. A copy of the written consent is available for review by the Chief Editor.

REFERENCES

1. Schlereth T, Dieterich M, Birklein F. (2009). Hyperhidrosis – causes and treatment of enhanced sweating. Dtsch Arztebl Int 106: 32 – 37.
2. Mo J, Huang L, Peng J et al. (2019). Autonomic Disturbances in Acute Cerebrovascular Disease. Neurosci Bull 35: 133 – 144.
3. Bassetti C, Staikov IN. (1995). Hemiplegia vegetativa alterna (ipsilateral Horner's syndrome and contralateral hemihyperhidrosis) following proximal posterior cerebral artery occlusion. Stroke 26: 702 – 704.
4. Schulz V, Ward D, Moulin DE. (1998). Segmental hyperhidrosis as a manifestation of spinal and paraspinal disease. Can J Neurol Sci / J Can des Sci Neurol 25: 325 – 327.

5. Compston A. (2008). The automatic bladder, excessive sweating and some other reflex conditions, in gross injuries of the spinal cord. By Henry Head, MD, FRS and George Riddoch, MD, Captain, Royal Army Medical Corps. (Officer in charge of the Empire Hospital, Vincent Square). Brain 1917; 40: 188 – 263. Brain 131: 2237 – 2239.
6. Kneisley LW. (1977). Hyperhydrosis in Paraplegia. Arch Neurol 34: 536 – 539.
7. Mon Y, Mizotani M. (1992). A case of hemi-hyperhidrosis and non-paralytic pontine exotropia due to brainstem infarction. Rinsho Shinkeigaku 32: 718 – 721.
8. Rey A, Martí-Vilalta JL, Abellán MT. (1996). Contralateral hyperhidrosis secondary to the pontine infarct]. Rev Neurol 24: 459 – 460.
9. Pellecchia MT, Criscuolo C, De Joanna G et al. (2003). Pure unilateral hyperhidrosis after pontine infarct. Neurology 61: 1305.
10. Kim BS, Kim YI, Lee KS. (1995). Contralateral hyperhidrosis after cerebral infarction. Clinicoanatomic correlations in five cases. Stroke 26: 896 – 899.
11. Rousseaux M, Hurtevent JF, Benaim C et al. (1996). Late contralateral hyperhidrosis in lateral medullary infarcts. Stroke 27: 991 – 995.
12. Faruqi S. (2004). Hemihyperhidrosis in cerebral infarction. Age Ageing 33: 514 – 515.
13. Labar DR, Mohr JP, Nichols FT. (1988). Unilateral hyperhidrosis after cerebral infarction. Neurology 38: 1679.
14. Minota K, Coon EA, Benarroch EE. (2019). Neurologic aspects of sweating and its disorders. Neurology 92: 999 – 1005.
15. Korpelainen JT, Sotaniemi KA, Myllylä V V. (1993). Asymmetric sweating in stroke: A prospective quantitative study of patients with hemispheral brain infarction. Neurology 43: 1211 – 1214.
16. Kim JM, Seo SD, Kim YW, Hwang YH. (2014). Contralateral hyperhidrosis in anterior thalamic infarction. Clin Auton Res 24: 311 – 313.
17. Smith CD, Criscuolo C, Joanna G De et al. (2001). A hypothalamic stroke producing recurrent hemihyperhidrosis. Neurology 56: 1394 –1396.
18. Sato K, Nitta E. (2000). Pontine hemorrhage presenting with Foville syndrome and transient contralateral hyperhidrosis. Rinsho Shinkeigaku 40: 271 – 273.
19. Wang J-C, Chan R-C, Chang P-Y et al. (2015). Profuse Unilateral Hyperhidrosis Induced by Urinary Retention in a Stroke Patient. Neurologist 19: 82 – 84.
20. Bernthal PJ, Koss MC. (1978). Some physiologic characteristics of the electrodermal reflex in the cat. Brain Res Bull 3: 437 – 441.
21. Bernthal PJ, Koss MC. (1984). Evidence for two distinct sympathoinhibitory bulbo-spinal systems. Neuropharmacology 23: 31 – 36.
22. Sakashita Y, Kakuta K, Kakuma K et al. (1992). Unilateral persistent hyperhidrosis after ischemic stroke. Rinsho Shinkeigaku 32: 454 – 456.
23. Fisher CM. (1977). Bilateral occlusion of basilar artery branches. J Neurol Neurosurg Psychiatry 40: 1182 – 1189.
24. Korpelainen JT, Sotaniemi KA, Myllyla V V et al. (1992). Hyperhidrosis as a Reflection of Autonomic Failure in Patients with Acute Hemispheral Brain Infarction an Evaporimetric Study. Stroke 23 (9): 1271 – 1275.
25. Siefferman JW, Lai G. (2015). Propranolol for Paroxysmal Sympathetic Hyperactivity with Lateralizing Hyperhidrosis after Stroke. Case Rep Neurol Med 2015: 421563.
26. Awada A, Ammar A, al-Rajeh S et al. (1991). Excessive sweating: an uncommon sign of basilar artery occlusion. J Neurol Neurosurg Psychiatry 54: 277 – 278.
27. Akbaş A, Kilinç F. (2018). Investigation on aetiological factors in patients with hyperhidrosis. Cutaneous and Ocular Toxicology: 1 – 6.