

Case Report

Acquired Idiopathic Generalised Anhidrosis: A human Equivalent to Equine Dry coat disease?

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Abstract

Case description: We report two male patients with acquired inability to sweat. One noticed this during military training, the other after a period of intense exercise.

Clinical findings: Neither of them had stigmata of ectodermal dysplasia or evidence of autonomic dysfunction. In case no.1, a biopsy revealed that sweat glands were present in the finger, a sweat duct was seen in the forearm skin, but no sweat glands or ducts were seen in the thigh skin specimen. No lymphocyte infiltrate was seen around the observed sweat glands. Case no.2 had declined to be biopsied.

Treatment and Outcome: No treatment has been ordered besides.

Clinical relevance: Acquired idiopathic generalized anhidrosis and equine dry coat disease have many features in common and these may be equivalent conditions in the two mammalian species. These two conditions display similarities in clinical presentation and circumstances preceding onset of the conditions. Pathogenesis of both conditions has not been clearly established. Current medical management of both conditions is neither specific nor satisfactory.

Keywords

- Anhidrosis
- Dry coat disease
- Equine
- Human

INTRODUCTION

Sweating has a variety of functions in humans and horses, the most important of which is thermoregulation.

Anhidrosis is the absence of sweating. The McGraw-Hill Concise Dictionary of Modern Medicine defines anhidrosis as "The lack of appropriate sweat production in response to thermal or pharmacologic stimulation, which may become a medical emergency with hyperthermia, heat exhaustion, heat stroke, and death. Anhidrosis may affect the entire body or be segmental in distribution. Causes of anhidrosis may be divided into structural – e.g. anhidrotic ectodermal dysplasia, or functional defects, often related to autonomic control which involve the central or peripheral nervous systems".

Dry coat or non-sweating disease of horses, also known as equine anhidrosis [1], develops in horses stabled and trained under hot, humid conditions. Affected animals are unable to sweat effectively despite appropriate stimuli. An earlier form of this condition, commonly known as "puff disease" or "the puffs", occurs in a proportion of horses in tropical weather during peak daytime temperatures. These horses are unable to thermoregulate effectively by sweating and resort to panting to dissipate excess heat. Onset of anhidrosis may be sudden, following a period of profuse sweating [1,2]; or gradual, with a slowly

declining sweat response over time [3]. In temperate regions, this condition appears in spring or summer, especially during unseasonable humid conditions, when there has been less time for the horses to adapt to the seasonal changes. This condition has a tendency to persist.

Acquired idiopathic generalized anhidrosis (AIGA) in humans has many features in common with dry coat disease and we postulate that these may be equivalent conditions in the two mammalian species. We would like to illustrate this with 2 of our own cases from hot, humid Singapore, a small country sitting on the equator that is hot and humid all year round.

CASE REPORTS

Case 1

A 21 year-old national serviceman was referred for a 1 year history of inability to sweat. He developed this anhidrosis suddenly during military training, and complained of fever after physical activity. There was no family history of a similar problem. Clinical examination was unremarkable. There were no stigmata of ectodermal dysplasia. A starch-iodine sweat test revealed only mild sweating on the face with deficiency of sweating over the trunk & limbs (Figure. 1). He was referred to a neurologist who found no evidence of autonomic dysfunction (Normal nerve conduction, no postural hypotension on standing

and on passive tilt till 60 degrees, blood pressure rises to isometric exercise and cold pressure stimulus, normal heart rate variability to change of posture, deep respiration and the vasalva manouever, sympathetic skin response is present on the palms and soles). Other investigations revealed high IgE levels of 736 IU/ml (Normal 0-87). Antinuclear antibodies (ANA) and extractable nuclear antigens (ENA) were negative. Full blood count and erythrocyte sedimentation rate (ESR) were unremarkable. A biopsy revealed that sweat glands were present in the finger, a sweat duct was seen in the forearm skin, but no sweat glands or ducts were seen in the thigh skin specimen. No lymphocyte infiltrate was seen around the observed sweat glands.

Case 2

A 49-year-old male sales & site supervisor presented with 1 month of inability to sweat. His wife gave a history that prior to the onset of anhidrosis, the patient attempted to exercise intensively and was noticed to have sweated profusely for a while before the onset of anhidrosis. On examination, there were no stigmata of ectodermal dysplasia. He was referred to a neurologist who found no evidence of autonomic dysfunction (details not available). The patient declined a skin biopsy.

DISCUSSION

Generalized anhidrosis (GA) may be congenital or acquired. Acquired generalized anhidrosis can be classified as secondary or idiopathic 4. Secondary acquired generalized anhidrosis is seen in systemic diseases such as diabetes mellitus, Sjogren's syndrome 5 and Fabry's disease 6. The pathogenesis of acquired idiopathic generalized anhidrosis (AIGA) is still unknown and no specific cause has been established.

We would like to propose that many cases of AIGA are the equivalents of equine dry coat disease, and that both conditions are the result of training under hot humid conditions before thermoregulatory conditioning. Most affected horses, and our patients, develop the anhidrosis during a period of intense physical training before they are thermoregulatorily fit. Onset of equine anhidrosis is sudden, occasionally preceded by a period of excessive sweating, similar to our two patients, with case 2 preceded by a period of excessive sweating.

Some areas are spared from anhidrosis in horses and humans affected by the conditions. Area of retained ability to sweat in horses affected by equine anhidrosis include the areas beneath the mane, saddle and halter areas as well as the axillary, groin & perineal regions. In our patients, areas spared from anhidrosis were the face and in Case 1, the iodine-starch sweat test showed a small patch of retained sweating ability over the sternum at the level of the nipples (Figure. 1).

Why do some horses develop equine anhidrosis and our two patients AIGA when thermoregulatorily stressed before they are fit remain unexplained, beyond the rather general impression of an unconditioned system breaking down when stressed.

A few studies have suggested that there is a hereditary predisposition for equine hyperhidrosis, related to the genetics of the water channel, aquaporin 5 7. There is evidence that aquaporins may also be important in human sweating. In Japan,



Figure 1 Starch-iodine sweat test of Case 1.

there has been a report of a patient with neuromyelitis optica with narcolepsy and anhidrosis, secondary to hypothalamic lesions associated with anti-aquaporin 4 antibodies 8. This patient's somnolence and anhidrosis had resolved after treatment with systemic corticosteroids.

Management of equine anhidrosis rests on preconditioning horses before subjecting them to training, by exercise only during cooler hours and adequate post-exertion cooling (hosing and subsequently standing in front of a fan) and a diet with less heat-producing food (low fibre, low protein). This prudent preconditioning may also be effective for humans, and should probably be informed to institutions where physical training takes place.

Veterinarians have no definitive treatment for equine anhidrosis after it has occurred. In humans, use of high dose corticosteroids have been reported to be effective [9,10], with some authorities thinking that AIGA may be an autoimmune process [11], but we remain unconvinced if this is the therapeutic avenue to follow. This article, we hope, will encourage responses.

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