

Blue-colored sweating: four infants with apocrine chromhidrosis

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Apocrine chromhidrosis is a very rare, idiopathic disorder of the sweat glands characterized by the secretion of colored sweat. Because hormonal induction increases sweating, the symptoms of apocrine chromhidrosis usually begin after puberty. Although treatment may not be necessary in some cases, capsaicin cream and 20% aluminum chloride hexahydrate solution have been successfully used to treat patients requiring intervention. Here we report four cases with apocrine chromhidrosis. To the best of our knowledge, our patients are the youngest cases reported in the literature.

Key words: chromhidrosis, colored sweating.

Apocrine chromhidrosis is a very rare disorder of the apocrine glands characterized by the secretion of sweat ranging in color from blue to black. The color results from lipofuscin pigment, and the disease can involve sweat glands in the axillae, neck or areola. It is a chronic disorder that usually begins during puberty and may slowly regress with age. Diagnosis is based on clinical features; skin biopsy is required only if the diagnosis is questionable. There is no specific therapy.

Case Reports

Case 1

A 40-day-old boy was admitted to our clinic with a chief complaint of blue-colored perspiration, which had first been recognized two weeks earlier. He was born at term via cesarean section after an uneventful pregnancy. He was exclusively breastfed. Neither the patient nor the mother had used any medications. The mother nor any other family members had any known metabolic or inherited disorders. On physical examination, he appeared healthy with a normal height, weight, and head circumference. There was no sign of child abuse. He had a normal skin color and pattern. The systemic evaluation was completely normal. The only abnormal finding was blue-colored spots on his clothing,

although similar spots were not detected on his diaper. The colored perspiration was not associated with odor, fever, pain, redness or other constitutional symptoms. There was no history of trauma, significant medical illnesses, known allergies to any foods or medicines, prior hospitalization or surgery. A complete blood count and coagulation panel, liver function analysis, adrenocorticotropic hormone and cortisol levels, iron panel and chrome, copper and ferritin levels were within normal ranges. The color of fresh and as well as stored urine was normal. The urinary homogentisic acid level was within the normal limits. Skin bacterial and fungal cultures were negative. The patient had no history of contact with copper or other heavy metals, and laboratory tests for heavy metals were negative. After excluding all other probable causes of colored sweat, the patient was diagnosed with apocrine chromhidrosis. The family did not give consent for a skin biopsy.

Cases 2 and 3

Fourteen-month-old female twins presented with a two-month history of blue spots on their napes. They both had uneventful medical and family histories. Neither the patients nor the mother had used any drugs or health foods

except for multivitamins. There were blue-colored droplets on the patients' necks and napes as well as their underclothing (Fig. 1a-1b). On physical examination, they appeared healthy, with normal heights, weights and head circumferences. They had no cyanosis, edema, acne, unusual bruising, odor, fever or pain. The systemic examinations of the twins were completely normal. There was no history of any significant medical illnesses, known allergies to any foods or medicines, prior hospitalization, trauma, surgery, dietary changes (blue-colored food) or application of any external substances (ruling out pseudochromhidrosis). The complete blood counts, coagulation panels, liver function analyses, adrenocorticotropic hormone and cortisol levels, iron panels and chrome, copper and ferritin levels were all within the normal ranges. The color of fresh as well as stored urine was normal for both children. The urinary homogentisic acid levels were within the normal limits. Skin bacterial and fungal cultures were negative. The patients had no history of contact with copper or other heavy metals, and the laboratory tests for heavy metals were negative. Based on the patients' histories and clinical examinations, a diagnosis of apocrine chromhidrosis was made. The family did not give consent for skin biopsies.

Case 4

A 13-month-old female infant was referred to our clinic for evaluation of colored sweat. A dark blue-colored sweat had first been detected six months earlier. She was otherwise healthy. She had been using vitamin D, but no other supplements or drugs, from the 15th day of

life. The mother had not noticed any unusual color or odor to her urine. The family history was unremarkable for any inherited metabolic disease that could be responsible for the secretion of colored sweat. On examination, the only significant finding was dark blue-colored spots on the patient's neck and the axillary areas of her clothing (Fig. 2). There was no history of significant medical illnesses, known allergies to foods or medicines, prior hospitalization, trauma, surgery, dietary changes (blue-colored food) or application of any external substances. A complete blood count and coagulation panel, liver function analysis, adrenocorticotropic hormone and cortisol levels, iron panel and chrome, copper and ferritin levels were within normal ranges. The color of the patient's fresh and stored urine was normal. The urinary homogentisic acid level was within the normal limits. Skin bacterial and fungal cultures were negative. The patient had no history of contact with copper or other heavy metals, and the laboratory tests for heavy metals were negative. With the reference of our other patients with apocrine chromhidrosis, and by excluding all other probable causes of colored sweat, the diagnosis of apocrine chromhidrosis was made.

