The Journal of Laryngology & Otology (2016), ***130***, *990–994*.

doi:10.1017/S0022215116008938

MAIN ARTICLE

Adenoidectomy can improve obstructive sleep apnoea in young children: systematic review and meta-analysis

L K RECKLEY[1], S A SONG[1], E T CHANG[1], B B CABLE[1], V CERTAL[2,3], M CAMACHO[1]

[1]*Otolaryngology – Head and Neck Surgery, Tripler Army Medical Center, Honolulu, Hawaii, USA,* [2]*Department of Otorhinolaryngology | Sleep Medicine Centre, Hospital CUF, Porto, Portugal, and* [3]*Centre for Research in Health Technologies and Information Systems (CINTESIS), University of Porto, Portugal*

Abstract

Objective: To systematically search for studies reporting outcomes for adenoidectomy alone as a treatment for paediatric obstructive sleep apnoea and use the data to perform a meta-analysis.

Methods: Nine databases, including PubMed and Medline, were systematically searched through to 1 April 2016. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement was followed.

Results: A total of 1032 articles were screened and 126 full texts were reviewed. Three paediatric studies (47 patients) reported outcomes. Overall, apnoea–hypopnoea index values decreased from 18.1 ± 16.8 to 3.1 ± 5.5 events per hour (28 patients). Random-effects modelling demonstrated a mean difference of −14.43 events per hour ($I^2 = 23$ per cent (low inconsistency)). The apnoea–hypopnoea index standardised mean difference was −1.14 (large magnitude of effect). The largest reduction in apnoea–hypopnoea index was observed in children aged less than 12 months (reduction of 56.6−94.9 per cent). Lowest oxygen saturation values improved from 80.0 ± 9.5 to 85.5 ± 6.0 per cent (13 children).

Conclusion: Adenoidectomy alone has improved obstructive sleep apnoea in children, especially in those aged less than 12 months; however, given the low number of studies, isolated adenoidectomy remains an area for additional research.

Key words: Adenoidectomy; Sleep Apnea Syndromes; Review, Systematic; Meta-Analysis

Introduction

Obstructive sleep apnoea (OSA) is a common disorder, with an estimated prevalence of 1–4 per cent in children.[1] In adults, there are several medical and surgical OSA treatment modalities available, such as myofunctional therapy,[2] oral appliances, positive airway pressure devices, soft tissue surgical procedures,[3–5] skeletal surgical procedures[6] and tracheostomy.[7] In children, adenotonsillectomy,[8] myofunctional therapy,[2] palate expansion[9] and allergy management are generally recommended. Recent studies have evaluated the effect of solitary procedures on OSA outcomes.[5,10] Given that adenotonsillectomy is amongst the most common procedures performed for paediatric OSA, the procedure has been systematically studied, by Brietzke and Gallagher,[8] Friedman *et al.*[11] and Lee *et al.*[12] Meta-analyses have demonstrated cure rates of between 51 and 82.9 per cent (defined by a post-adenotonsillectomy apnoea–hypopnoea index of less than one event per hour).

Although adenotonsillectomy has been very successful in treating OSA, one of the concerns, especially in children aged less than three years, is the potential for post-operative bleeding. Therefore, in the presence of small tonsils and large adenoids, some have advocated that adenoidectomy alone could be performed as a first procedure. Tonsillectomy could be performed once the child has grown more. Thus far, there have been three review articles evaluating the effects of adenoidectomy alone as a treatment for paediatric OSA.[13,14] However, to our knowledge, a systematic review with meta-analysis has not been performed. In order to determine the isolated effect of adenoidectomy on paediatric OSA, a search of multiple publication databases without regard to language would need to be performed.

This study aimed to systematically search the international literature for studies reporting outcomes for adenoidectomy alone as a treatment for paediatric OSA, and use the data to perform a meta-analysis with random-effects modelling.

Accepted for publication 28 May 2016 First published online 6 October 2016

Materials and methods

Three authors (MC, SAS and LKR) independently and systematically searched the international literature (in all languages), from the inception of each database through to 1 April 2016.

The databases searched included: PubMed/Medline; Cochrane Library; Web of Science; Book Citation Index – Science; Cumulative Index to Nursing and Allied Health Literature ('CINAHL'); Scopus; Embase; Google Scholar; and Conference Proceedings Citation Index – Science.

