

# Late recurrence in surgically managed pediatric atypical mycobacterial lymphadenitis: A case report and review of the literature

Katerina Green<sup>1</sup>  | Alexa Denton<sup>2</sup> | Jeffrey Graves<sup>3</sup>  | Joshua Wiedermann<sup>4</sup>

<sup>1</sup>Department of Plastic and Reconstructive Surgery, University of Pittsburgh School of Medicine, Pittsburgh, Pennsylvania, USA

<sup>2</sup>Herbert Wertheim College of Medicine, Florida International University, Miami, Florida, USA

<sup>3</sup>Alix School of Medicine, Mayo Clinic, Rochester, Minnesota, USA

<sup>4</sup>Department of Otolaryngology-Head and Neck Surgery, Mayo Clinic, Rochester, Minnesota, USA

## Correspondence

Joshua Wiedermann, Department of Otolaryngology-Head and Neck Surgery, Mayo Clinic, 200 1st St SW, Rochester, MN 55905, USA.

Email: [Wiedermann.Joshua@mayo.edu](mailto:Wiedermann.Joshua@mayo.edu)

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## Abstract

**Objective:** The purpose of this study is to identify existing literature on recurrent atypical mycobacterial cervicofacial lymphadenitis to augment our understanding of a unique patient who presented to our tertiary-care center 5-years posttreatment with recurrence following curettage.

**Data Sources:** OVID Medline, Scopus, and Web of Science.

**Methods:** A literature search was conducted yielding 49 original articles which were screened twice by two independent reviewers resulting in 14 studies meeting inclusion criteria for data extraction using Covidence software. Two independent reviewers extracted data on recurrence of atypical mycobacterial cervicofacial lymphadenitis and consensus was reached on data points from all included studies.

**Results:** This study illuminated the paucity of recurrence reporting in the literature regarding atypical mycobacterial lymphadenitis. Sixteen studies identified in our review included discussions on recurrence with few elaborating beyond the rate of recurrence to describe their management. Fourteen out of sixteen studies provided recurrence rates for their cohort, 11 out of 14 specified the initial treatment modality, and only five out of eight studies that described initial treatment with surgery differentiated recurrence rates between complete and incomplete excision. The mean length of follow-up in the included studies was 20 months. There was one previously reported case of late recurrence at 5-years.

**Conclusions:** We identified few reports that discussed the management of recurrence of atypical mycobacterial cervicofacial lymphadenitis. There was minimal data on recurrence rates between surgical treatment modalities. The case discussed in our study showcases that treatment with curettage has the potential to present with late recurrence.

## KEYWORDS

atypical mycobacteria, case report, curettage, nontuberculosis mycobacteria, pediatric lymphadenitis, pediatric lymphadenopathy, recurrent nontuberculosis mycobacterial infection

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## INTRODUCTION

Atypical, nontuberculosis mycobacterial (NTM) cervicofacial lymphadenitis is an infection that typically occurs in immunocompetent children between the ages of 1 and 5 years old. Cervicofacial lymphadenitis most commonly presents with local signs and symptoms, including painless neck swelling progressing to violaceous discoloration, fluctuance, and eventually drainage.<sup>1</sup> Diagnosis of NTM is controversial and was reviewed in a 2018 systematic review by Willemse et al.<sup>2</sup> They described the literature regarding the diagnosis of NTM lymphadenitis to be scarce and lacking high methodological quality. There is no gold standard for diagnosis of NTM, therefore diagnosis typically relies on the clinical judgment of the treating clinician. A weakly positive purified protein derivative (PPD) skin test can be predictive of NTM infection. Other purified protein-derivative tests such as interferon- $\gamma$  release assays (quantiferon gold) appear to have good sensitivity and specificity preoperatively for mycobacterial tuberculosis, but its utility in identifying NTM is limited.<sup>2</sup>

Imaging is not necessary for routine evaluation for suspected NTM lymphadenitis. Imaging studies such as ultrasound or CT scan are useful to exclude alternative differential diagnoses of cervical adenitis. Imaging studies often show necrosis or fistulization of the lymphadenopathy that usually involves perifacial and/or intraparotid lymph nodes in cases of NTM lymphadenitis. It may be instinctive for physicians to treat and diagnose with incisional biopsy, but this should be avoided when possible as incision and needle aspiration can lead to fistulization and spread of disease. This may also blur planes of dissection adding to the risks of definitive management. Excisional biopsy is the standard of care when there is clinical suspicion of NTM lymphadenitis for diagnostic and curative intent.<sup>2</sup> Additionally, the diagnosing physician should consider other infectious etiologies before committing to surgical excision.

