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The truth comes out of children's mouths

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The truth comes out of children's mouths.

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Reims, July 20th 2021

Dr G Agnelli
Editor in chief
European journal of Internal Medicine

Dear Dr Agnelli

Please find enclosed of a manuscript (internal medicine flashcard; one figure) entitled : “The truth comes out of children’s mouths.”, by Bénédicte Decoudier and colleagues, that we would like to submit to your consideration for publication in « **European journal of Internal Medicine** ». This typescript has not been published elsewhere and is not submitted in another journal. We think that this typescript is quite didactic and that it could be of interest for the readership of « **European journal of Internal Medicine** ». It showed unexplained patchy darkish pigmented macules of the oral mucosa and unnoticed hyperpigmentation of the palmar creases and of the dorsum of the hands, which did not allow to diagnose Addison Disease before the onset of Adrenal Crisis in a high skin phototype teenager. All authors had full access to all the data and the corresponding author had the responsibility for the decision to submit this work for publication with the agreement of all the co-authors. Please note that both authors equally contributed to this work. Thank you for considering our paper.

Yours sincerely,

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14 The truth comes out of children's mouths.
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17 (398 words)
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22 Bénédicte Decoudier^{1*}, Yohan N'Guyen^{2*}.
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Main section (174 words)

A 16 year-old patient of Hawaiian ancestry was admitted for confusion in April 2018. He had no relevant past medical history except he went to the dentist few months ago for patchy darkish pigmented macules of the oral mucosa (Figure 1A). His mother reported that he complained of pharyngitis and diarrhoea since one week. Blood pressure, body temperature and heart rates at admission were 80/50 mm Hg, 38.3°C and 163 beats per min respectively. Oxygen saturation and glycemia were estimated to 100% room air and 3.2 mmol/L using pulse oximeter and glucometer respectively. The patient did not exhibit neck stiffness, physical examination was normal except localised skin pigmentation disorders (Figure 1B). Screening of drugs of abuse or alcohol yielded negative results. White blood cells count was increased with left shift (21 000/mm³, 68% neutrophils) and haemoglobin level was 15.9g/dL. Platelets count was within normal range and Procalcitonin value was 4.78ng/mL. Blood chemistry results before and after crystalloid fluid resuscitation started immediately after admission, are shown in Figure 1C. What is the most plausible diagnosis?

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Discussion section: (225 words)

Blood cortisol level at 08.00 am was 57 nmol/L (normal value >275nmol/L) and antiadrenal antibody test was positive at 1 in 100 dilutions (normal value ≤ at 1 in 10 dilutions), in line with the diagnosis of Addison's disease (AD). Moreover, Adrenal crisis was confirmed here by the presence of shock with inappropriate natriuresis [1]. Interestingly, hypoglycemia and hyperkalemia were not constantly present during Adrenal crisis, as suggested by the blood levels of glucose or potassium here. The outcome was favourable (normal consciousness and temperature) after hydrocortisone supplementation.

This case illustrates that a subset of AD are still revealed by adrenal crisis, because the onset of asthenia (which is the key symptom of AD) is so insidious that it is not reported by the patient or his relatives [1]. Hyperpigmentation of the palmar creases and of the dorsum of the hands with accentuation over the interphalangeal joints (Figure 1B), which is one of the earliest symptom of AD in connection with Adenocorticotrophic Hormon and Proopiomelanocortin secretion [2], went unnoticed (as well as vitiligo of the thigh, Figure 1B) in this patient with high skin phototype. Conversely, darkish pigmentation of the oral mucosa (Figure 1A) remained unexplained before the onset of adrenal crisis. Except AD, the main diagnoses to consider in case of patchy darkish pigmentation of the oral mucosa are Peutz-Jeghers Syndrome and Laugier-Hunzicker Syndrome [3].

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Contributors: BD and YNG take care of the patient. YNG wrote the manuscript that was critically revised by BD.

Declaration of interest: the authors report no conflict of interest.

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Figure caption:

A. Patchy darkish pigmented macules of the oral mucosa (lower lips). **B.** Localised skin pigmentation disorders. Upper panel: hyperpigmentation of the palmar creases. Lower left panel: vitiligo of the thigh. Lower right panel: hyperpigmentation of the dorsum of the hands with accentuation over the interphalangeal joints. **C.** Blood chemistry results before and after crystalloid fluid resuscitation started immediately after admission. Admission time is quoted as Hour 0 (H0). H1: 1 hour after admission. H4: 4 hours after admission.

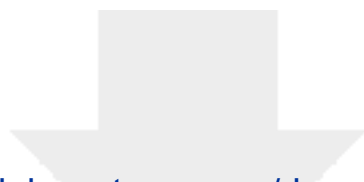


C

Crystalloid fluid resuscitation →

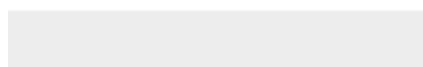
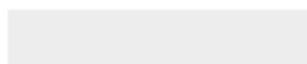
Chemistry tests	Admission (H0)	H1	H4
Blood sodium level (mmol/L)	127		129
Blood potassium level (mmol/L)	4.3		4.6
Blood creatinine level ($\mu\text{mol/L}$)	160		99
Venous lactate level (mmol/L)	2.92		0.77
Single Urine Sample sodium level ((mmol/L)		43	





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