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SHORT COMMUNICATION

Idiopathic Localised Unilateral Hyperhidrosis in a 7-year-old Girl: A Case Report

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Sweating is a physiological process, which is vital for maintaining a constant body temperature and good grip function. Excessive sweating (hyperhidrosis) can on the contrary lead to severe social problems for the affected individual with a decreased quality of life (1). Primary or essential hyperhidrosis is seen symmetrically located to palms, feet, axillae and groins/anogential or generalised in larger areas such as head and trunk. Emotional as well as heat and effort trigger hyperhidrosis in genetic predisposed individuals (2). Essential hyperhidrosis must be differentiated from secondary hyperhidrosis, which is caused by disorders such as infection, malignancy, medication, neurological, or endocrine diseases (2). While axillary hyperhidrosis is quite common with a prevalence of about 1.4 % (3), hyperhidrosis on the body unilaterally or lesionally is very rare. It has been termed localised unilateral hyperhidrosis (LUH) (4) and has been associated with organic disorders such as malignancies (5) or neurological disorders (6), but it has also been reported after a trauma (4) or as idiopathic (7).

CASE REPORT

A 7-year-old girl was referred to our clinic from the paediatric ward. She was otherwise healthy and thriving when she suddenly developed a unilateral thoracic pronounced hyperhidrosis (Fig. 1) on the right side of her chest. Four weeks before the hyperhidrosis began, the girl had symptoms of a common cold; these symptoms had stopped when the hyperhidrosis started.

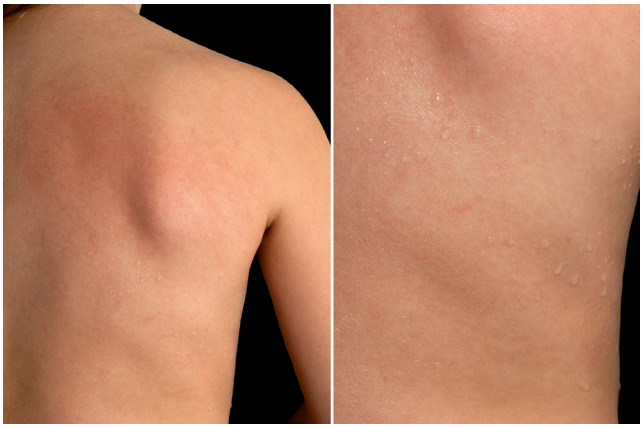

Fig. 1. Clinical manifestation of localised unilateral hyperhidrosis on the right side of the back in a 7-year-old girl.

She never had any skin problems or signs of herpes zoster, and the family history was unremarkable. When she was referred to our department, the hyperhidrosis had lasted for one year. She has approximately 6 daily episodes of hyperhidrosis, each episode lasting about one hour. During the outbreaks the skin in the involved area became slightly erythematous, but she felt no discomfort, only a little warm. The only observed possible triggering factor was stress. However, she also had outbreaks during sleep. We used the iodine starch test to visualise and map the involved area and found that the hyperhidrotic area was sharply demarcated and localised to the right T2–T9 dermatomes (Fig. 2). The area measured 14×28 cm^2. Using gravimetric testing, we quantified her sweat production to 1.6 mg/cm^2/min. No increase in the sweat production was observed on the contralateral side. At the department of paediatrics she went through numerous diagnostic tests in search of the cause of the hyperhidrosis. Her physical and neurological examination was without any pathological findings, including equal pupillary responses and tendon reflexes. Blood tests showed

Fig. 2. Starch-iodine test. An iodine solution is applied. After it dries, starch is sprinkled on the area. The starch-iodine combination turns black wherever there is excess sweat.

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normal endocrine function, haematology and plasma chemistry. Magnetic resonance imaging (MRI) scans of her cerebrum, paravertebral area, and cervical and thoracic spine were also completely normal. The family refused a skin biopsy and any treatment attempts of the hyperhidrosis. Based on patient history, clinical and paraclinical features the diagnosis of idiopathic LUH was made. All known differential diagnoses were excluded; Ross syndrome was excluded due to a normal neurological examination (8).

DISCUSSION

LUH is a rare disorder and the pathogenesis is still unknown. It was first reported in 1947 and since then it has only been reported in the literature occasionally (9). It is characterised by attacks of profuse sweating, usually in sharply demarcated areas, and is most often localised to the upper extremities and forehead. It can be precipitated with heat, and in some cases also triggered by emotional stress and gustatory stimulation. In some cases the sweating is seen without any stimulus at all (4).

LUH has been reported in association with diseases of the nervous system such as cerebral infarction (10) or spinal cord injuries (11). Furthermore, it has been associated with intrathoracic tumours such as mesotheliomas (5). In our case, the MRI scans were without any pathological findings, making major diseases of the nervous system an unlikely cause. Based on the clinical evaluation, the laboratory test results, the MRI scans, the age of the patient, and the fact that the hyperhidrosis has lasted for almost 2 years and she is still healthy and thriving, makes a malignant cause very unlikely.

LUH has also been reported after traumas (4). Kreyden et al. suggested a possible mechanism of a misdirected reconnection of the sympathetic nerve fibre network after injury, similar to Frey syndrome. Frey syndrome was first described by Polish neurologist Lucie Frey, in 1923, and is characterised by sweating, flushing, a sense of warmth, and occasional pain in the temporal area, following the production of a strong salivary stimulus (12). The syndrome's pathophysiology is believed to be explained on the basis of damage to the auriculotemporal nerve, e.g. during parotid surgery, and subsequent re-innervation of sweat glands by parasympathetic (salivary) fibres and some form of transaxonal excitation from adjacent fibres or ganglion (12).

LUH caused by increased size of sweat glands (13) and eccrine hamartoma (14) have been reported. No skin biopsy was performed; however, the extent of the affected area, the sudden onset, and the fact that there was no visible skin lesion make these diagnoses unlikely. Idiopathic LUH have previously only been reported in a single child, a 4-year-old girl (7). Like our patient she had no associated disorder. The pathogenesis in these cases remains unknown. It has been suggested that idiopathic LUH probably is a result of dysregulation of the autonomic nervous system (15), although not yet possible to detect.

At present, the family in our case does not wish to pursue treatment. Treatment with botulinum toxin has been reported to be successful in LUH (4, 15). Topical treatment with aluminium chloride hexahydrate would probably not be sufficient.

The authors declare no conflict of interest.

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