Article type : Regular Article

Title Page

What does the world think of ankyloglossia?

Authors

Ruilin R JIN[1]

Alastair SUTCLIFFE[2]

Maximo VENTO[3]

Claudelle MILES[4]

Javeed TRAVADI[5]

Kumar KISHORE[6]

Keiji SUZUKI[7]

David TODD[8]

Susanne WOODERSON[5]

Azanna Ahmad KAMAR[9]

Li MA[10]

John SMYTH[1,4]

Ju Lee OEI[1,4]

Affiliations

[1]School of Women's and Children's Health, University of New South Wales, Randwick, NSW, Australia

[2]Department of Paediatrics, University College London, London, United Kingdom

[3]Division of Neonatology, University and Polytechnic Hospital La Fe, Valencia, Spain

[4]Clinical Midwifery Consultant for Lactation Services, Royal Hospital for women, Randwick, NSW, Australia

[5]Department of Neonatology, John Hunter Children's Hospital, Newcastle, NSW,

This article has been accepted for publication and undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the Version of Record. Please cite this article as doi: 10.1111/apa.14242

[6]Cloudnine Hospitals, India

[7]Department of Pediatrics, Tokai University School of Medicine, Isehara, Kanagawa, Japan

[8]Department of Neonatology, Canberra Hospital, Garran, ACT, Australia

[9]Department of Neonatology, University of Malaya Medical Centre, University of Malaya Medical Centre, Kuala Lumpur, Malaysia

[10]Department of Paediatrics, Shijiazhuang Children's Hospital, Shijiazhuang, Hebei Province, China.

Corresponding author

Dr Ju Lee Oei, Department of Newborn Care, The Royal Hospital for Women, Barker Street, Randwick, NSW, Australia, 2031. [Phone: 61 2 9382 6152, Fax: 61 2 9382 6191, Email: j.oei@unsw.edu.au

Short title: Tongue tie survey

Abstract

Aim:

The diagnosis of tongue-tie (or ankyloglossia) has increased more than 10-fold in some countries. Whether this is a global phenomenon or related to cultural and professional differences is uncertain.

Methods:

An online survey in English, Japanese, Mandarin Chinese and Spanish was disseminated between May to November 2016 via 27 international professional bodies to >30 clinical professions chosen *a priori* to represent occupations involved in the management of neonatal ankyloglossia.

Results:

A total of 1,721 responses came from nursing (51%), medical (40%), dental (6%) and allied health (4%) clinicians. Nurses (40%) and allied health (34%) professionals were more likely than doctors (8%) to consider ankyloglossia as important for lactation problems, as were Western (83%) compared to Asian (52%) clinicians. Referrals to clinicians for ankyloglossia management originated mainly from parents (38%). Inter-professional referrals were not clearly defined. Frenotomies were most likely to be performed by surgeons (65%) and dentists (35%), who were also less likely to be involved in lactation support. Clinicians performing frenotomies were more likely to consider analgesia as important compared to those not performing frenotomies.

Conclusion:

The diagnosis and treatment of ankyloglossia varies considerably around the world and between professions. Efforts to standardize management are required.

Keywords: Ankyloglossia, frenotomy, opinion, survey

Key Notes

- The diagnosis and treatment of infants with ankyloglossia has increased significantly in the last decade.
- Our study identified considerable professional and geographical differences in opinion about the management of infants with ankyloglossia with little awareness, acknowledgement and utilisation of clinical practice guidelines.
- The great differences in opinion indicate a potential for either under or over treatment of infants and consequently, to potential complications in lactation.

List of Abbreviations

HATLFF: Hazelbaker's assessment of lingual frenulum function

BTAT: Bristol Tongue Assessment Tool

CPG: Clinical practice guidelines

Introduction

Ankyloglossia, or tongue-tie, is a congenital anomaly characterized by an abnormally short, thickened, or tight sublingual frenulum (1). The frenulum grows with age but a significant proportion (25-44%) of infants with ankyloglossia may have early functional problems, especially in breast-feeding (2-4). Ankyloglossia prevents the tongue from extending beyond the lip, resulting in ineffective latching, sucking and swallowing (all actions necessary for successful breast-feeding). As a result, the mother experiences nipple pain, the infant fails to thrive and in many cases, gives up breast-feeding (5).