The management of NTM lymphadenitis is also controversial and several treatment options have been adopted and studied over time. The literature has shown that among the options for management of NTM cervicofacial lymphadenitis, cure rates and treatment side effects are highly variable.<sup>3</sup> Previously utilized and researched treatment options have included nonintervening "Wait-And-See" methods, antibiotic treatment regimens, and varying degrees of surgical excision. Cohort studies by Lindeboom et al.<sup>4</sup> have compared treatment outcomes along with morbidities associated with different modalities of treatments. When comparing conservative Wait-And-See therapy to antibiotic treatment with clarithromycin and rifabutin in a randomized control trial, they found no statistical difference in clinical improvement rates, lasting 36–40 weeks in both groups, indicating no superiority of antibiotic treatment over non-intervention.<sup>4</sup> Although the Wait-And-See approach leads to comparable resolution, it is generally discouraged due to the associated morbidity caused by a draining lesion on the face or neck for several months which can be intolerable by the patient or parent. In a randomized control trial comparing the same antibiotic regimen to lymph node excision, in a cohort of 100 participants using

intention-to-treat analysis they again found that surgery was a more effective treatment than antibiotic therapy. In this study, they also considered incidence of adverse effects of antibiotics and surgical complications.<sup>5</sup> They described a surgical complication rate of 28%, including wound infections and one permanent facial marginal branch injury. They also brought attention to the high rate of adverse effects with the antibiotic regimen of 78%, including four patients who subsequently discontinued treatment.<sup>5</sup> In a following study, esthetic scar-scores were compared between antibiotics and surgical excision. The median esthetic outcome in surgical patients was found to be significantly better than the group treated with antibiotics alone.<sup>6</sup>

Studies discuss the use of varying surgical treatment modalities with heterogeneous results. In the systematic review of management of NTM cervicofacial lymphadenitis conducted by Zimmermann et al., including 1906 cases, less than 20% received treatment that did not include surgery ( $n = 328$ ). But without surgery, confirmation of diagnosis becomes difficult and therefore poses challenges in determining the superior treatment method.<sup>3</sup> Among surgical management options, there are several arguments for and against complete versus incomplete excision. Approaches for incomplete excision include incision and drainage, partial excision, and curettage. Inherent arguments against complete excision are those of surgical morbidity, including facial nerve paresis, due to the disease's proximity to this nerve. Incomplete excisional techniques are often used to reduce instances of these complications and outcomes, however, at the cost of failure to heal, recurrence, and reoperation.<sup>7,8</sup> In a review by Willemse et al. investigating long-term outcomes of NTM cervicofacial lymphadenitis with 10 years follow-up, recurrence rates were reduced in patients treated with initial complete excision. In this cohort, there was a 98.7% mean cure with complete surgical excision while 34.2% of patients treated with curettage experienced recurrence of symptoms within 3 weeks after surgery, and 7.9% additionally experiencing recurrence in the following year.<sup>9</sup> Herein, we describe a case of recurrent NTM 5-years posttreatment with curettage, a novelty in the literature due to the length of time to recurrence. We also describe the current gap in the literature describing the incidence of recurrence of this disease, including how recurrence presents and is typically managed.

## Case

An otherwise healthy 2-year-old male presented to his primary care physician for evaluation of a facial lesion. The lesion was described as a violaceous discoloration on his left cheek with a "jelly-like" consistency and did not fistulize. He had not experienced any constitutional symptoms. At this time, he was diagnosed with presumed atypical NTM infection due to a mildly positive PPD skin test (without a history of BCG vaccination), negative chest X-ray, and no known risk factors for tuberculosis. Head and neck CT scan revealed central low attenuating lesion with peripheral enhancement along the anterior aspect of the left masseter muscle along with reactive cervical adenopathy, consistent with the presumed NTM

infection. Given the clinical presentation and lack of alternative confirmatory testing, NTM remained at the top of the differential. He was treated with azithromycin and trimethoprim/sulfamethoxazole for several months.

