Surgery for Cervicofacial Nontuberculous Mycobacterial Adenitis in Children

An Update

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Objective: To assess optimal surgical treatment with excision or curettage techniques in children with cervicofacial nontuberculous mycobacterial (NTM) adenitis.

Design: Retrospective case series.

Setting: Tertiary university-based pediatric referral center.

Patients: Patients younger than 18 years diagnosed as having cervicofacial NTM adenitis by positive mycobacterial cultures or stains, or by histopathologic evaluation.

Interventions: Fine-needle aspiration biopsy for diagnosis, surgical excision and/or curettage of head and neck lesions for treatment.

Main Outcome Measures: Number of procedures per patient, complications, resolution of mass.

Results: A total of 32 surgical procedures were performed in 25 children with cervicofacial NTM adenitis (mean, 1.3 procedures per patient; range, 1-3): 19 excisional and 13 curettage procedures. The 14 children who had excision as an initial procedure required no additional surgery. Of 11 children who had curettage as an initial procedure, 6 (55%) required additional procedures. Three of these children had additional surgery as planned staged procedures. Excisional surgery after initial curettage (5 patients) was simplified by initial débridement and secondary healing. No complications of curettage were noted. Transient marginal mandibular nerve weakness was seen in 4 patients who had excision. Fourteen of 16 fine-needle aspiration biopsy specimens were diagnostic for NTM adenitis.

Conclusions: Cervicofacial NTM adenitis can be treated with excision or curettage. Excision remains the treatment of choice because of the high cure rate with a single procedure. We now consider curettage as a staged procedure for lesions in proximity to the facial nerve or with extensive skin necrosis, with initial curettage simplifying subsequent excision and wound closure. Preoperative counseling should include discussion of planned or unplanned revision surgery after curettage. Fine-needle aspiration biopsy allows early diagnosis of NTM adenitis.

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Nontuberculous mycobacterial (NTM) infection remains a common cause of persistent head and neck masses in children. In 1995, Tunkel and Romaneschi reported our experience with diagnosis and treatment of these lesions. We emphasized expedited diagnosis using fine-needle aspiration biopsy (FNAB) to obtain material for culture, stains, and cytopathologic analysis. Children with NTM adenitis of the head and neck were treated with surgical excision or curettage. This article reports further follow-up of those patients and our center’s experience with additional patients with surgically treated NTM adenitis.

RESULTS

Twenty-five children (17 girls and 8 boys) were diagnosed as having cervicofacial NTM adenitis and treated during the review period. The median age of these children was 28 months (range, 17-78 months). Twenty children were white and 5 were African American. The incidence of cervicofacial NTM adenitis in children appears stable, as about 3 patients were treated each year of the study period.

The diagnosis of NTM adenitis was made on the basis of positive mycobacterial cultures in 23 patients and histopathologic assessment with positive mycobacterial stains in 2 patients.
PATIENTS AND METHODS

Medical records of children with the diagnosis of cervicofacial NTM adenitis were retrospectively reviewed. These patients were treated at Johns Hopkins Hospital, Baltimore, Md, between July 1991 and September 1998. Demographic information was recorded, as were the types of diagnostic and surgical procedures. The outcomes of surgical procedures were documented, detailing complications and need for additional surgery.

The diagnosis of NTM adenitis was made when a cervicofacial mass persisted for several weeks and 1 or more of the following was present: (1) positive mycobacterial cultures from FNAB or surgical specimens; (2) positive mycobacterial staining of biopsy specimens; (3) granulomatous inflammation with caseating necrosis on histopathologic evaluation of biopsy specimens. None of the patients had features suggestive of Mycobacterium tuberculosis infection on patient history, examination, or family history.

Fine-needle aspiration biopsy was performed as previously described. Aspirated material was prepared for routine cytopathologic analysis, stained for mycobacteria by the Ziehl-Neelsen and/or fluorescent chromate techniques, and cultured for bacterial and mycobacterial species. All children had FNAB performed in the ambulatory clinic setting under local anesthesia using a 2.5% lidocaine and 2.5% prilocaine hydrochloride cream (Emla cream; Astra, Westborough, Mass) and/or lidocaine infiltration, and a few young children required sedation with oral midazolam maleate or chloral hydrate.

Curettage was performed as previously described. A small incision was made over the fluctuant area of the lesion, and orthopedic curettes were used to thoroughly remove necrotic tissue. Skin was not excised, but the undersurface of the skin was curetted to remove adherent debris. All wounds were drained or packed overnight, and allowed to heal secondarily.

Additional surgery was recommended for children with masses, skin discoloration, or nonhealing wounds that persisted without improvement for a minimum of 4 to 6 weeks after surgery.

Symptoms and signs at presentation included focal swelling of the face and neck in all 25 patients (100%), and changes in overlying skin color in 19 patients (76%). Severe skin involvement, such as necrosis or fistula formation, was seen in 13 patients (52%). No child had fever, weight loss, or other systemic complaints. The mean duration of symptoms prior to presentation was 8 weeks (range, 1-26 weeks). Children with more severe skin involvement tended toward later presentation compared with children without skin changes (9.2 weeks vs 6.1 weeks).

