International Journal of Pediatric Otorhinolaryngology 82 (2016) 102–106

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Non-tuberculous mycobacterial head and neck infections in children: Analysis of results and complications for various treatment modalities

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ARTICLE INFO

Article history:
Received 18 November 2015
Received in revised form 30 December 2015
Accepted 31 December 2015
Available online 7 January 2016

Keywords:
Non-tuberculous mycobacteria
Child
Lymphadenitis
Treatment

ABSTRACT

Objectives: Analyze the results and complications of various surgical interventions in a large cohort of children with non-tuberculous mycobacterial (NTM) head and neck infections and suggest a heuristic treatment protocol for managing this condition while aiming to maximize cure and minimize complications.

Methods: Retrospective chart review of 104 consecutive patients diagnosed with head and neck NTM at a tertiary paediatric hospital between January 1994 and December 2013 inclusive.

Results: 104 patients ranged in age between 8 months to 15 years (mean age 27 months) were reviewed and 97 patients were included in the final analysis. 6 patients excluded due to lack of follow-up and one excluded due to systemic immunocompromised condition. Sub-sites of NTM infections were submandibular ($n = 48$, 46%), cervical ($n = 40$, 38%), parotid ($n = 18$, 17%) and submental ($n = 4$, 4%). Some patients had more than one lesion so counted twice. Higher cure rates were demonstrated for primary excision (81%, $p < 0.01$) versus incisional interventions (44%, $p < 0.01$). Marginal mandibular nerve palsy following surgery was seen in 7 patients (7.2%). This was permanent in 4 patients (4%) and temporary in 3 patients (3%). All children who were complicated with marginal mandibular palsies had lesions in the submandibular region. The rate of palsy for submandibular disease alone was 15%, while 8% presented permanent palsy and 6% temporary. Marginal mandibular nerve palsy was more likely following excisional compared to incisional procedures (6 versus 1 patient, $p < 0.01$). Hypertrophic scarring occurred in 7 patients: 3 patients following excision and 4 patients after an incisional procedure. One patient suffered long term spinal accessory nerve damage presented as winged scapula.

Conclusions: Excision of NTM provides better cure rates compared to incision although at the expense of long term post-surgical morbidity. Excision should probably be the first line of treatment when the risk for neural damage is low. Incision and drainage with or without antimycobacterial treatment may be the preferred option for at-risk sub-sites (submandibular or parotid) in order to reduce long term morbidity.

1. Introduction

Non-tuberculous mycobacteria (NTM) may cause a variety of diseases both in immunocompromised and immunocompetent patients. The most common presentation in healthy children is cervical lymphadenitis [1] which is most frequently caused by *Mycobacterium avium* intracellular complex (MAIC).

NTM are ubiquitous in the environment, existing in soil and water (including tap water), and ingestion of contaminated material is thought to be the main route of head and neck infection in children [2].

Affected children are typically healthy one to five years of age with painless lymphadenitis which involves the skin [3]. This infection commonly involves the cervical, submandibular or pre-auricular regions and does not present with systemic signs or symptoms. Diagnosis is commonly based on a constellation of epidemiological, clinical and bacteriological/histological findings [3].

NTM is slowly progressive and diagnosis may be delayed after treatment failure of topical or systemic antibiotics. Surgical intervention is often necessary and tissue can stain positive to acid-fast bacilli yet might not grow in culture [4]. Histology may reveal necrotizing granulomatous inflammation, occasionally accompanied by caseous necrotic areas. NTM can be treated by incision and drainage, surgical excision or systemic antibiotic treatment and cure rates vary between these modalities according to different previous reports [5–8,20]. Surgical excision is

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http://dx.doi.org/10.1016/j.ijporl.2015.12.026
0165-5876/

considered to have the highest cure rates (up to 92%), although it carries a risk of significant complications such as permanent facial nerve branch palsy [9–11].

This report analyzes the results and complications of different surgical modalities in a large cohort of patients treated in a tertiary paediatric hospital.

2. Material and methods

This study is a retrospective chart review on all children diagnosed with Head and Neck NTM that were admitted to Starship Children's Hospital, a paediatric tertiary-level hospital in Auckland, New Zealand between January 1994 and December 2013, inclusive. 104 patients were identified.

Children were assigned a diagnosis of head and neck NTM infection if they met one of the following criteria: (1) sample from a cervical LN positive for acid-fast bacilli (AFB) microscopy and/or positive NTM culture.

(2) Positive histological confirmation from a cervical LN that showed characteristic findings (well-formed large granulomas composed of epithelioid histiocytes, multinucleated giant cells, acellular necrosis – often caseous, mononuclear inflammatory cells) with or without positive culture for NTM [12,13].