Over the next 6 months, the lesion was noted to expand despite medical intervention, leading to referral to infectious disease and otolaryngology departments at an outside institution. The patient continued to be asymptomatic. One week before scheduled surgical resection, the lesion ruptured. The patient subsequently underwent surgical curettage of the 2 cm lesion. Gram staining of the lesion showed rare leukocytes with no organisms, fungal and acid-fast bacilli smears were negative, and aerobic, anaerobic, fungal, and mycobacterial cultures were negative. A 4-week course of azithromycin and trimethoprim/sulfamethoxazole was initiated postoperatively. The patient was seen in follow-up with ENT and noted to have a persistent lesion that eventually healed, leaving behind slightly pink scar tissue.

Five years after the initial treatment, the patient returned to his primary care physician complaining of a nontender, palpable, solid bump in his left cheek, underlying the scar from the previous curettage. In the interim, the patient's family noted that small pustule-like lesions formed intermittently around the outer edges of the scar that would occasionally drain oily, milky fluid. Intermittent cervical adenopathy was also noted throughout this time. Recurrence of his NTM infection was suspected, and another course of azithromycin and trimethoprim/sulfamethoxazole was initiated.

Five months after his course of antibiotics, the patient was referred to infectious disease and the otolaryngology department at our institution for further evaluation. Head and neck CT revealed a 12 mm calcified nodule along the anterior inferior left masseter with an apparent tract extending from the superior aspect of the mass to the skin surface (Figure 1). The mass was noted to have a homogenous soft tissue-like appearance with no evidence of drainable fluid collection. Chest X-ray, PPD, and QuantiFERON tests were negative. Differentials included NTM, fistulized inclusion cyst or a sinus tract with debris.

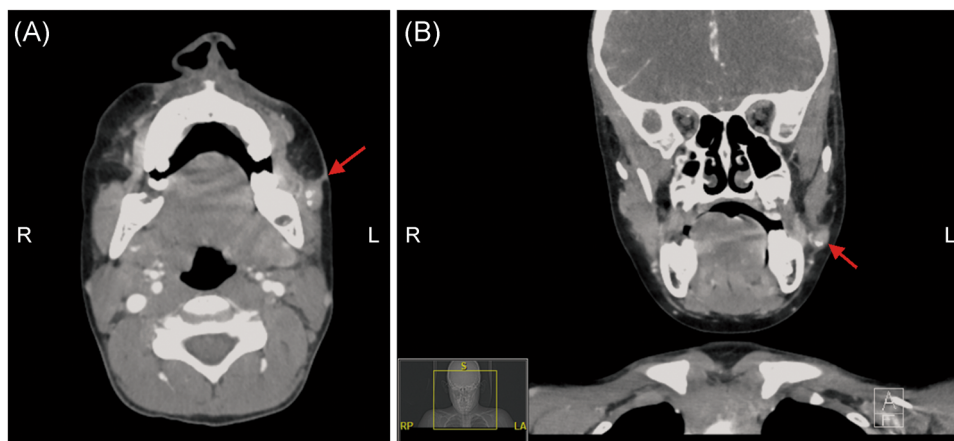
The patient's family consented to complete surgical excision due to the recurrent and persistent nature of the infection, understanding the surgical risks including the increased risk of damage to the facial nerve in comparison to their previous surgical treatment. The patient underwent transoral transbuccal excision of the residual cyst and cystic tract in January 2022 (Figure 2) (Supplemental Video Content). Biopsy of the lesion showed granulomatous inflammation and was positive on acid-fast staining, confirming recurrence of the NTM infection. He was not given postoperative antibiotics following complete surgical resection. The patient recovered well from the procedure and has no residual symptoms at the most recent follow-up 8 months post-op including no facial nerve injury (Tables 1 and 2).

## MATERIALS AND METHODS

We conducted a literature review using OVID Medline, Scopus (Elsevier), and Web of Science from as early as 1946 to April 2022. The search targeted cases of pediatric atypical mycobacterial infections of the head and neck that recurred. Search terms included: "nontuberculosis mycobacterium," "atypical mycobacterium," "recurrence," "curettage," "surgery," and "pediatric." An English limitation was added to the search and duplicates were removed resulting in 51 total references (Figure 3). Covidence review software was used for



**FIGURE 2** Intraoperative image of necrotic, calcified lymph node.



**FIGURE 1** CT head and neck revealing cutaneous fistulizing tract and left buccal calcifications (arrow). (A) Transverse view of CT head and neck. (B) Coronal view of CT head and neck.