The location of cervicofacial NTM adenitis lesions were as follows: submandibular/facial, 9 patients; parotid, 7 patients; parotid and submandibular/facial, 1 patient; upper neck (jugulodigastric), 4 patients; submental, 3 patients; and lower neck, 1 patient. The submandibular/facial lymph nodes (n = 10, 40%) and intraparotid nodes (n = 8, 32%) were the most frequent sites of involvement. All lesions were unilateral. One child had 2 nonadjacent masses, a large parotid lesion, and a smaller ipsilateral submandibular/facial lesion. Three submental masses were clinically similar to thyroglossal duct cysts, and were diagnosed at surgery (FNAB was not performed preoperatively).

Chest radiographs were normal in all 20 children who had this test. Tuberculin skin testing was performed in 20 children; 8 results were positive (≥10 mm) (40%), 2 were "intermediate" (5-9 mm) (10%), and 10 were negative (<5 mm) (50%). One child had a positive purified protein derivative-Battey test result (skin test specific for NTM organisms) at another institution before presentation.

Sixteen children underwent FNAB for cytopathologic analysis, staining, and cultures. Fourteen FNAB specimens (87%) provided information diagnostic of or highly suggestive of NTM infection. Ten FNAB specimens had positive mycobacterial cultures, and 4 of these had positive mycobacterial stains. Of the 6 FNAB specimens that did not produce positive mycobacterial cultures, 4 showed granulomatous inflammation on cytopathologic analysis and 2 were nondiagnostic. There were no complications of FNAB.

Twenty-three children (92%) had positive cultures for NTM species. Cultures were not sent for evaluation in 1 patient. Mycobacterium avium complex was isolated in 21 children, Mycobacterium chelonae in 1 child, and Mycobacterium xenopi in 1 child.

All patients were treated surgically. Five patients received antituberculous drugs before surgery until NTM species were cultured. Five patients were treated with oral clarithromycin for wound problems or persistent mass effect after curettage; all required additional surgery.

A total of 32 surgical procedures were performed on these 25 children (mean, 1.3 procedures per patient; range, 1-3). These procedures included 19 excisional procedures and 13 curettage procedures. Fourteen children had excision of the entire mass with any involved skin as initial surgical treatment. Eleven children had curettage performed as initial surgical treatment. Curettage was preferred for lesions with extensive skin necrosis and for fluctuant parotid lesions.

All 14 children who had excision of the NTM adenitis lesions as the first surgery were cured, without additional medical or surgical treatment. Six (35%) of the 11 children initially treated with curettage required additional surgery: excision of the lesion in 4 (3 planned and 1 unplanned), repeated curettage in 1 (unplanned), and repeated curettage and subsequent excision in 1 (unplanned). Three of the children (27%) who required more than 1 procedure had the second procedure done as a planned excision 2 to 10 weeks after initial curettage. Curettage was done in these patients to debride extensive lesions with severe skin necrosis to simplify subsequent excision with primary wound closure. Revision surgery was performed at a mean of 11 weeks after the previous procedure (range, 2-6 weeks). The indications for the 4 additional procedures in the 3 children with unplanned...
additional surgery were recurrent fluctuant mass \((n = 2)\) and persistent wound drainage and crusting \((n = 2)\).

Complications of excision procedures included transient marginal mandibular nerve weakness in 4 children \((21\%)\), all of which resolved within 3 months. The only complications of curettage procedures were the need for unplanned additional surgery for persistent disease in 3 children \((27\%)\).

**REPORT OF CASES**

**PATIENT 1**

An 18-month-old white female toddler presented with a 1-month history of an enlarging right parotid mass. The patient had no fever, weight loss, cough, or history of tuberculosis exposure. At initial examination, a 3-cm parotid mass was noted with overlying skin erythema, as well as a smaller 1.5-cm mass in the area of the right mandibular notch (Figure 1). Computed tomography revealed a low-density mass within the right parotid gland (Figure 2). An FNAB of the larger parotid mass was performed, and granulomatous inflammation with necrosis was noted. Mycobacterial cultures from this specimen yielded *M avium* complex.

The patient was brought to the operating room and both lesions were curetted. The wounds healed secondarily within 3 weeks, and the underlying masses resolved over 6 weeks. The patient has no evidence of recurrence 1 year after surgery (Figure 3).

**PATIENT 2**

A 3-year-old white girl was referred with a 10-day history of an enlarging mass near the angle of the left mandible. A 4-cm mass with extensive erythema and attenuation of overlying skin was seen (Figure 4). An FNAB was performed, and acid-fast bacilli were identified with fluorochrome stain. A chest radiograph and tuberculin skin test were negative.

Because of the extensive skin involvement, and the likely need for local flap coverage if the mass was excised primarily, we planned curettage followed by excision of any residual mass after several weeks of secondary healing. Curettage was performed, and the mass decreased with dramatic resolution of skin erythema. One month later, a residual 2-cm mass was excised, and the wound was closed primarily. Subtle weakness of the marginal mandibular branch of the facial nerve resolved over 8 weeks after this procedure. Her wound healed well and she is healthy 9 months after excision (Figure 5).