Simple ankyloglossia can be treated with a frenotomy. This entails snipping or division of the shortened frenulum, usually in a conscious infant with or without local or mild analgesia (6). Frenotomies can be performed in various locations (in or out of hospital) using a variety of implements, including sharpened fingernails (7), scissors and specially designed implements (8). In experienced hands, frenotomies are relatively quick procedures but serious complications, such as scarring, bleeding and infection, are rare but not unknown (9).

The diagnosis and treatment of ankyloglossia have been controversial for millennia (10). Despite the relatively recent advent of various diagnostic tools designed to assess ankyloglossia severity (11, 12), the natural history of ankyloglossia and the outcomes (both long and short term) of infants treated with frenotomies remain unknown (1, 13). In the last decade, the number of infants diagnosed with ankyloglossia (and thus, treated with frenotomies) has increased by >70% in countries like Canada (14). In the USA, data suggest an increase of 866% in the number of frenotomies performed over the last decade (15).”(15).

The cause of this rise is unknown but important to determine. Untreated ankyloglossia may lead to breast feeding problems, with all its attendant consequences including loss of protective immunity for the child (16) and possibly, even cognitive loss (17). However, subjecting infants to a surgical procedure, no matter how minor, causes pain, stress, possibly longer-term neurological harm (10, 18).

We therefore sought to determine if professional and global opinion varied in the diagnosis and treatment of ankyloglossia, as professional practice is one of the most powerful determinants of patient care and service utilization (19). We hypothesized that 1. Opinions about ankyloglossia will vary between countries and professions 2. The number of professions involved in managing infants with ankyloglossia are numerous and 3. The majority of clinicians would not use established tools to guide management of babies with suspected ankyloglossia.

Methods

We disseminated an online web-based survey (www.surveymonkey.com) to health professionals determined *a priori* to be involved in the care of infants with breast-feeding problems and of ankyloglossia. The survey was developed in English and translated and back-translated into Mandarin Chinese, Japanese and Spanish by native speakers.

The link to the survey was sent three times to professional organizations for dissemination to their members between May to November 2016. Respondents were asked to state their professional background in broad terms (e.g. neonatologist, nurse, lactation consultant) as specific designations may have differed between countries (e.g. attending specialist in North America versus consultant physician in Australia). They were also asked to disseminate the survey to colleagues who were involved in the management of infants with ankyloglossia.

Survey Questions

The survey consisted of 20 questions (see supplemental table 1) seeking information about the caseload seen by the respondents, their opinions on ankyloglossia and frenotomy, the referral pathway for infants with possible ankyloglossia, frenotomy practice, knowledge and use of clinical practice guidelines (CPGs) and breastfeeding and ankyloglossia assessment tools. Fields for free text comments were also provided.

Statistical analyses

Descriptive statistics were used. The Chi-square test was used to evaluate proportions between groups. Responses were analysed according to professions and to country of origin. A p-value of <0.05 was considered significant.

Ethical consideration

This study was approved as a quality improvement survey by the Hunter New England Research Ethics Committee on 24th March 2016.

Results

A total of 1721 responses were received from 28 countries and >30 different professions (see Table 1). Nursing-related professions (nurses, midwives and lactation consultants) accounted for 51% of the responses while medical, (paediatricians, neonatologists, obstetricians, general practitioners and surgeons), dental and allied health professionals comprised the remainder. Most (79%) of the respondents were female and half (50%) were from Australia and New Zealand. Most (82%) managed <10 cases of suspected ankyloglossia per month (see Table 1).

1. The relationship between ankyloglossia and breast-feeding difficulties.

Opinions about the relationship between ankyloglossia and breast-feeding difficulties varied greatly between professions and geography. For example, nursing (40%) and allied health (34%) professionals were more likely than doctors (8%) or dentists (19%) to strongly agree that feeding difficulties were caused by ankyloglossia. Professionals from Western countries (83%) were more likely than those from Asian countries (52%) to agree about this relationship. Clinicians with a shorter working life (<10 years; 65%) and those managing higher case-loads (>30 per month; 84%) were more likely than their older colleagues (47%) or than those who managed fewer babies (<1 per month; 40%) to agree to this association.

