New evidence that chronic ulcerative stomatitis is an autoimmune disease

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In the first study investigating the origins of a little-known condition called chronic ulcerative stomatitis (CUS), researchers at Tufts University School of Dental Medicine provide evidence that an autoimmune response contributes to the painful oral sores that characterize the disease. The study findings support the classification of CUS as a new autoimmune disease.

Chronic ulcerative stomatitis is characterized by painful, recurring sores in the mouth. Thus far, it has been diagnosed most frequently in white women in their 40's and 50's and may appear similar to oral erosive lichen planus. Only 39 cases of CUS have been reported in the English-language medical literature since it was identified as a clinically distinct condition in 1989, but it is likely under-diagnosed because of low awareness among clinicians and the extensiveness of the testing that would confirm its presence.

"Currently, diagnosing CUS requires a surgical biopsy which then must be sent to an outside lab for special processing for immunofluorescence microscopic examination. Accurate diagnosis is important because the usual treatment option for immunologically-mediated diseases, corticosteroids, is often not effective in treating CUS," said senior author, Lynn Solomon, DDS, MS, associate professor in the department of oral and maxillofacial pathology at Tufts University School of Dental Medicine (TUSDM).

"In previous studies, we identified that CUS patients had specific

autoantibodies –antibodies produced by an immune response to the body's own tissue – but we weren't sure whether these autoantibodies were contributing to CUS or part of a benign biological process. In this study, we determined that autoantibodies fulfill the criteria of pathogenetic antibodies and do contribute to the disease," she continued.

In this in vitro study, the researchers applied antibodies from four CUS patients to Human Skin Equivalents (HSEs), a three-dimensional model of skin tissues. At low concentrations, the CUS antibodies appeared to have no effect. At higher concentrations, however, the researchers reported complete detachment of the surface layer of tissue, known as epithelium.

The researchers found that the CUS autoantibodies do not cause damage to the surface epithelial cells, but cause a change in the cell-binding proteins that allow the surface layer to attach to the connective tissue beneath them. This weakened cohesion results in breakdown of the tissue, which would result in the sores that characterize CUS.

"In our future research, we would like to gain a better understanding of the mechanisms linking the autoimmune response to ulcerative sores so that we can optimize approaches to managing the condition. Additional data will help us evaluate hydroxychloroquine therapy, an antimalarial drug used off-label that provides relief in many cases, but which is not well-tolerated by some patients and which may have serious side effects," said Solomon.

More information: Carlson MW, Garlick JA, Solomon LW. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. "Chronic ulcerative stomatitis: Evidence of autoimmune pathogenesis" Published online April 4, 2011, doi: 10.1016/j.tripleo.2010.12.020

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