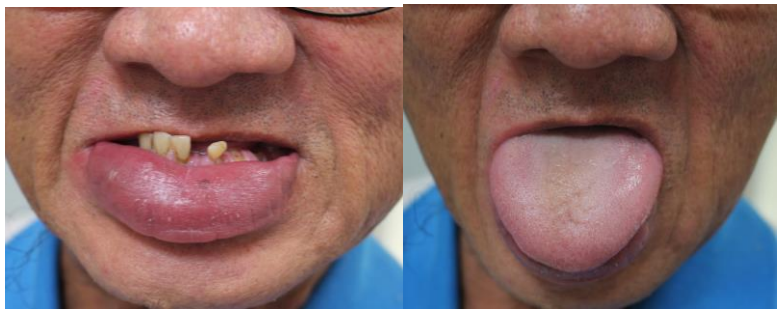
Case 14.1
A 61-year-old man, from Nan.
Chief complaint: Lower lip swelling for 1 year



Present illness: He had persistent lower lip swelling wi
dryness, pain or burning sensation for a year. His sympt
was not related to any condition. He denied fever, any
gastrointestinal symptom and also facial palsy. There wa
history of applying irritant agents, trauma, or filler inject

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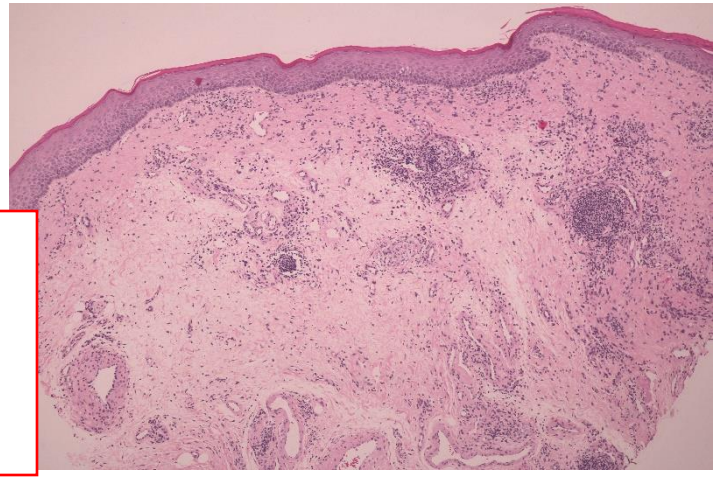
Past history
No known underlying disease

Physical examination
HEENT: not pale conjunctiva, anicteric sclera
Lymph node: no palpable lymph nodes
Lung: good air entry, clear
Heart: regular, no murmur
Abdomen: not tender, no hepatosplenomegaly
Neurological: no muscle weakness, no facial palsy

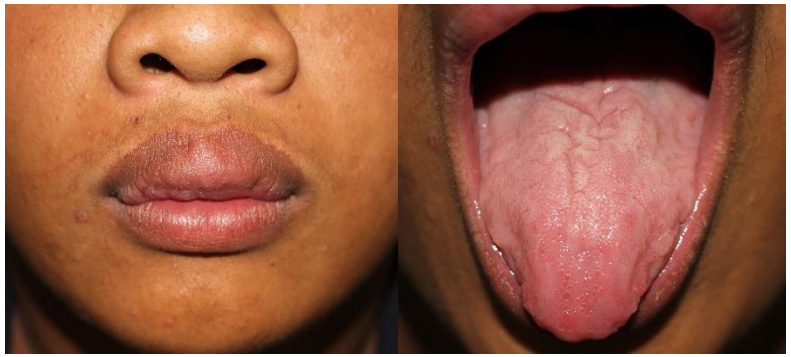
Skin examination
Lower lip swelling
No fissuring of tongue
No gingival abnormalities

Histopathology (Case14.1: S15-20540A, Lower lip)



- Perivascular and nodular inflammatory cell infiltrate of lymphocytes admixed with a few histiocytes, some of which tend to form small tuberculoid granuloma

Case 14.2
A 20-year-old man, from Kamphaengphet
Chief complaint: Swollen lip for 4 months