Where follow-up information was missing the patient's general practitioner was contacted. Wounds were regarded as healed if there was no underlying mass or skin involvement after surgery. Inclusion criteria were all children who were assigned a diagnosis of cervical NTM according to the criteria above. Exclusion criteria included immunocompromised patients or active TB. Imaging was not routinely performed to establish the diagnosis.

All patients were treated with one or a combination of surgical modalities which included excision of lesion, incision and drainage, incision and curettage or aspiration. None of the patients were treated exclusively with antimycobacterial drugs.

All patients were examined by an Otolaryngologist prior and after the procedures and surgical complications were recorded in patients' medical records. A paediatric Otolaryngology clinic follow up appointment was offered to all patients. Children were evaluated in regard to recurrent/residual disease and treatment-related complications. Patients with residual or recurrent disease were offered further surgical management.

Institutional review board approval was obtained for this study.

3. Statistical analysis

Non-parametric data: Independent-Samples Kruskal–Wallis test. Categorical data: Pearson Chi-Square Test (and Fishers Exact test). Two-tailed calculations performed in all analyses. p Value < 0.05 accepted as statistically significant.

4. Results

104 patients ranged in age between 8 months and 15 years were reviewed. One patient was excluded due to a systemic immunocompromised condition (Kawasaki disease). Six patients failed to attend follow up appointments and were excluded so a total of 97 patients were eventually analyzed.

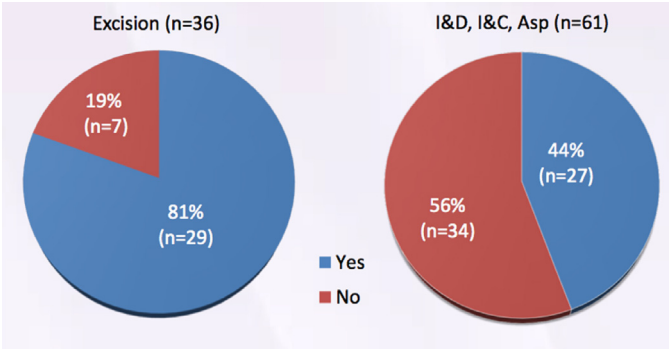

Diagram 1. Healed after 1st operation ($p = 0.01$). Patients who did not have follow-up were excluded from this analysis.

Table 1 summarizes the demographic and clinical data for this study cohort. Sub-group analysis was performed for primary excision, incision and drainage, incision and curettage and aspiration. The mean duration of follow up was 5.5 months.

There were no significant differences between the groups in terms of age, gender, affected site, average duration of the nodal mass or length of follow-up.

Sub-sites of NTM infections were submandibular ($n = 48$, 46%), cervical ($n = 40$, 38%), parotid ($n = 18$, 17%) and submental ($n = 4$, 4%). Some patients had more than one lesion so counted twice.

Cure rates following the first procedure for patients who had primary surgical excision were compared to cure rates of the other procedures. Diagram 1 shows statistically significant higher cure rates for primary excision (81%, $p < 0.01$) versus incisional interventions (44%, $p < 0.01$).

4 patients (4%) had not healed at the last follow-up and caregivers refused additional interventions. These four treatment failures followed incisional interventions (Tables 2 and 3).

Patients who had a primary excision of the lesion were less likely to require additional procedures (primary excision failure $n = 7$, 19% versus incisional $n = 34$, 56%, $p < 0.01$). Seven patients who needed secondary excisions following primary excisions did not need further treatments. All patients who failed primary excisions underwent a secondary excision. Analyzing this cohort of patients who failed their first excision did not show them to be different from the rest of the children: average age, time to nodal presentation, involvement of different neck sub-sites were similar. Regarding complications, one patient presented with temporary marginal mandibular palsy, one with hypertrophic scar and one patient from this sub-group presented with an injury to the spinal accessory nerve following the second procedure. It is reasonable to assume that a second procedure may be more prone for complications.

Most patients who had a primary incisional procedure (ID/IC) needed further surgery ($n = 34$, 51%, $p < 0.01$). 26 patients from this group needed a second procedure and 8 patients needed a third. 11 patients who had a primary incisional procedure ended

Table 1
Patient demographics.

Criteria	Excision	Incision and drainage	Incision and curettage	Aspiration	p Value
	$n = 36$	$n = 34$	$n = 31$	$n = 2$	
Median age (months)	27.0	24.5	29.0	31.0	0.642
Gender (male:female)	21:15	11:23	19:12	1:1	0.069
Median duration of nodal mass (weeks)	8.0	7.5	6.0	5.5	0.267
Median duration of follow up (months)	4.0	7.0	6.0	1.0	0.237
Did not have follow up	1 (2.7)	2 (5.9)	2 (6.4)	1 (50)	0.087