An example of a search strategy for PubMed/Medline is: 'adenoidect*' together with 'sleep apnea', 'sleep apnoea', 'apnea-hypopnea', 'apnoea-hypopnoea', 'respiratory disturbance', 'sleep study' or 'polysomnography'. A separate search was also performed in each database with the following search strategy: 'isolated adenoidectomy', 'adenoidectomy alone', 'adenoidectomy as the sole', 'adenoidectomy as an isolated' or 'only adenoidectomy', together with 'sleep apnea', 'sleep apnoea', 'hypopnea', 'apnoea-hypopnoea', 'respiratory disturbance', 'sleep study' or 'polysomnography'.

The Preferred Reporting Items for Systematic Reviews and Meta-Analysis ('PRISMA') statement was adhered to as much as possible in the performance of this review.[15] As this was a systematic review, and the searches were in publically available databases, with publically available articles, the study was exempt from review by the institutional review boards.

Study selection

The inclusion criteria were as follows: the studies selected needed to comprise children (aged under 18 years) with OSA, who underwent isolated adenoidectomy, in which pre- and post-adenoidectomy (quantitative) data were compared and sleep study outcome data assessed. The articles, published and unpublished, could be written in any language, and the studies could be of any design (case report, case series, cohort study, case–control study or randomised trial). Studies were excluded if: additional surgical procedure(s) were performed and there were no individual data for adenoidectomy-only patients; only qualitative data were available; or they concerned central sleep apnoea patients.

Quality assessment

The National Institute for Health and Care Excellence (NICE) quality assessment tool was used to determine the quality of the included studies, as it allows for the assessment of case series.[16]

Statistics

The null hypothesis was that there is no difference in sleep study outcomes pre- and post-adenoidectomy.

SPSS software (version 20.0; IBM, Armonk, New York, USA) was used for statistical analysis. Two-tailed, paired *t*-tests were employed; a *p*-value of <0.05 was used to determine statistical significance.

The Cochrane Collaboration's Review Manager software ('RevMan'), version 5.3, was used for sub-analyses with random-effects modelling.[17] The 95 per cent confidence intervals (CIs), mean differences and standardised mean differences were calculated using the Review Manager software. The magnitudes of effect for standardised mean differences were assigned values of 0.2 (small), 0.5 (medium) or 0.8 (large), based on Cohen's guidelines.[18] The Cochran Q statistic was employed to evaluate heterogeneity; a *p*-value of ≤ 0.10 was used to determine significant heterogeneity.[19] Review Manager software was utilised to calculate the inconsistency (I^2 statistic). Levels of inconsistency guidelines were used to assign levels: low inconsistency = 25 per cent, moderate inconsistency = 50 per cent and high inconsistency = 75 per cent.[20] As part of the assessment to determine which study was the source of heterogeneity and inconsistency (if present), the Review Manager software sensitivity analysis function was used; this function allows the removal of one study individually to determine which one(s) was the cause.

If apnoea–hypopnoea index and respiratory disturbance index values were reported, then the apnoea–hypopnoea index was used for the meta-analysis. If only the respiratory disturbance index was reported, the polysomnography scoring criteria were reviewed. If the polysomnography scoring criteria were consistent with the apnoea–hypopnoea index, it was combined with apnoea–hypopnoea index and if consistent with the respiratory disturbance index, a separate analysis was planned for the respiratory disturbance index.

Bias was assessed as recommended by the Cochrane Collaboration, in that at least 10 studies were needed in order to assess the funnel plot for any given variable. If any additional data were needed for the meta-analysis, we planned to contact the corresponding author at least twice to try and obtain the data.

Results

The systematic review searches provided a total of 1032 potentially relevant studies, of which 125 were downloaded for full text review. After reviewing the studies, references and the citing studies, 1 additional study was added, for a total of 126 studies. The final review yielded a total of three studies (47 patients) that reported outcomes for sleep study data pre- and post-adenoidectomy alone (Figure 1).[21–23] The NICE quality assessment tool was utilised to determine the quality of the data (Table I).