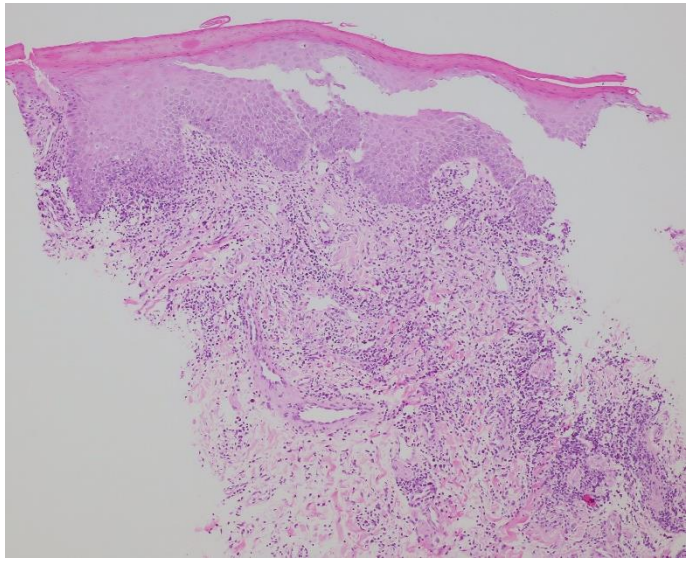


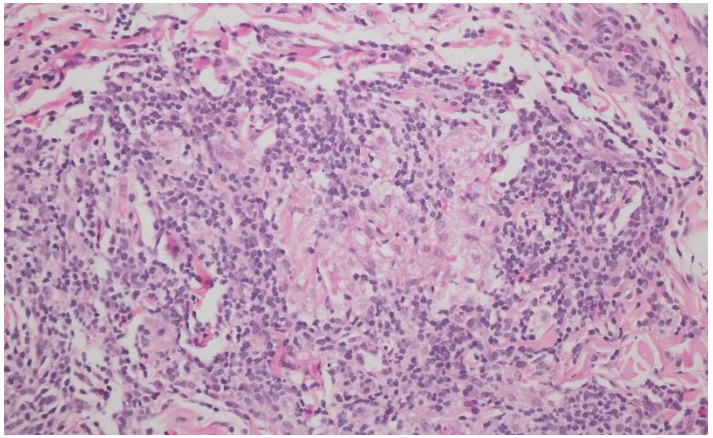
Present illness: He noticed swollen lip without any symptoms for 4 months. Initially, the symptom was wax and wane, and seems to relate with fermented food consumption. Therefore, he tried avoiding fermented food, but there was no improvement.
Later, his upper lip became persistently swollen.
He had no fever, no gastrointestinal symptoms, or facial nerve palsy.
There was no history of irritants application, local trauma, or filler injection.
He also denied eating raw food.

Past history
No known underlying disease

Physical examination
HEENT: not pale conjunctiva, anicteric sclera
Lymph node: not palpable
Lung: normal breath sound, no adventitious sound
Heart: regular, no murmur
Abdomen: not tender, no hepatosplenomegaly
Neurological: no muscle weakness, no facial palsy
Skin examination
Markedly erythema and swollen upper lip
Fissuring of tongue
No gingival abnormalities

Histopathology (Case14.2: S15-17505A, Lower lip)





- Confluent parakeratosis and epidermal hyperplasia
- Perivascular and nodular inflammatory cell infiltrate of lymphocytes admixed with a few histiocytes, some giving the features of small tuberculoid granuloma

Diagnosis: Cheilitis granulomatosa

Treatment: Intralesional injection of Kenacort A (10mg/ml) 0.4 ml in both cases.

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Discussion:

Cheilitis granulomatosa or Miesher's cheilitis is a rare inflammatory disorder with clinical characteristics of chronic swollen lips.[1]

Cheilitis granulomatosa was first described in 1945 by Miescher. There is no age, racial, or sexual predilection in cheilitis granulomatosa.[2] The incidence is about 0.08% in general population[3]

Clinical presentations of cheilitis granulomatosa are chronic, recurrent, and painless swollen of one or both upper and lower lip due to granulomatous inflammation, with normal tongue.[4, 5] It can occur by itself or as the most common finding in Melkersson-Rosenthal syndrome, the granulomatous inflammatory disorder with classical triad of lip swollen, plicated tongue (lingua plicata) or fissured tongue, and recurrent facial palsy.[3]

Both syndrome are considered subsets of orofacial granulomatosis group (OFG), which is an entity of non caseating granulomatous inflammation in absence of systemic disease with orafacial swelling.[6]

The etiology of cheilitis granulomatosa is unknown, but some hypotheses suggest it may be associated with allergic reactions to cinnamon and benzoates, chronic orodental infection, or autoimmune process, such as Crohn's disease and sarcoidosis.[3]

Histopathology shows non-caseating granulomas without foreign materials and any organism.[3]

There is no specific treatment for cheilitis granulomatosa. But from many studies, corticosteroids, topical or intralesional injection (triamcinolone acetonide 40 mg/ml weekly for 3 weeks), are effective in reducing lip swelling with long term remission 10 -12 months.[7] Other treatment options that have been reported as small series and case reports, are thalidomide, minocycline, metronidazole, clofazimine, roxithromycin, dapsone combine with intralesional steroid injection and more recently, monoclonal antibodies against TNF-α, adlimumab.[6, 8-11] Cheiloplasty, surgical treatment for cosmetic problems due to fibrosis in long standing case.[12] Benzoate and cinnamon-free diet also showed satisfactory result in cheilitis granulomatosa even in patient with negative patch test result to cinnamaldehyde and benzoate.[13, 14]

Both of our patients presented chronic persistent lip swelling without any associated symptoms. Their physical examinations were unremarkable, except their swollen lower lips and fissured tongue in the second case. Histopathological findings showed non-caseating granulomas that compatible with cheilitis granulomatosa.

Despite of fissured tongue in the second case, criteria diagnoses of Melkersson-Rosenthal syndrome were not complete. Both of them were treated with intralesional injection of corticosteroids.

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