**COMMENT**

Nontuberculous mycobacterial adenitis continues to be a common cause of persistent cervicofacial masses in children. As demonstrated in this series, this disease usu-
ally affects healthy immunocompetent children between the ages of 1 and 5 years.\textsuperscript{3,4,7} Nontuberculous mycobacterial adenitis presents as unilateral swelling, usually in the submandibular/facial nodes or the parotid area.\textsuperscript{8} Skin involvement can be absent, but as the disease progresses, overlying skin becomes indurated and necrosis can occur. The organism responsible for the vast majority of these infections is \textit{M. avium} (or \textit{Mycobacterium avium-intracellulare}) complex.\textsuperscript{1,3,4,9}

Delay in diagnosis remains common. We have seen no change in the duration of symptoms before presentation for otolaryngology evaluation (8 weeks) between this series and our earlier series. All children were referred after evaluation and treatment for presumptive diagnoses of reactive adenopathy, bacterial adenitis, cat-scratch disease, parotid abscess, and even malignant neoplasm. The children with later presentation tended to have more extensive involvement of overlying skin. This is significant, as almost all lesions without skin involvement can be excised primarily with a very high rate of cure.

Fine-needle aspiration biopsy was a rapid and safe method of obtaining material for culture, staining, and cytologic analysis. Fine-needle aspiration biopsy was performed at the initial otolaryngology evaluation in most cases, to quickly establish the likelihood of NTM infection and to rule out malignancy. Chest radiographs were invariably normal, and tuberculin skin tests showed reaction results of 5 mm or greater in half of the children. Nadel et al\textsuperscript{10} reviewed magnetic resonance imaging and computed tomographic scans of children with NTM adenitis, and described subcutaneous fat stranding, thickening and enhancement of overlying skin, obliteration of tissue planes, and multichambered masses. While we have noted similar findings on computed tomographic scans of NTM adenitis, such imaging assisted in surgical planning but did not provide characteristic diagnostic features.

This series demonstrates the high cure rate for excision of NTM adenitis of the head and neck. Children who underwent excision of involved lymph nodes, affected salivary tissue, and overlying skin required no additional medical or surgical treatment.

In our previous report,\textsuperscript{4} curettage appeared to have a high rate of cure for treatment of NTM adenitis, supporting the favorable results of Olson\textsuperscript{11} and Kennedy\textsuperscript{6} with this technique. In the present report, many of the patients who underwent curettage as initial surgical therapy underwent additional surgical treatment. All of these subsequent procedures were simpler and shorter than the surgery that would have been required for primary excision. Specifically, no rotation flaps were required for closure and skin excision was minimal. A common finding at the second procedure was a small area of crusting at the curettage incision, attached to a deeper small lymph node involved with granulomatous inflammation. With additional experience, curettage has been used as a staged surgical approach to simplify wound closure and improve cosmesis after subsequent excision.

The need for additional treatment after curettage may be due to several factors. (1) Additional follow-up periods may detect persistent or recurrent disease. Two of the 6 patients who had additional surgery had treatment after 6 months of follow-up. (2) We may be using curettage on more extensive lesions, lesions that we may have primarily excised in the past. This was the case in the 3 patients who had planned staged surgery—curettage and secondary healing followed by excision. Our experience with planned and unplanned “revision” excisional surgery is that the dissection is easier and the problem of wound closure (eg, need for flaps) does not exist. (3) Our wound debridement with curettage may be incomplete. Treatment of NTM adenitis with simple incision and drainage often causes persistent sinus tract formation.\textsuperscript{12} Lesions that are not fluctuant may be difficult to debride completely with curettes. (4) We may be performing additional surgical procedures too soon. Four of the 7 additional procedures were performed within 2 months of curettage. Some of these lesions may have resolved without additional surgery.

The role of medical therapy for NTM adenitis is not well established.\textsuperscript{1} Hawkins et al\textsuperscript{13} recommended multidrug antituberculous therapy for all suspected mycobacterial adenitis, with surgery used for lesions that enlarge, fluctuate, or develop skin necrosis. Clarithromycin, an oral agent...
macrolide with activity against M. avium complex, has been used successfully in one child with persistent drainage and one child with persistent swelling after simple incision and drainage of NTM adenitis lesions. We used clarithromycin to treat persistent mycobacterial infection after curettage, but all 5 of these patients required additional surgery for cure. We recognize that the children in this series probably represent the more severe manifestations of NTM adenitis, as they were referred for surgical evaluation and treatment. Spontaneous resolution of NTM adenitis may occur if tissue necrosis and suppuration is limited, and there may be a role for medical treatment of children with limited disease.

Cervicofacial NTM adenitis should be suspected in young children based on characteristic clinical features. Use of FNAB has allowed early diagnosis of these infections. Excision of NTM adenitis lesions was curative in this series, and thus remains the treatment of choice. For lesions with extensive skin necrosis or when facial nerve injury is of concern, curettage is an excellent treatment option. We counsel the parents of affected children that initial treatment with curettage may require a second surgical procedure for cure. This staged approach may optimize wound healing and cosmesis in extensive lesions.

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REFERENCES


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