Adenoid size was reported by Shatz; all the children had adenoid enlargement causing more than 50 per cent narrowing of the nasopharynx.[22] However, the other two studies did not specify adenoid size.[21,23]

The apnoea–hypopnoea index values decreased from a mean ± standard deviation (SD) of 18.1 ± 16.8 (95 per cent CI = 11.9–24.3) to 3.1 ± 5.5 (95 per cent CI = 1.1–5.1) events per hour (28 patients) (Table II).

https://doi.org/10.1017/S0022215116008938 Published online by Cambridge University Press

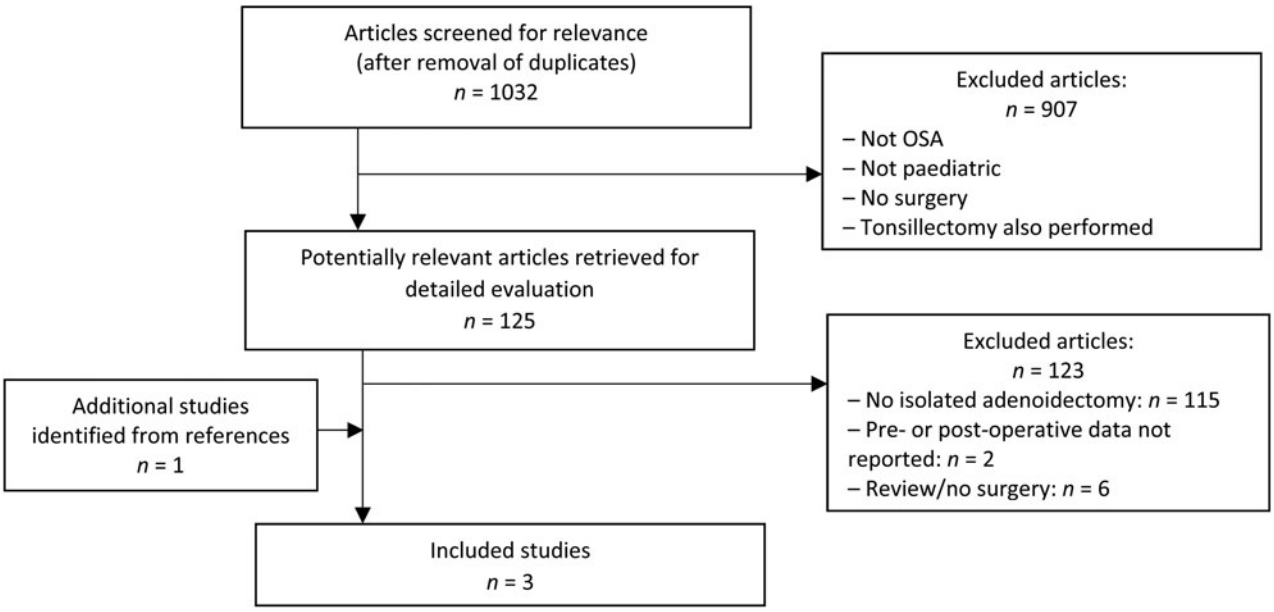

FIG. 1
Flow chart for study selection. OSA = obstructive sleep apnoea

A sub-analysis using random-effects modelling demonstrated a mean difference of −14.43 (95 per cent CI = −21.83 to −7.04) events per hour, with an overall effect Z score of 3.83 ($p = 0.0001$), Q statistic of $p = 0.25$ (no statistically significant heterogeneity) and I^2 value of 23 per cent (low inconsistency). The standardised mean difference for the apnoea–hypopnoea index values demonstrated a large magnitude of effect (−1.14 (95 per cent CI = −1.71 to −0.57), with an overall effect Z score of 3.91 ($p < 0.0001$), Q statistic of $p = 0.52$ (no statistically significant heterogeneity) and I^2 value of 0 per cent (no inconsistency). The largest reduction in apnoea–hypopnoea index was observed in children aged less than 12 months (reduction of 56.6–94.9 per cent).

