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CASE REPORT

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Oculosympathetic hyperactivity in idiopathic hyperhidrosis

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Abstract We describe a patient suffering from idiopathic hyperhidrosis, more pronounced on the right side, who presented with intermittent oculosympathetic hyperactivity (mydriasis, lid retraction and more rapid pupillary dilatation) on the same side. Mydriasis was induced by stress, loss of sleep and cold pressor test. The clinical features in this patient suggest an involvement of the hypothalamic structures.

Key words anisocoria · idiopathic hyperhidrosis · pupils · sympathetic nervous system

Case report

Idiopathic hyperhidrosis is a condition that is characterized by an excessive, uncontrolled production of sweat, typically on the axillae, palms and soles [1, 5]. The cause of this dysfunction is unknown. A positive family history is present in some cases. We present the case of a 27-year-old man suffering from idiopathic hyperhidrosis who was referred to our outpatient unit because he had noticed that his pupils were of an unequal size (anisocoria). He had suffered from excessive sweating since childhood, but he did not know exactly when the pupillary alterations had developed. His mother remembered having noticed transient anisocoria during her son's childhood.

When examined by us, the patient presented with marked axillar, palmar and plantar hyperhidrosis, which was more pronounced on the right side. His pupils were equal in size and the marginal reflex distance between the lids was normal. His medical history, clinical features, blood pressure and heart rate values, endocrinologic assessments and total body computed tomography were all normal, thereby supporting the diagnosis of idiopathic hyperhidrosis, and not of a secondary form. The patient did not suffer from diabetes, alcoholism or any other disorders that may affect the autonomic nervous system. He had no history of migraine or any diseases known to induce permanent or transient anisocoria [8]. The patient was given a diary in which he was asked to record the presence and duration of anisocoria, as well as the situations in which it usually occurred. A 3-month follow-up with frequent examinations was scheduled.

During the follow-up, anisocoria was observed in 6 out of 12 examinations (performed once a week). When present, the anisocoria was always associated with severe hyperhidrosis. Brain, spinal cord, neck and lung MR did not reveal any abnormalities. Carotid echo-color-Doppler sonography and cerebral MR angiography were also normal.

Computerized pupillometry, performed twice when the pupils were of both equal and unequal size, revealed that the direct and consensual light reflexes were always normal. No signs of Horner's syndrome, such as a reduced marginal reflex distance between the lids and exophthalmos, were apparent, when the pupils were of equal size. A degree of lid retraction was observed on the mydriatic pupil when anisocoria occurred. A dysfunction of the oculosympathetic system was hypothesized in this subject.

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In accordance with previous studies performed in the headache field [3, 4], oculosympathetic function was tested by pupillary instillation of 5 % tyramine to assess post-ganglionic function, and 1 % phenylephrine to assess adrenoceptor hypersensitivity. Pupillary changes, measured using a pupillometer (Fairville Medical Optics), were assessed twice when the pupils were of equal size, and twice when they were of unequal size.

When the pupils were of equal size (both 4 mm in diameter), tyramine induced a bilateral, equal pupillary dilatation of 3 mm on both sides (both 7 mm); phenylephrine instilled under similar conditions did not modify the pupillary diameters. When the pupils were of unequal size (right pupil 6 mm and left pupil 4 mm), tyramine induced a bilateral pupillary dilatation of 3 mm on both the right and left sides (right pupil 9 mm, left pupil 7 mm); phenylephrine did not modify the pupillary diameters. When repeated a second time, pupillary testing yielded similar results. The symmetric mydriatic responses to tyramine indicated the integrity of the third order sympathetic neuron, while the lack of any response to 1 % phenylephrine excluded the presence of adrenoceptor hypersensitivity in our patient. Since "the redilatation phase of the light reflex can shed light" on sympathetic function, including its possible hyperactivity, this parameter was investigated [7]. Computerized pupillometry revealed that the pupil redilatation after contraction was more rapid on the right side than on the left side (redilatation velocity: right pupil 3.6 mm/s, left pupil 2.4 mm/s) when anisocoria occurred (right pupil 7.2 mm, left pupil 6.5 mm). By contrast, the redilatation velocity was similar for both pupils (right pupil 2.4 mm/s, left pupil 2.44 mm/s) when they were of equal size (diameter 6.6 mm). Similar results were obtained in the second assessment.

The diary kept by the patient revealed that anisocoria occurred 11 times (6 of which we also observed) during the 3-month follow-up. Anisocoria lasted from 1 to 5 days and was always associated with particularly stressful events (family and work) or psychophysical tiredness (particularly loss of sleep).

Cold pressor test, repeated twice, promptly induced anisocoria, with a more marked, rapid dilatation of the right pupil (from 4 mm to 8 mm – maximal dilatation in 20 seconds) and a mild, slower dilatation of the left pupil (from 4 mm to 5.5 mm – maximal dilatation in 30 seconds). Our findings are indicative of oculosympathetic hyperactivity on the right side.

Increased excitability of the sympathetic sudomotor pathways has been demonstrated in idiopathic hyperhidrosis [6]. Abnormalities in cardiac parasympathetic function have also been described in this condition [2]. In our patient, oculosympathetic hyperactivity was associated with idiopathic hyperhidrosis and was visible on the side with more pronounced hyperhidrosis. The clinical features in this subject suggest an involvement of hypothalamic structures. The fact that both anisocoria and marked sweating were induced by sympathetic activation due to stress, loss of sleep and the cold pressor test lends support to this interpretation.

Further studies on pupils, as well as on other autonomic parameters, are warranted in patients with idiopathic hyperhidrosis to clarify the involvement of autonomic pathways other than those related to sweating in this disorder.

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