2. The referral pathway to and between health professionals

The referral pathway to and between professions for ankyloglossia was complex and without any clearly defined hierarchy. Parents were the most common initial referral source (38%) and more than half (57%) of the professionals agreed that parents would seek alternative assistance if they did not provide a frenotomy service. The professions also avidly cross-referred, mainly to lactation consultants (35%), paediatricians (32%), surgeons (31%) and dentists (21%) (Quotes #1&2). North American respondents were unique in utilizing the services of alternative health therapists, including chiropractors, bodyworkers, myo-functional therapists and orofacial mycologists (Quotes #3&4).

3. Knowledge and opinion of clinical practice guidelines (CPG)

Nursing and allied health professions (63%) were more likely to be aware of, and to utilize CPGs for the diagnosis and treatment of ankyloglossia than doctors and dentists (39%. Most (62%) used local (e.g. hospital or community) CPG and some respondents (mostly doctors) commented that current CPGs were of low quality, not evidence based and not useful for their practice (Quotes #5-7).

4. Diagnosing ankyloglossia.

Most of the respondents would diagnose ankyloglossia on tongue appearance (64%) and a history of maternal nipple pain (60%) and feeding difficulty (43%) (Quotes 8&9). Few respondents, especially doctors, used formal assessment tools, including the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) (11) (see Figure 1). Over 20 different methods and tools were described, including the Bristol Tongue Assessment Tool (BTAT) (12), LATCH (Jensen et al., 1994), the Carole Dobrich Frenotomy Decision Rule for Breastfeeding Infants (20) and the Martinelli Tongue-tie Tool (21)

5. Frenotomies

Only 24% of the respondents performed frenotomies. It was most commonly performed by surgeons (65%) and dentists (35%, see Figure 2). Only 8% of Asian respondents performed frenotomies while most doctors (71%) and nurses (95%) did not perform frenotomies. The most common reasons for this were: the lack of training (>70%), the belief that breast-feeding education and support was more important (>40%) and the belief that frenotomy had no impact on lactation (31%). Some doctors commented that ankyloglossia was "over-diagnosed" and led to unnecessary frenotomies (Quote #10-11) but several nurses expressed wishes for more education so that they could perform frenotomies themselves (Quotes #12-13). Respondents who performed frenotomies were more likely to consider ankyloglossia as a cause of lactation difficulties.

Nurses (45%) were more likely than doctors (22%) to advocate for early (<7 days) frenotomy. Sucrose was advocated as the analgesia of choice by 45% of respondents. Respondents who performed frenotomies (33%) were more likely to recommend analgesia for the procedure compared to those who did not perform frenotomies (16%).

Discussion

Our study demonstrates considerable variation between health professionals in the diagnosis and treatment of ankyloglossia in the newborn infant. Nurses and allied health professionals were more likely than physicians to attribute breast-feeding problems to ankyloglossia and to advocate early (<7 days) frenotomies. This discovery is not new. Disagreement and competition between midwives (who were almost always women) and surgeons (who were almost always men) have existed since Biblical times (10) and in communities all over the world (8, 22), especially in traditional settings where the care of the mother and infant remains primarily a female responsibility (7, 23).

These differences may lead to significant management variations and a disconnection between the services provided by professionals who perform frenotomies and those that provide lactation support. Doctors may be more cautious in recommending frenotomies because they may not be the primary clinician involved in lactation support. However, they may be more likely to have to treat complications caused by frenotomies e.g. haemorrhage and damage to anatomical structures (6) and may therefore be more reticent in recommending a surgical procedure over education and support for lactation difficulties.

Furthermore, in North America, alternative health professionals like bodyworkers and myofacial therapists also appear to be involved in the treatment of ankyloglossia. Whether such management will lead to better outcomes are as yet, unknown. Further study of these services and efforts to connect the follow-up practices of clinicians performing frenotomies to those referring for frenotomies are needed to reduce economic waste or exposure of patients to potentially futile or harmful treatments.

Our survey also highlights differences in how ankyloglossia is managed and perceived around the world. Asian respondents were less likely to consider ankyloglossia as a cause of lactation difficulties. They were also less likely to perform frenotomies even in countries with high breast-feeding rates, such as Japan (24) and Malaysia (25). In Asian countries with low breast-feeding rates (e.g. Hong Kong and China, 6.2% at 6 months, (26)), the lack of support and emphasis on lactation (27, 28) may lead to under-diagnosis of ankyloglossia.