The study by Shintani *et al*. reported lowest oxygen saturation values, which improved in the 13 children from a mean ± SD of 80.0 ± 9.5 per cent (95 per cent CI = 74.5–85.2) to 85.5 ± 6.0 per cent (95 per cent CI = 82.2–88.8).[23]

Discussion

This systematic review and meta-analysis revealed four main findings. The first finding was that isolated adenoidectomy performed in paediatric OSA patients can significantly improve obstructed breathing during sleep. In the 47 patients identified in the literature, adenoidectomy alone improved the apnoea–hypopnea index values. Shatz reported a decrease in apnoea–hypopnoea index values by 94.9 per cent for 15 patients under the age of 12 months,[22] and Shintani *et al*. noted that apnoea–hypopnoea index values decreased by 66 per cent in 13 paediatric patients.[23] Robison *et al*. did not cite individual patient data and instead reported the change in apnoea–hypopnoea index values for various age ranges.[21] Only patients aged 18–24 months demonstrated an increased apnoea–hypopnoea index value at 3 per cent, and it is unclear how many of the 19 patients in this study fell within this age range. The apnoea–hypopnoea index values for all other age ranges improved, with the smallest improvement at

TABLE I
GENERAL CHARACTERISTICS AND QUALITY CRITERIA OF INCLUDED STUDIES

Study (year)	Study location	Evidence level	Outcomes analysed	Quality assessment of included studies*							
				1	2	3	4	5	6	7	8
Robison *et al*.[21] (2013)	USA	4	AHI	No	Yes	Yes	No	No	No	Yes	No
Shatz[22] (2004)	Israel	4	AHI	No	Yes	Yes	Yes	No	No	Yes	No
Shintani *et al*.[23] (1998)	Japan	4	AHI	No	Yes	Yes	Yes	Yes	No	Yes	Yes

*Quality assessments of case series were conducted using the National Institute for Health and Care Excellence checklist: (1) were the case series data collected in more than one centre (i.e. multi-centre study)?; (2) is the hypothesis/aim/objective of the study clearly described?; (3) are the inclusion and exclusion criteria (case definition) clearly reported?; (4) is there a clear definition of the outcomes reported?; (5) were data collected prospectively?; (6) is there an explicit statement that patients were recruited consecutively?; (7) are the main study findings clearly described?; and (8) are outcomes stratified (e.g. by abnormal results, disease stage, patient characteristics)?[16] AHI = apnoea–hypopnoea index

https://doi.org/10.1017/S0022215116008938 Published online by Cambridge University Press

TABLE II
DEMOGRAPHIC AND QUANTITATIVE SLEEP STUDY DATA PRE- AND POST-ADENOIDECTOMY FOR INCLUDED STUDIES

Study (year)	Patient age (months)	Number of patients	AHI (mean ± SD (95% CI))		AHI change (%)	Lowest O_2 saturation (mean ± SD (95% CI))	
			Pre-op	Post-op		Pre-op	Post-op
Robison *et al.*[21] (2013)	Overall	19	NR	NR	−17.7	NR	NR
	3–5				−86.3		
	6–11				−56.6		
	12–17				−10.5		
	18–24				+3.0		
	>24				−43.8		
Shatz[22] (2004)	5 ≤ 12	15	19.57 ± 19.28 (9.8–29.3)	0.99 ± 0.64 (0.7–1.3)	−94.9	NR	NR
Shintani *et al.*[23] (1998)	NR*	13	16.5 ± 14.0 (8.9–24.1)	5.5 ± 7.5 (1.4–9.6)	−66.7	80.0 ± 9.5 (74.5–85.2)	85.5 ± 6.0 (82.2–88.8)
Total		47	18.1 ± 16.8 (11.9–24.3)	3.1 ± 5.5 (1.1–5.1)	−55.9	80.0 ± 9.5 (74.5–85.2)	85.5 ± 6.0 (82.2–88.8)

*The age of children undergoing adenoidectomy alone was not reported; however, the age range for all patients (including adenotonsillectomy patients) was one to nine years. AHI = apnoea–hypopnoea index; SD = standard deviation; CI = confidence interval; pre-op = pre-operative; post-op = post-operative; O_2 = oxygen; NR = not reported

10.5 per cent and the greatest improvement at 86 per cent.[21] While the apnoea–hypopnoea index, as demonstrated by a post-operative sleep study, is the 'gold standard' for assessing OSA improvement, there are many other measurements that can demonstrate OSA improvement.

