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Commentary on: Thogmartin JR, Wilson CI, Palma NA, Ignacio SS, Shuman MJ, Flannagan LM. Sickle cell trait-associated deaths: a case series with a review of the literature. J Forensic Sci 2011;56(5):1352–60.

Sir,

We read with great interest the article by Thogmartin et al. (1) describing a case series of sickle cell trait (SCT)-associated deaths with a review of the literature. It is noticeable that a great majority of the 16 documented cases were deaths arising in a context of sport activity (i.e., exercise-related) or physiological stress, "which lasted from several minutes to several hours" (1, p. 1356). The same was found in another case series by Scheinin and Welti (2). Without criticizing any part of these articles, and based on our own forensic experience, we would like to insist on the fact that sudden death may occur also in SCT patients far away from any physical activity (3).

In 2002, a Black-African (Cameroon) 30-year-old clergyman suddenly collapsed at the end of a normal meal, far away from any air travel, physical activity, or physiological stress. A short resuscitation was realized, without any success. His height was 1.82 m and he weighed 72 kg (body mass index = 22). Autopsy revealed no particular injuries. Heart showed left ventricle concentric hypertrophy (maximal thickness: 1.7 cm; heart weight: 415 g). Microscopic examination of the viscera showed the presence of an intravascular sickling associated with focal acute myocardial ischemia, interpreted as the cause of this sudden death. Vascular congestion with sickled erythrocytes was found in many organs (brain, lungs, liver, spleen, kidneys, testicles, pancreas, tongue, adrenal glands, and prostate). Coronary arteries were free of any atherosclerosis lesion. Hemoglobin electrophoresis (on peripheral blood) revealed HbS = 58%, HbA1 = 37.6%, and HbA2 = 4.4%. Extensive toxicological testing failed to show the presence of alcohol and drugs.

If all the cases presented by Thogmartin et al. (1), as well as other case reports of sudden deaths associated with SCT (2–8), were related to sport activities or physiological stress, our forensic case clearly confirms that such sudden death may also occur during quiet and rest periods.

As a matter of fact, in our case, usual complication factors (dehydratation, hyperthermia, hypoxic states, poor physical

conditioning, and comorbidity) (9,10) were not found; in fact, no one cause of acute sickling was found.

Last, our case highlights the fact that intravascular sickling as an underlying cause of death requires systematic histological examination after any autopsy, particularly in a forensic context (11).

References

- Thogmartin JR, Wilson CI, Palma NA, Ignacio SS, Shuman MJ, Flannagan LM. Sickle cell trait-associated deaths: a case series with a review of the literature. J Forensic Sci 2011;56(5):1352–60.
- Scheinin L, Wetli CV. Sudden death and sickle cell trait. Medicolegal considerations and implications. Am J Forensic Med Pathol 2009;30(2):204–8.
- 3. Rosenthal MA, Parker DJ. Collapse of a young athlete. Ann Emerg Med 1992;21:1493–98.
- Eichner ER. Fatal collapse in athletes with sickle cell trait. Sports Med Digest 2000;22:109–18.
- Pretzlaff RK. Death of an adolescent athlete with sickle cell trait caused by exertional heat stroke. Pediatr Crit Care Med 2002;3:308–10.
- Wirthwein DP, Spotswood SD, Barnard JJ, Prahlow JA. Death due to microvascular occlusion in sickle-cell trait following physical exertion. J Forensic Sci 2001;46:399–401.
- Thogmartin JR. Sudden death in police pursuit. J Forensic Sci 1998;43:1228–31.
- 8. Kerle KK, Nishimura KD. Exertional collapse and sudden death associated with sickle cell trait. Military Med 1996;161:766–7.
- McCormick WF. The pathology of sickle cell trait. Am J Med Sci 1961;241:329–36.
- Lorin de la Grandmaison G. Sickle cell hemoglobinopathy and sudden death. In: Plasmar RL, editor. Focus on sickle cell research. New York, NY: Nova Science Publishers, 2004;125–30.
- Lorin de la Grandmaison G, Charlier P, Durigon M. Usefulness of systematic histological examination in routine forensic autopsy. J Forensic Sci 2010;55(1):85–8.

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