There was a low rate of awareness and acknowledgement of CPGs for ankyloglossia. CPGs are designed to inform on best practice, especially in areas of medicine with little definitive evidence (29) but our survey found that clinicians, especially physicians, did not consider either CPGs or diagnostic

tools to be informative or helpful. Less than 50% of respondents, for example, used the HATLEFF tool (11), >20 assessment tools were used by the respondents and many respondents had little confidence in CPGs, commenting that they were “contradictory” and “poorly done”.

Finally, it must be recognized that frenotomy, regardless how minor a procedure, causes not only pain but potential complications, which are, fortunately, uncommon (6). Practitioners must be encouraged to exclude potential problems, such as inherited coagulopathies, prior to the procedure. It is also important to acknowledge that pain, no matter how seemingly brief, may not be entirely innocuous, especially during critical periods of neurodevelopment (18, 30). Further study into the use of analgesia for frenotomy or to the long-term (including neurodevelopmental) impact of the procedure is warranted. Hence, the decision to do a frenotomy should not be hastened simply because of its perceived simplicity and must also take into account possible health risk factors as well as the financial and emotional burden of the procedure on the parents.

Limitations

Our survey has several limitations. Since it was anonymous, non-respondents could not be chased up, although repeat email was sent to the professional bodies to encourage replies. Furthermore, it is likely that response rates were lower in clinicians who did not consider frenotomy or ankyloglossia to be important in lactation success.

Conclusion

In conclusion, our survey highlights the vast differences in opinion and management strategies for ankyloglossia, which are affected by factors such as profession, geography, personal experience and

caseload. There is a need to evaluate current diagnostic tools and CPGs in order to uniformly diagnose and treat this condition to prevent potential lactation difficulties.

Funding and Conflict of Interest Statement

The authors declare that there are no known conflicting interests and funding in the preparation of this manuscript.

References

1. Francis DO, Krishnaswami S, McPheeters M. Treatment of ankyloglossia and breastfeeding outcomes: a systematic review. *Pediatrics* 2015;13

2. Hogan M, Westcott C, Griffiths M. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. *J Paediatr Child Health* 2005;41:246-50.

3. Messner AH, Lalakea ML, Aby J, Macmahon J, Bair E. Ankyloglossia: incidence and associated feeding difficulties. *Arch Otolaryngol Head Neck Surg* 2000;126: 36-9.

4. Segal LM, Stephenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia Methodologic review. *Can Fam Physician* 2007;53: 1027-33.

5. Power R, Murphy J. Tongue-tie and frenotomy in infants with breastfeeding difficulties: achieving a balance. *Arch Dis Child* 2014.

6. NICE. Division of ankyloglossia (tongue-tie) for breastfeeding. Interventional procedures guidance: *NICE* 2005 [cited 2017 Aug 7] Available from: URL: http://www.nice.org.uk/Guidance/IPG149

7. Opera P, Eke G. Tongue-Tie in Children; Mothers Perceptions, Attitudes and Practices in Port Harcourt, Nigeria. *Glob J Med Res* 2013;13(5).

8. Brinkmann S, Reilly S, Meara J. Management of tongue-tie in children: A survey of paediatric surgeons in Australia. *J Paediatr Child Health* 2004;40: 600-5.

9. Academy-of-Breastfeeding-Medicine. Protocol # 11: Guidelines for the evaluation and management of neonatal ankyloglossia and its complications in the breastfeeding dyad: *Academy of Breastfeeding Medicine* 2004 [cited 2017 Aug 24] Available from: http://www.bfmed.org/Media/Files/Protocols/ankyloglossia.pdf

10. Obladen M. Much ado about nothing: two millenia of controversy on tongue-tie. *Neonatology* 2009;97:83-9.

11. Hazelbaker AK. The assessment tool for lingual frenulum function (ATLFF): *Use in a lactation consultant private practic*e. Passadena, CA: Pacific Oaks College 1993.

12. Ingram J, Johnson D, Copeland M, Churchill C, Taylor H, Emond A. The development of a tongue assessment tool to assist with tongue-tie identification. *Arch Dis Child Fetal Neonatal Ed* 2015. 2014.

13. O'Shea JE, Foster JP, O'Donnell CP, Breathnach D, Jacobs SE, Todd DA, et al. Frenotomy for tongue-tie in newborn infants. *The Cochrane Library* 2014.