**TABLE 1** Literature review table summary.

Reference	Study design	Country	Cases (total N)	N treated medically	N treated surgically only	N treated by complete surgical excision	N treated by incomplete surgical excision <sup>a</sup>	N treated by combination of medicine and surgery (in any order)	N recurrences
Willense et al. <sup>10</sup>	Case-control	Netherlands	103	0	103	N/R	N/R	N/R	N/R
Neven et al. <sup>7</sup>	Case series	Belgium	40	0	0	27	13	40	5
Loizos et al. <sup>11</sup>	Case series	Canada	22	0	0	0	0	22	N/R
Reuss et al. <sup>12</sup>	Case series	Germany	61	1	29	23	38	31	4
Deichmueller et al. <sup>13</sup>	Case series	Germany	29	0	0	24	5	29	5
Blyth et al. <sup>14</sup>	Cohort	Australia	68	2	3	N/R	N/R	63	11
Rustom et al. <sup>15</sup>	Case series	UK	3	0	3	0	3	0	1
Fraser et al. <sup>16</sup>	Case series	UK	31	4	27	27	0	0	7
Luong et al. <sup>17</sup>	Case series	United States	55	30	5	N/R	N/R	20	5
Mushtaq and Martin <sup>18</sup>	Case series	Australia	118	0	109	47	71	9	6
Fergusson and Simpson <sup>19</sup>	Case series	Australia	10	0	10	0	10	0	1
Wright <sup>20</sup>	Case series	Australia	67	4	63	55	8	0	3
Hawkins et al. <sup>21</sup>	Cohort	United States	82	23	0	3	56	59	13
Harris et al. <sup>22</sup>	Cohort	United States	32	0	32	19	13	0	4
Willense et al. <sup>9</sup>	Case series	Netherlands	189	0	0	151	38	0	4
Trebruegge et al. <sup>1</sup>	Cross-sectional	Australia	107	0	107	104	3	14	19

<sup>a</sup>Incomplete surgical excision including curettage.

**TABLE 2** Reports of recurrence.

Cases (total N)	N treated medically	N recurrences	N treated surgically only	N recurrences	N treated by combination	N recurrences	Complete: incomplete excision (ratio)	N recurrences (ratio)	N reported recurrences
103	0	N/A	103	N/R	0	N/A	N/R	N/A	N/R
40	0	N/A	0	N/A	40	4	27:13	0:4	4
22	0	N/A	0	0	22	N/R	N/R	N/A	N/R
61	1	1	29	0	31	3	23:38	N/R	4
29	0	N/A	0	N/A	29	5	24:5	N/R	5
68	2	N/R	3	N/R	63	N/R	N/R	N/A	11 <sup>a</sup>
3	0	N/A	3	1	0	N/A	0:3	N/A	1
31	4	0	27	7	0	N/A	27:0	N/A	7
55	30	1	5	1	20	3	N/R	N/A	5
118	0	N/A	109	N/R	9	N/R	47:71	2:4	6 <sup>a</sup>
10	0	N/A	10	1	0	N/A	0:10	N/A	1
67	4	N/R	63	3	0	N/A	55:8	1:2	3
82	23	N/R	0	N/A	59	N/R	3:56	N/R	13 <sup>a</sup>
32	0	N/A	32	4	0	N/A	19:13	0:4	4
189	0	N/A	189	N/R	0	N/R	151:38	1:3	4
107	0	N/A	93	15	19	4	104:3	N/R	19

Note: Chart created from lit review table in order they appeared—not including add ons. Abbreviations: N/A, not applicable; N/R, not recorded.

