Images in Rheumatology

Raynaud Phenomenon With Lingual Involvement in a 6-year-old Female

Julia A. Hunter, MB BCH BAO; Deborah M. Levy , MD, MS, FRCPC; Roberta A. Berard , MD, MSc, FRCPC, Division of Pediatric Rheumatology, Western University, London, Ontario, Canada. Address correspondence to Dr. R. Berard, Children's Hospital, London Health Sciences Centre, London, ON N6A5W9, Canada. Email: Roberta.berard@lhsc.on.ca. Written and verbal consent were provided by the patient's mother to publish this case. Ethical board approval was not required according to the authors' institution, as this was an image for which we have received consent from the patient's family.

Raynaud phenomenon (RP) may affect the tongue as has previously been reported in cases of secondary RP^{1,2,3}. We are not aware of lingual involvement being previously described in individuals with primary RP, particularly in children.

A 6-year-old prepubertal, previously healthy female was referred for rheumatological assessment. Following cold exposure, she described a typical triphasic RP of the fingers. After initial episodes involving the digits, she subsequently experienced episodes of nonpainful blanching of the anterior left one-third of the tongue associated with tingling. These episodes were precipitated by exposure to cold weather with an open mouth, but not ingestion of cold liquids or foods. Exact episode duration is unknown,

though there was rapid resolution after mouth closure. There was no history of lingual ulceration or other associated symptoms. Lingual and digital episodes did not occur concurrently (Figures 1A,B).

History and examination, including nailfold capillaroscopy were normal. Laboratory workup revealed positive antinuclear antibodies (ANA; titer of 1:2560 in a mitotic pattern). Extractable nuclear antigen (ENA) panel was negative. The patient was evaluated 9 months later, with repeat ANA revealing a titer of 1:160 (mitotic). ENA remained negative. There was no evolution in symptoms to suggest secondary RP.

Retrospective data have shown that ANA positivity in chil-

Figure 1. A 6-year-old female with a history of blanching and tingling of fingers developed anterior predominantly left-sided tongue discoloration consistent with lingual RP. (A) Superior surface of the tongue. (B) Typical blanching of the fingers as seen in RP. RP: Raynaud phenomenon.

dren with RP is associated with evolution to involve other features of a systemic autoimmune disease⁴. Despite not having secondary RP at the time of assessment, given this patient's elevated ANA and unusual lingual presentation, she will be followed closely for development of an associated rheumatic disease.

REFERENCES

1. Lioger B, Diot E. Raynaud's phenomenon of the tongue: uncommon presentation of a classical sign. *QJM* 2012;106:583-4.
2. Katada Y, Tanaka T. Lingual Raynaud's phenomenon. *N Engl J Med* 2012;366:e12.
3. Cohen JC, Palomba ML, Morris LG. Raynaud's phenomenon of the tongue. *J Rheumatol* 2013;40:336.
4. Falcini F, Rigante D, Candelli M, Martini G, Corona F, Petaccia A, et al. Anti-nuclear antibodies as predictor of outcome in a multi-center cohort of italian children and adolescents with Raynaud's phenomenon. *Clin Rheumatol* 2015;34:167-9.