Discussion

Chromhidrosis is a very rare idiopathic disorder of the eccrine or apocrine sweat glands characterized by yellow, blue, green or black pigmented secretions¹. The color of perspiration varied from blue to dark blue in all of our patients. In most cases, chromhidrosis appears to be unrelated to diet



Fig. 1a. Case 2: blue-colored staining on the neck of the shirt.



Fig. 1b. Case 3: blue colored stain on the neck of the shirt.



Fig. 2. Case 4: blue-colored staining on the underclothing.

or systemic or metabolic abnormalities. Several etiologies for chromhidrosis exist. True eccrine chromhidrosis is associated with water-soluble pigments excreted by the eccrine glands. Pseudochromhidrosis is the result of colorless perspiration mixing with external chromogens, such as dyed clothing². Several extrinsic causes may be involved, such as chromogenic bacteria and dyes such as bromophenol blue and copper salts³. Differential diagnoses include hyperbilirubinemia, *Pseudomonas* infection, bleeding diathesis (red-colored perspiration and hematohidrosis), alkaptonuria (ochronosis) and some types of poisoning^{4,5,6}. In our four patients, we excluded the use of external colored agents, systemic and metabolic factors and child abuse as possible causes.

Apocrine chromhidrosis results from a higher than normal concentration of lipofuscin granules in the apocrine glands⁷. Apocrine sweat glands are located in the axillae, anogenital skin, mammary areolae and the skin of the trunk, face and scalp. A diagnosis is based primarily on the patient's history and physical examination. If the diagnosis is unclear, the presence of lipofuscin granules in apocrine cells in a skin biopsy can be used to confirm the diagnosis. In a patient with apocrine chromhidrosis, the apocrine glands appear normal in size and morphology, but the number of glands varies. An increased number of yellow-brown lipofuscin granules are observed in the cytoplasm of secretory cells. The lipofuscin granules are responsible for the pigmented sweat⁸. Lipofuscin is a yellow pigment that is not specific to apocrine glands. In apocrine chromhidrosis, the lipofuscin is

present in a higher concentration or higher state of oxidation than in normal secretions, adopting a darker coloration, such as blue, green or black¹. After excluding the other etiologies of colored perspiration, we were confident about the diagnosis of apocrine chromhidrosis. However, in these cases, the families did not give consent for an invasive test such as a skin biopsy.

Carman et al.⁹ reported a 9-month-old girl with infant chromhidrosis. Physical examination and signs were similar to those of our patients. Griffith¹⁰ reported an 11-year-old girl with isolated areolar apocrine chromhidrosis. To the best of our knowledge, our cases are the youngest patients with apocrine chromhidrosis to be reported in the literature. The presence of colored sweat at a prepubertal age is another striking feature of our cases.

Satisfactory therapy for this condition remains a challenge. Success has been reported with capsaicin cream and a 20% aluminum chloride hexahydrate solution¹¹. Patients may become symptom-free after manual or pharmacologic emptying of the glands secondary to depletion of the pigment from the apocrine glands, but the pigment reaccumulates within 48 to 72 hours^{11,12}. Botulinum toxin type A has been shown to be effective for facial chromhidrosis^{13,14}. Although apocrine glands are thought to be unresponsive to cholinergic stimulation, some authors have demonstrated a response to local administration of cholinergic compounds¹⁵. Our patients did not use any of these medications.

In summary, we report four cases of apocrine chromhidrosis in infants. Our patients are unique, because in each case the secretion of colored sweat was detected at a very young, prepubertal age. In fact, to the best of our knowledge, these cases are the youngest patients to be reported in the literature. By presenting these cases, we would like to draw attention to the fact that chromhidrosis can occur in young, prepubertal children.

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