The second finding was that lowest oxygen saturation values can also improve in children after isolated adenoidectomy. According to Shintani and colleagues' data, isolated adenoidectomy without tonsillectomy or other intervention improves lowest oxygen saturation in paediatric patients with OSA.[23] In 13 paediatric patients, Shintani *et al.* found that pre-operative lowest oxygen saturation was 80.0 ± 9.5 per cent, whereas post-operative lowest oxygen saturation improved to 85.5 ± 6.0 per cent.[23] While the oxygen nadir is clearly improved, the duration of the desaturations was not reported. Generalisations regarding the lowest oxygen saturation cannot be made given that there is currently only one publication reporting this outcome. To help determine the true effect of adenoidectomy alone, future studies could report the time spent below 90 per cent oxygen saturation, the duration of the lowest oxygen saturation, the oxygen desaturation index and the mean oxygen saturation.

The third finding was that all three studies demonstrated that isolated adenoidectomy in paediatric OSA patients can significantly improve OSA, while avoiding the additional risks associated with tonsillectomies. The avoidance of post-operative tonsillectomy bleeding improves patient safety, while at the same time isolated adenoidectomy can potentially treat OSA. The current literature indicates that children aged less than 12 months have the biggest apnoea–hypopnoea index improvement. More studies are required to determine which paediatric patients will benefit from adenoidectomy without tonsillectomy. Regardless, the higher oxygen nadir and the apnoea–hypopnoea index improvement achieved with adenoidectomy alone is promising for future treatment of OSA in paediatric patients.

The fourth finding was that while the studies included in this meta-analysis suggest that adenoidectomy alone can benefit paediatric patients with OSA, there are limitations. The number of patients across the three publications is relatively low (totalling 47 patients) compared to other, larger meta-analyses. Therefore, this meta-analysis is constrained by the limited information presented in the three articles reviewed. In addition, only Shatz reported adenoid size (more than 50 per cent obstruction at the level of the nasopharynx).[22] Furthermore, the studies included only provide level 4 evidence; none is a case–control study, cohort study or randomised trial. In addition, Robison *et al.* did not report individual post-operative apnoea–hypopnoea index values, but rather reported a per cent change in apnoea–hypopnoea index values based on age ranges.[21] Robison *et al.* also did not

https://doi.org/10.1017/S0022215116008938 Published online by Cambridge University Press

differentiate which patients had additional medical co-morbidities.[21]

- **This study reviewed isolated adenoidectomy for obstructive sleep apnoea (OSA) in paediatric patients**
- **Adenoidectomy alone can significantly improve obstructed breathing during sleep, as measured by apnoea–hypopnoea index**
- **Lowest oxygen saturation can also improve in children after isolated adenoidectomy**
- **Adenoidectomy alone can significantly improve OSA in children, while avoiding tonsillectomy-associated risks**

Future research is needed. We did not identify any studies that compared adenoidectomy alone to adenoidectomy with intracapsular coblation tonsillectomy (tonsillotomy) as treatment for OSA. A recent meta-analysis by Wang *et al*. indicated that tonsillotomy (when compared to tonsillectomy) had advantages in the short term, which included 'lower hemorrhage rate, shorter procedure time and reduced pain'.[24] Therefore, in younger children with larger tonsils, if the surgeon and/or the patient's family is against a tonsillectomy, a tonsillotomy could be considered.

Conclusion

The international literature indicates that adenoidectomy improves OSA in children, especially in those aged less than 12 months. However, given the low number of studies, adenoidectomy alone remains an area for additional research.