<sup>a</sup>Recurrence after unknown treatment.

literature review. Two further duplicates were removed, and 49 studies were screened by two independent reviewers for relevance, leading to consensus for exclusion of 29 further studies. Two independent reviewers then completed full-text reviews and excluded another six studies, leaving 14 studies for inclusion in our review for data extraction. Two additional studies not previously identified were later added to the literature review, including a paper by Willemse et al. published after the time of the literature search.<sup>1,9</sup>

## RESULTS

In our review of the literature, we searched for similar cases of recurrent and persistent pediatric NTM infections. Notably, recurrence was rarely commented throughout our literature review. Two of the 16 included papers did not mention the recurrence rate in their cohort, 3 of 14 mentioned their rate of recurrence of the entire cohort but provided no details on how they were initially managed. The remaining 11 of 14 studies provided recurrence rates for a specified treatment method (medical, surgical, or combination), and only 5 of 8 papers which described use of complete and incomplete excision techniques specified the recurrence rates in each group. Discussion surrounding recurrence in the included studies most commonly described recurrence presenting with recurrent lymphadenopathy or abscess formation with or without a

draining fistula. The duration of time from initial treatment to recurrence ranged from as short as 4 weeks to the longest recurring after 5 years, with the majority recurring in the first 6 months. The largest study reported on 189 participants and found a 34.2% recurrence rate within 3 weeks and 7.9% recurrence rate within the first year in the curettage group, compared with 1.3% recurrence in the group who underwent complete surgical excision.<sup>9</sup> In the present case, infection presumably recurred after curettage and antibiotic treatment and persisted for many years before fistulizing and presenting 5 years later. Reuss et al. presented 61 cases of NTM which were treated in a multitude of ways.<sup>12</sup> Of four cases which recurred, one case of recurrence presented 5 years later, along with ours, this is the longest length of time to recurrence identified in the existing literature. The patient in this study was initially treated in a combination method with complete excision followed by 12 months of rifabutin. At the time of recurrence this patient was treated with a 10-month regime of clarithromycin, rifabutin, and ethambutol.<sup>13</sup> All studies described treating recurrence in a broad variety of ways. Secondary surgeries for recurrences or following initial treatment failure were described in all 16 papers to varying degrees. Neven et al. described that 54% ( $n = 7$ ) of cases treated with incomplete excision in their cohort required a second surgical intervention for complete excision due to persistent infection or recurrence occurring between 18 days and 25 months after the initial intervention.<sup>7</sup> Many of the included papers discussed the trend of initial complete excision being associated with