14. Joseph K, Kinniburgh B, Metcalfe A, Razaz N, Sabr Y, Lisonkova S. Temporal trends in ankyloglossia and frenotomy in British Columbia, Canada, 2004-2013: a population-based study. *CMAJ open* 2016;4:E33.

15. Walsh J, Links A, Boss E, Tunkel D. Ankyloglossia and lingual frenotomy: National trends in inpatient diagnosis and management in the United States, 1997-2012. *Otolaryngol Head Neck Surg* 2017;156:735-40.

16. WHO, UNICEF. Baby-friendly hospital initiative: revised, updated and expanded for integrated care. In: UNICEF WHO. *Geneva: World Health Organization* 2009.

17. Horta BL, Loret de Mola C, Victora CG. Breastfeeding and intelligence: a systematic review and meta-analysis. *Acta Paediatr* 2015;104:14-9.

18. Schwaller F, Fitzgerald M. The consequences of pain in early life: Injury-induced plasticity in developing pain pathways. *Eur J Neurosci* 2014;39:344-52

19. Reeves S, Perrier L, Goldman J, Freeth D, Zwarenstein M. Interprofessional education: effects on professional practice and healthcare outcomes (update). *The Cochrane Library* 2013.

20. Srinivasan, A., Dobrich, C., Mitnick, H., & Feldman, P. Ankyloglossia in breastfeeding infants: the effect of frenotomy on maternal nipple pain and latch. *Breastfeeding Medicine 2006. 1*, 216-224.

21. Martinelli, R., Marchesan, I., & Berretin-Felix, G. Lingual frenulum protocol with scores for infants. *Int J Orofacial Myology 2012. 38*, 104-112.

22. Messner AH, Lalakea ML. Ankyloglossia: controversies in management. *Int J Pediatr Otorhinolaryngol* 2000; 54:123-31.

23. Kraft JM, Wilkins KG, Morales GJ, Widyono M, Middlestadt SE. An evidence review of gender-integrated interventions in reproductive and maternal-child health. *J Health Commun* 2014;19:122-41.

24. Inoue M, Binns CW, Otsuka K, Jimba M, Matsubara M. Infant feeding practices and breastfeeding duration in Japan: A review. *Int Breastfeed J* 2012;7:15.

25. Tan KL. Factors associated with exclusive breastfeeding among infants under six months of age in peninsular Malaysia. *Int Breastfeed J* 2011;6:2.

26. Ouyang Y-Q, Su M, Redding SR. A survey on difficulties and desires of breast-feeding women in Wuhan, China. *Midwifery* 2016; 37:19-24.

27. Tarrant M, Lok KY, Fong DY, Lee IL, Sham A, Lam C, et al. Effect of a hospital policy of not accepting free infant formula on in-hospital formula supplementation rates and breast-feeding duration. *Public Health Nutr* 2015;18:2689-99.

28. Li Y, Kong L, Hotta M, Ushijima SAW. Breast-feeding in Bangkok, Thailand: Current status, maternal knowledge, attitude and social support. *Pediatr Int* 1999;41:648-54.

29. Lohr KN, Field MJ. Clinical practice guidelines: directions for a new program: *National Academies Press* 1990.

30. Taddio A, Goldbach M, Ipp M, Stevens B, Koren G. Effect of neonatal circumcision on pain responses during vaccination in boys. *Lancet* 1995;345:291-2.

Table 1

Respondent characteristics

Professions		
Category	**Profession**	**Number of respondents (%)**
Medical Professionals	Paediatricians	277 (16%)
	Neonatologists Obstetricians	238 (14%)
	General Practitioners	97 (6%)
	Paediatric Surgeons General Surgeons ENT Surgeons Plastic Surgeons	68 (4%)
Nursing Professionals	Midwives Nurses Neonatal Nurses Child and Family Nurses	528 (31%)
	Lactation Consultants IBCLC- International Board Certified Lactation Consultants La Leche League Leader	298 (17%)
	Multi-professional nurses: more than one midwifery, nursing and lactation qualifications	52 (3%)
Dental Professionals	Dentists Paediatric Dentists Dental Surgeons Orthodontists	100 (6%)
Allied Health Professionals	Speech Pathologists/ Speech and Language Therapists	39 (2%)
	Dental Therapists Physiotherapists Occupational Therapists Dietitians Paediatric Dieticians	16 (1%)
	Alternative medicine therapists Chiropractors Orofacial myofunctional therapists/ myologists Craniosacral therapists Osteopaths Bodyworkers	8 (0.5%)
Regions		
Region	**Countries**	**Number of Respondents (%)**
Oceania	Australia New Zealand	393 (23%) 460 (27%)
UK	UK	39 (2%)
North America	North America	258 (9%)
Asia	Japan Others (China, Singapore, Malaysia, India, etc.)	78 (4%) 50 (3%)
Europe	Spain Others (Belgium, Denmark, Netherlands, Ireland,	97 (6%) 33 (2%)