References

1 Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. *Proc Am Thoracic Soc* 2008;**5**:242–52
2 Camacho M, Certal V, Abdullatif J, Zaghi S, Ruoff CM, Capasso R *et al*. Myofunctional therapy to treat obstructive sleep apnea: a systematic review and meta-analysis. *Sleep* 2015;**1**:669–75
3 Certal VF, Zaghi S, Riaz M, Vieira AS, Pinheiro CT, Kushida C *et al*. Hypoglossal nerve stimulation in the treatment of obstructive sleep apnea: a systematic review and meta-analysis. *Laryngoscope* 2015;**125**:1254–64
4 Camacho M, Certal V, Capasso R. Comprehensive review of surgeries for obstructive sleep apnea syndrome. *Braz J Otorhinolaryngol* 2013;**79**:780–8
5 Senchak A, McKinlay A, Acevedo J, Swain B, Tiu MC, Chen BS *et al*. The effect of tonsillectomy alone in adult obstructive sleep apnea. *Otolaryngol Head Neck Surg* 2015;**152**:969–73
6 Camacho M, Liu SY, Certal V, Capasso R, Powell NB, Riley RW. Large maxillomandibular advancements for obstructive sleep apnea: an operative technique evolved over 30 years. *J Craniomaxillofac Surg* 2015;**43**:1113–18
7 Camacho M, Certal V, Brietzke SE, Holty JE, Guilleminault C, Capasso R. Tracheostomy as treatment for adult obstructive sleep apnea: a systematic review and meta-analysis. *Laryngoscope* 2014;**124**:803–11
8 Brietzke SE, Gallagher D. The effectiveness of tonsillectomy and adenoidectomy in the treatment of pediatric obstructive sleep apnea/hypopnea syndrome: a meta-analysis. *Otolaryngol Head Neck Surg* 2006;**134**:979–84
9 Villa MP, Malagola C, Pagani J, Montesano M, Rizzoli A, Guilleminault C *et al*. Rapid maxillary expansion in children with obstructive sleep apnea syndrome: 12-month follow-up. *Sleep Med* 2007;**8**:128–34
10 Camacho M, Riaz M, Capasso R, Ruoff CM, Guilleminault C, Kushida CA *et al*. The effect of nasal surgery on continuous positive airway pressure device use and therapeutic treatment pressures: a systematic review and meta-analysis. *Sleep* 2015;**38**:279–86
11 Friedman M, Wilson M, Lin HC, Chang HW. Updated systematic review of tonsillectomy and adenoidectomy for treatment of pediatric obstructive sleep apnea/hypopnea syndrome. *Otolaryngol Head Neck Surg* 2009;**140**:800–8
12 Lee CH, Hsu WC, Chang WH, Lin MT, Kang KT. Polysomnographic findings after adenotonsillectomy for obstructive sleep apnea in obese and non-obese children: a systemic review and meta-analysis. *Clin Otolaryngol* 2015. Epub 2015 Oct 5
13 Black AP, Shott SR. Is adenoidectomy alone sufficient for the treatment of airway obstruction in children? *Laryngoscope* 2014;**124**:6–7
14 Caldwell P, Hensley R, Machaalani R, Cheng A, Waters K. How effective is adenoidectomy alone for treatment of obstructive sleep apnoea in a child who presents with adenoid hypertrophy? *J Paediatr Child Health* 2011;**47**:568–71
15 Moher D, Liberati A, Tetzlaff J, Altman DG; PRISMA Group. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Medicine* 2009;**6**:e1000097
16 National Institute for Health and Clinical Excellence. *Methods for Development of NICE Public Health Guidance*. London: National Institute for Health and Clinical Excellence, 2006
17 Review Manager (RevMan), version 5.3 [computer program]. Copenhagen: Nordic Cochrane Centre, Cochrane Collaboration, 2014
18 Cohen J. *Statistical Power Analysis for the Behavioral Sciences*. Hillsdale, NJ: Erlbaum, 1988
19 Lau J, Ioannidis JP, Schmid CH. Quantitative synthesis in systematic reviews. *Ann Intern Med* 1997;**127**:820–6
20 Higgins JP, Thompson SG, Deeks JJ, Altman DG. Measuring inconsistency in meta-analyses. *BMJ* 2003;**327**:557–60
21 Robison JG, Wilson C, Otteson TD, Chakravorty SS, Mehta DK. Analysis of outcomes in treatment of obstructive sleep apnea in infants. *Laryngoscope* 2013;**123**:2306–14
22 Shatz A. Indications and outcomes of adenoidectomy in infancy. *Ann Otol Rhinol Laryngol* 2004;**113**:835–8
23 Shintani T, Asakura K, Kataura A. The effect of adenotonsillectomy in children with OSA. *Int J Pediatr Otorhinolaryngol* 1998;**44**:51–8
24 Wang H, Fu Y, Feng Y, Guan J, Yin S. Tonsillectomy versus tonsillotomy for sleep-disordered breathing in children: a meta analysis. *PloS One* 2015;**10**:e0121500

Address for correspondence:
Dr Lauren K Reckley,
Otolaryngology – Head and Neck Surgery,
Tripler Army Medical Center,
1 Jarrett White Road,
Honolulu,
HI 96859, USA

E-mail: lauren.k.reckley.mil @mail.mil

Dr L K Reckley takes responsibility for the integrity of the content of the paper
Competing interests: None declared

https://doi.org/10.1017/S0022215116008938 Published online by Cambridge University Press