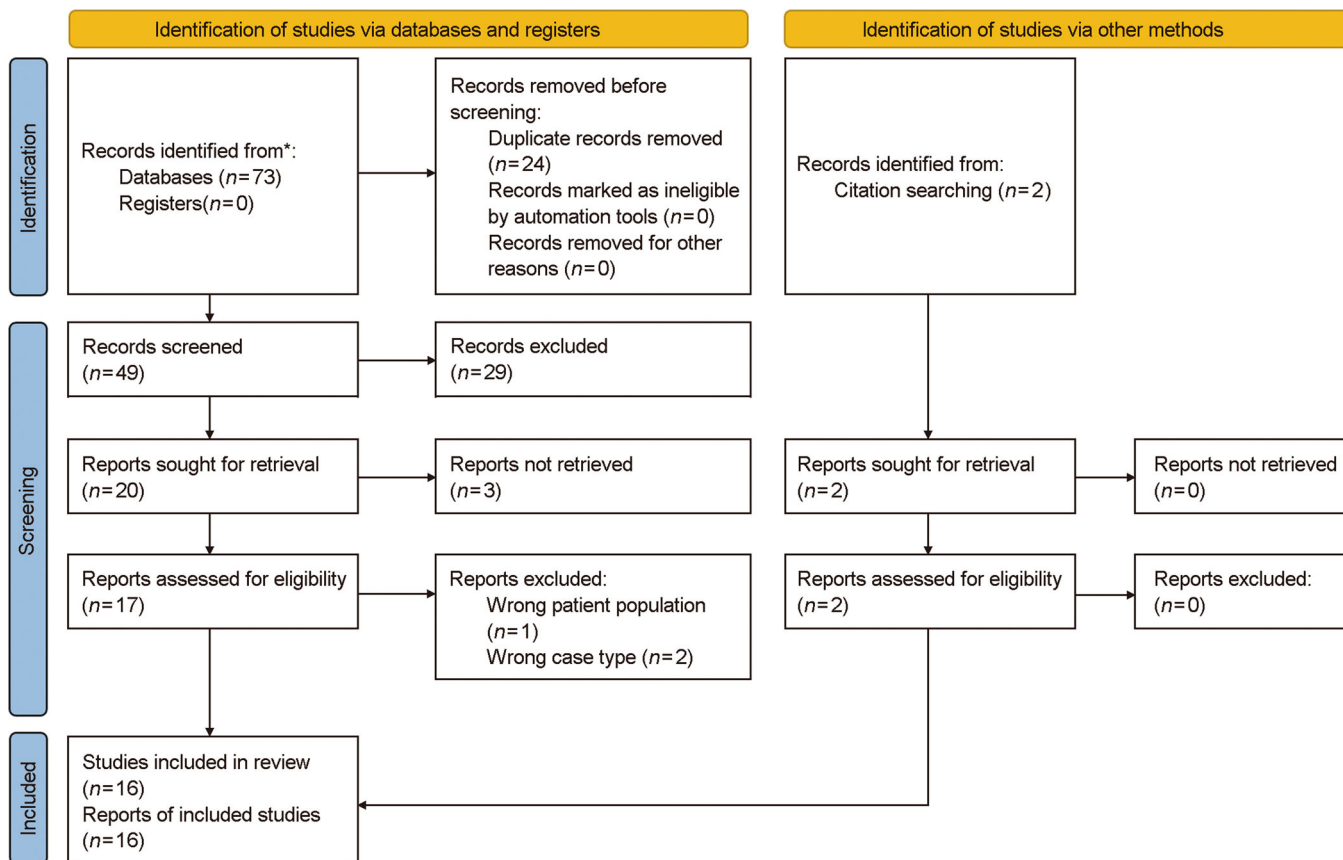


FIGURE 3 CONSORT diagram.

lower rates of recurrence.<sup>1,7,9,13,14,18,22</sup> Another frequent discussion was the association between previous biopsy (incision and drainage or FNA) being associated with higher rates of recurrence, even after complete excision.<sup>7,16</sup> This has previously been acknowledged in the literature and thus it is widely advised to avoid incisional biopsies of any kind in cases with clinical suspicion of NTM lymphadenitis.<sup>3,23</sup>

In this review, we identified 100 reported cases of recurrence of NTM cervicofacial lymphadenitis. Recurrence rates varied with initial treatment modality with the highest incidence of recurrence following incomplete surgical excision (6.6%) and the lowest incidence of recurrence with complete surgical excision (0.8%) (Table 3).

## DISCUSSION

The present case describes a patient who underwent prolonged medical treatment and curettage for NTM lymphadenopathy which recurred after 5 years.

Curettage has been an accepted method of surgical treatment for NTM cervicofacial lymphadenitis for many years.<sup>19</sup> Curettage has been thought to have benefits over complete surgical excision in cases of NTM lymphadenitis because of its reduced risk to facial nerve damage and its potentially more desirable natural scarring.<sup>24</sup> In some cases, these benefits outweigh the reduced cure rate associated with

curettage in comparison to complete surgical excision.<sup>19,25</sup> By treating NTM lymphadenitis with curettage, the contents of the infected, necrotic node and potentially fistulizing tract are scraped out using a curette, with an approach from the surface of the skin. Continuous back-and-forth scraping is continued until normal uninfected lymph node tissue is met. The resulting specimen can be sent for pathology, usually yielding the clinical diagnosis of NTM with necrotizing granulomas, however without the confirmation of negative margins. Curettage is a minimally invasive surgical procedure, and thus quick, cost-effective, and with few surgical complications. The wound created by curettage is cauterized and left open to healing by secondary intention. Some surgeons believe this provides a natural and cosmetically appealing scar.<sup>24</sup> Healing times with curettage are known to be variable, with a mean healing time of  $11.4 \pm 5.1$  weeks.<sup>26</sup>

Although curettage reduces the risk of transient facial nerve paralysis, the reduced cure rate, complications, persistent infections, and recurrence rates are consequences of treating NTM lymphadenitis with curettage. Beginning with cure rates, in the meta-analysis by Zimmerman et al.,<sup>3</sup> curettage had an adjusted mean cure rate of 87.9%. This was lower than the adjusted mean cure rate for complete surgical excision by >10% (98.7%), with 18% of cases requiring an additional surgical procedure to cure.<sup>3</sup> Furthermore, cure rates in curettage are determined clinically by resolution of infection because without complete excision and pathological determination of total infection