	Greece etc.)	
Others	Other (e.g. Chile, South Africa, Argentina etc.) Not stated	6 (0.3%) 407 (24%)

Table 2: *Selected free-text responses*

Referrals

Quote #1: "Someone experienced in tongue tie - assessing both appearance and function - this may be dentist, lactation consultant, surgeon - but related to their individual experience" – Australian GP

Quote #2: "Depending on the locally skilled personnel available, and the degree of difficulty when assessed" – New Zealand Midwife

Alternative therapists

Quote #3: "Don't officially refer, but give information about resources including laser dentist, pediatric chiropractor, CST [craniosacral therapists], SLP [speech pathologists], etc." – US Lactation Consultant

Quote #4: "Every baby should be followed up with bodywork as I have seen countless times that tie releases do not solve all symptoms. The restrictions often extend deeper into the body. Infants often need correct rest insulation to allow for better ROM and neural connections" – US Registered Massage Therapist

Guidelines:

Quote #5: "I've seen all sorts of guidelines that are contradictory….so I don't use them" -- Australian Neonatologist

Quote #6: "I keep appraised of the literature but there are no good guidelines that I refer to regularly." -- Australian Paediatrician

Quote #7: "As most practitioners I send families to are in private practise, their guidelines are their own. I think this needs to be formalised as we see some very badly done frenotomies!!" -- Australian Lactation consultant

Assessment Elements:

Quote #8: "We have developed our own process based on experience with many of the above criteria. None of the tools are useful on their own" --Lactation consultant

Quote #9: "I look at limitation of tongue movement combines with feeding difficulty (significant pain on mother, suboptimal weight gains etc.). Not sure if there is a scale for these to what it is called." -- New Zealand Paediatrician

Over-diagnosis and Over-procedure

Quote #10: "Depends on who is making the diagnosis - my opinion is that it is way over-diagnosed by lactation consultants and midwives." -- Australian Paediatrician

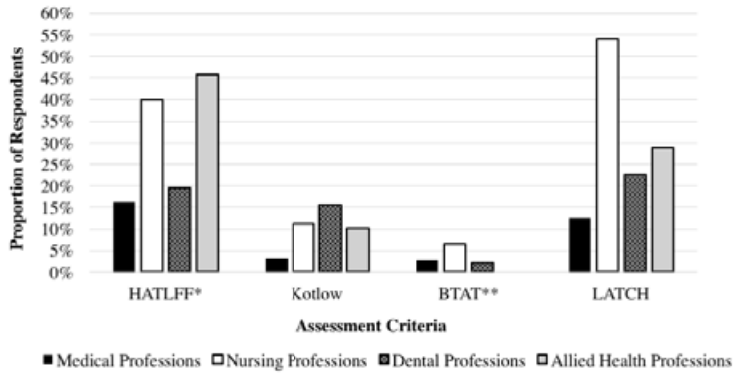
Quote #11: "The biggest issue I have is that there seems to be no exclusion criterion…. But I mean in healthy babies, everyone who is referred to lactation consultant gets a snip." – New Zealand Midwife

Nursing professionals' scope of practice

Quote #12: "As an IBCLC in Canada, it is not within my scope of practice to perform frenotomy. If I were allowed to learn and perform this procedure myself, I would." – Canadian Lactation Consultant

Quote #13: “I don’t do frenotomies because there are very limited training opportunities in Australia, and no-one available locally to teach and supervise.” – Australian Midwife and Lactation Consultant

Figure 1: *Ankyloglossia assessment tools utilized by the respondents*



* HATLFF: Hazelbaker Assessment Tool for Lingual Frenulum Function
**BTAT: Bristol Tongue Assessment Tool

Figure 2: *Respondents performing frenotomies in daily practice*