Commentary: Controversies in Thrombosis and Hemostasis Part 1—Hematidrosis: "Blood, Sweat and Fears" or A "Pigment of Fertile Imaginations?"

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Now that we have captured your attention with our "snappy" title, we should apologize if the title suggests we are trivializing a potentially serious, and certainly emotive and controversial topic. Hematidrosis (also called blood sweat), is reported to be a very rare condition in which a human "sweats blood." Hematidrosis is purportedly caused by the rupture of capillaries that feed the sweat glands, causing them to include blood elements in the secreted sweat. The condition most frequently seems to occur at times of substantial physical or emotional stress and fear. In the Christian faith, Jesus is said to have undergone hematidrosis before the crucifixion, during the Agony in the Garden (New Testament; Luke 22:44). Hematidrosis has also been proposed as a possible explanation for "stigmata."

Hematidrosis is a condition that only a portion of medical professionals believe in. Certainly, as a condition, there is both plausibility and plausible deniability, with many alternate explanations, potentially including self-inflicted or other injuries (e.g., Munchausen's syndrome or Munchausen's by proxy). Neither of the authors to the current commentary, also guest editors to the current issue of the journal, have ever been personally involved in such a case, although perhaps pertinent, in terms of "similar" physical etiology, we have seen cases of "skin hemorrhages," albeit always associated with hemorrhagic conditions, either congenital or acquired.

Nevertheless, a proposed case of hematidrosis is described in this issue of the journal,[1] and the authors of that case report certainly believe they are describing a case of hematidrosis. This case report was sent to several hematologists for review ahead of publication, and skepticism was expressed, having like us, never seen such a case during their careers. Some of the potential differential diagnoses and concerns expressed in these reviews are identified in ►**Table 1**. Despite this healthy skepticism, we decided to permit the publication of this case in this issue of the journal. The intention here is to permit potential future dialogue not only on hematidrosis but also on other potential controversies in thrombosis and/or hemostasis, as the situation may present in the future. Our take on the topic of hematidrosis follows.

A search of the literature uncovers only sporadic publications on the topic, with the search term "hematidrosis" or "haematidrosis" in PubMed uncovering a total of only 22 reports, with an additional report on "hematidrosis" in Russian. Of the total 23 reports, 10 were in a non-English language source, and another 3 appeared in English within Indian journals (i.e., tallying more than half of the total). The majority of the publications appear in dermatological or pediatric journals, with one article published in a psychological journal, and only a single report published in a hematological journal. Among some recent articles were a case published in 2017 involving a 10-year-old girl with mixed anxiety and depressive disorder,[2] another case of a young girl with epilepsy who was successfully treated using an anti-epileptic drug (i.e., oxcarbazepine) published in 2015,[3] and a case published in 2013 in an American Society of Hematology (ASH) Image Bank article.[4] No other publication in the hematology literature. Not many publications in English geography journals. Apart from two short (single page) publications in Blood[4] and the British Medical Journal (BMJ),[5] no publication in a high-impact journal is available. Of further interest, the BMJ report (in a newborn) was published exactly 100 years ago![5] No surprise, then, that hematologists, particularly in developed English geography locations, may not believe hematidrosis to be real.

Why is hematidrosis plausible? As mentioned, hematidrosis is reportedly caused by the rupture of capillaries that feed

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Table 1 Plausible deniability for hematidrosis[a]

• Rare; most of us have never seen such a case
• Unconscious "avoidance" of pathologies for which there is no clear cause or treatment
• Most patients are under stress (therefore high probability of self-inflicted wounds)
• Munchausen's syndrome or Munchausen's by proxy
• Most reports are from older literature, in foreign language, in nonhematology journals, and there is a lack of publications in high-quality journals (limited "trust" factor for modern day hematology workers)
• Self-inflicted injuries/ecchymoses never occur at sites not reachable by the hands, such as in between the scapula
• Most reported cases seem to be from tropical areas–significance?
• Insufficient scientific proof that hematidrosis is an entity

[a]Including alternate explanations offered by reviewers of the case study.

the sweat glands, particularly under periods of stress, causing them to include blood elements in the secreted sweat. This is perhaps why most hematidrosis cases are published in dermatology journals. We are not dermatologists, nor experts in sweat gland physiology. However, that capillaries feed the sweat glands, and may rupture under periods of stress, certainly sounds plausible to us. For the case published in Blood: "Physical examination showed no signs of self-inflicted lesions. All laboratory, image, and coagulation tests, including platelet aggregation, were normal. More than 30 bleeding episodes were witnessed during hospitalization and samples of the fluid contained all normal blood cells."[4] That case was successfully managed using propranolol. For another reported case of a 12-year-old girl: "During hospitalization she had more than 10 instances of spontaneous intermittent bleeding per day that was evidenced by almost all of our on-duty doctors and nurses."[6] Again, laboratory testing failed to identify any hemostatic or other defect, and the case was again successfully managed using propranolol.

Importantly, hematidrosis is not the only kind of "blood leakage" phenomenon. For example, hemolacria is a condition in which tears are partially composed of blood, and otorrhagia represents bleeding from the ear. A PubMed search of "hemolacria" and "haemolacria" identified 17 articles, unsurprisingly with most in ophthalmology journals. More interestingly, a PubMed search of "otorrhagia" uncovered 69 publications, and we suspect that otorrhagia is more common than either "hematidrosis" or "hemolacria," and thus more plausibly acceptable to workers in hematology.

Why is hematidrosis plausibly deniable? Any rare condition, should it truly exist, will rarely be seen by workers in the field. Neither of the authors of this commentary nor the reviewers to the case study have ever personally seen a typical case of hematidrosis. Should we then send the case for review by someone who has seen such a case? Naturally, given the proposed rarity, that would be difficult; however, we would suspect in this situation that the reviews would be more favorable at least. The literature on hematidrosis is scarce, not generally published in the English literature and in high impact journals. Working in developed countries, both authors have personally not seen many of the more established rare bleeding disorders, and yet, we believe they exist. Nevertheless, with hematidrosis, there are also plausible alternative diagnoses or explanations (►**Table 1**). So, we can personally never be sure of the truth or otherwise of such a diagnosis in any given individual.

We hope that this commentary, and the accompanying case report, encourage further dialogue, not only in regards to hematidrosis but also as related to other potential controversies in thrombosis and/or hemostasis, as the situation may present in the future.

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