**TABLE 3** Summary of reports of recurrence in the literature.

Total cases	N treated medically	N	recurrences (%)	N surgically only	N	recurrences (%)	N treated by combination	N	recurrences (%)	N treated by complete excision	N	recurrences (%)	N treated by incomplete excision	N	recurrences (%)	Unaccounted recurrences
1020	64	2	(3.1)	666	32	(4.8)	292	15	(5.1)	480	4	(0.8)	258	17	(6.6)	30

removal cannot be confirmed. In terms of complications specific to curettage, it should be noted that it has been found to have a significantly longer healing period than nodal excision. Curettage heals by primary intention, taking nearly four-times longer to heal at an average of 11.4 weeks in comparison to 3.6 weeks for lymph node excision.<sup>26</sup> Scars created by curettage often remain large, discolored, and cosmetically unappealing. Last, as the current study has described, incomplete surgical excisions, such as curettage have a higher rate of recurrence than alternative surgical options (6.6% vs. 0.8%). The curettage procedure leaves the infected lymph node in situ, leaving a site for the remaining infection to recur. Recurrences typically present similarly to original presentation with recurrent discoloration, swelling, and drainage. Although disease persistence or recurrence typically presents within 6 months of original treatment and resolution is determined after this time period has passed, curettage has the potential to recur up to 5 years post-presumed cure, as in our case.<sup>12</sup>

Complete surgical excision is widely accepted as the standard of care for NTM cervicofacial lymphadenitis when possible.<sup>9</sup> As previously mentioned, it is sometimes avoided due to proximity of infection to the facial nerve as to avoid temporary or permanent facial nerve paralysis. The frequency of this occurrence has been examined by many studies and is the main argument against performing complete excision on every patient. Complete surgical excision has the highest cure rate<sup>3</sup> with reduced rates of secondary surgeries.<sup>7</sup> Esthetics of scars are preferable to those of curettage, antibiotic treatment, and Wait-And-See approaches. Intuitively, scars have the best appearance if the infection is caught and treated early. Although some patients respond to conservative approaches, surgical options can become more complicated as infection spreads. Most surgeons advocate for early surgical intervention secondary to this. Scar-improving techniques have also been described by Willemse et al.<sup>9</sup> by which they made their incision in a near-by skin crease in healthy skin, rather than the traditional approach over the site of infection to remove affected skin. They found that over several months, the initially purple or red affected skin resolved and the patient was left with a smaller, hidden scar in a natural skin crease.<sup>9</sup> Last, although recurrence can still occur after complete excision, it is rare. Complete surgical excision has an adjusted mean cure rate of 98.7%.<sup>3</sup> In the largest study reviewing recurrence in NTM lymphadenitis, Willemse reported a recurrence rate of 1 out of 151 patients after receiving complete excision (<0.007%) with minimum of 10 years follow-up.<sup>9</sup> To prevent recurrence, maximize the opportunity for cure, and optimize esthetic outcomes, complete excision should be employed in appropriate patients when feasible.

## CONCLUSION

There is limited evidence that guides the treatment of recurrent NTM cervicofacial lymphadenitis largely because the literature frequently neglects to report recurrences and treatment of such presentations. We found few reports addressing the complication which occurred in our case report, of late recurrence following curettage. Incomplete surgical excision techniques such as curettage have a propensity to

recur and the potential to present with late recurrence. Based on the current literature and our experience, we recommend complete excision to minimize recurrence, maximize the opportunity for cure and optimize esthetic outcomes.

#### AUTHOR CONTRIBUTIONS

All authors significantly contributed to and agree with the content of this manuscript in its current state.

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None.

#### CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

#### DATA AVAILABILITY STATEMENT

The data that supports the results of this study are available upon reasonable request from the corresponding author.

#### ETHICS STATEMENT

Informed consent was obtained. This study was exempt by the Mayo Clinic Institutional Review Board.

#### ORCID

Katerina Green  <http://orcid.org/0000-0001-5937-4822>

Jeffrey Graves  <http://orcid.org/0000-0003-3347-0546>

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#### SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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