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Tongue-tie in Newborns – a History of Changing Medical Attitudes

Wędzidełko języka u noworodków w perspektywie historycznej

Usque adeone scire tuum, nihil est, nisi te scire hoc, sciat alter?
Persius

SUMMARY

This historical article discusses changes in opinions about tongue-tie (ankyloglossia) over two millennia. The sudden surge in interest, since 1990, in the impact of tongue-tie on tongue functions during breastfeeding and during speech development has renewed the historical controversies in contemporary terms and circumstances. A feature has been, and continues to be, the persistence of differences of opinion between and within the various medical professions and laypeople about the significance of tongue-tie in infancy, the indications for its division, and who should perform this procedure and under what conditions. We also consider presumed reasons for frequently extreme divergences of opinions in the historical span, and the difficulty of expecting clinical recommendations to be supported by evidence at the accepted level.

Key words: frenotomy, history of medicine, lingual frenulum, tongue-tie

STRESZCZENIE

Niniejszy artykuł o charakterze historycznym omawia zmiany w opiniach na temat krótkiego wędzidełka języka (ankyloglosja) na przestrzeni dwóch tysiącleci. Nagły wzrost zainteresowania od 1990 roku wpływem krótkiego wędzidełka języka na funkcjonowanie języka podczas karmienia piersią i w okresie rozwoju mowy odnowił historyczne kontrowersje we współczesnych

uwarunkowaniach. Cechą charakterystyczną było i jest nadal utrzymywanie się różnic w opiniach różnych zawodów medycznych, i w ich obrębie, oraz laików na temat znaczenia krótkiego wędzidełka języka w okresie niemowlęcym, wskazań do jego podcięcia, a także kto powinien ten zabieg wykonać i w jakich warunkach. Rozważamy również przypuszczalne przyczyny krańcowych rozbieżności opinii oraz trudności związane z oczekiwaniami co do wsparcia klinicznych rekomendacji dowodami na odpowiednim poziomie.

Słowa kluczowe: frenotomia, historia medycyny, wędzidełko językowe, krótkie wędzidełko języka

INTRODUCTION

Over the last thirty years, several hundred scientific papers have been published about the lingual frenulum in newborns, the potential developmental consequences of doing nothing and recommendations for either conservative management or surgical release (Bin-Nun 2017). Conferences of organisations providing education pertaining to breastfeeding and human lactation now almost universally have at least some content addressing this topic. A sudden proliferation of articles and opinions about a small part of the human body, which was previously treated as a minor congenital anomaly of no significance, may be puzzling to a medical historian. Was its significance suddenly discovered? What caused this increased interest: new diagnostic possibilities, or perhaps a different perspective on the chain of causes and effects in certain clinical problems? What pathways did doctors' opinions on this topic follow for two millennia? Even a cursory review of contemporary literature confirms the existence of controversies regarding many aspects of the lingual frenum. Representatives of individual medical specialties still differ, also within their own groups, in their views on the impact of tongue-tie in feeding disorders in infants and speech disorders, as signaled in the study of Ann Messner and Lauren Lalakea and other articles (Messner 2000; Levkovich 2017; Jin 2018). Was the question of the frenulum of the tongue clearer in the past? Attempts have been made, previously, to answer these questions. Some authors of paediatric texts about the so-called short lingual frenum evoked the opinions of historical medical authorities, but usually they used a random and very selective basis. The subject rarely seemed important enough to them to deserve a reliable description: 'This is too trifling [a] matter to dwell upon.' proclaimed Michael Underwood (1736–1820) (Underwood 1784). Two noted Polish physicians of the nineteenth century – Jędrzej Śniadecki and Teofil Matecki did not mention it in their otherwise judicious writings on infant feeding and diseases of children. Another author claimed that 'much entertaining nonsense' was written about the tongue-tie (Wallace 1963), a subjective statement.

More recently, historical aspects of tongue-tie are the main topic of two published papers by two retired neonatologists – Peter Dunn (1929–2021) and Michael Obladen, who, due to seniority, can be considered as witnesses to history. Peter Dunn was the first to notice differences in opinions regarding the significance of the lingual frenum between ancient authors, on the one hand, and various medical specialists in the twentieth century who insisted that the frenum had no clinical significance (Dunn 1995). He observed in 1957 that the opinions then in force during postgraduate education were not compatible with his observations in the neonatal ward. His experimental research, the first in this matter and conducted in cooperation with Brian Speidel, supported his conviction about the need for surgical intervention in some cases of tongue-tie (*ibidem*). Michael Obladen did not describe any personal experiences with tongue-tie, and in the final paragraph of his paper acknowledged 'tongue manipulation' as a culturally determined phenomenon among other rites in the neonatal period (Obladen 2010). The authors of the present paper became convinced regarding the clinical significance of neonatal tongue-tie diagnosis and necessary remediation relatively late in their professional careers, TS in 1998 after encountering articulatory problems in his own children and VT in 1995 for professional reasons, since when she has additionally observed feeding difficulties in tongue-tied infants drinking from bottles.

The aims of this study are to supplement the sources given in the above-mentioned medico-historical articles and to describe in greater detail the controversial issues found in the historical context, prior to the twenty-first century. The authors will also attempt to determine the causes of both persistence and changes in attitudes of physicians during the two millennia towards the relevance of the lingual frenum to problems observed in their patients.

TERMS AND DEFINITIONS

The subject of this study is lingual frenum (*frænum s. vinculum linguæ*, string *or* bridle of the tongue, lingual frenulum) (Dorland's 2012; Mosby's 2018). Its modern definitions may be twofold – either anatomical or functional. The former states that it is a tissue fascial fold between the floor of the mouth and the lower surface of the tongue and in connection with the lingual medial septum, and the latter that it is an embryological remnant without any useful function but which may impair tongue functions which are essential for individual development. The less precise descriptions given by various authors in the past usually fall into one of these two concepts. The functional definition implies that the lingual frenum attracts medical interest only when it creates a dysfunction. Authors who are more anatomically oriented will write about tongue-tie, *lingua frænata* or

an abnormal frenum of the tongue (or use respective words in their languages[1], and those more functionally oriented will use various terms derived from Greek-Latin *ankyloglossia*[2], implying "stiffness" of the tongue with 'impeded motion' (Dunglison 1876), or simply "tongue-tie".

While opinions among authors using the functional orientation are broadly compatible, the anatomically oriented authors have had problems in precisely describing the clinical situation they observed. Most of them described the abnormal frenum as being short, without indicating what is actually short in the abnormal frenum, and individual doctors described it as 'bigger than it should be' (d'Acquapendente 1620), long (Zwinger 1722; Roche 1828; Billard 1850; Sołowij 1922; Kowalski 1929) or broad (Butlin 1885). In fact, the frenum is a three-dimensional fold, and it should be described as such even more precisely than it was done by Jakób Szymkiewicz (1775–1818): 'The tongue fraenulum suffers from various defects, it can be too short, too wide, too long or unnaturally thick and fleshy.' (Szymkiewicz 1806). These minor congenital defects were recognized as anomalies of formation processes (Wendt 1835) within a broader term of lingual adhesion (*adhesio linguæ*). Two types of lingual adhesion were distinguished at first: that caused by the frenum and that very rare type with bands fixing the sides of tongue to the walls of the oral cavity (the buccal tie), which was described first by André Levret (Levret 1751; Levret 1781). A complete or partial adherence of the tongue to the floor of the oral cavity – which may be called 'true' lingual adhesion – is extremely rare (Fitzwilliams 1927, 36–37). Actually, that floor in neonates, which serves as an anchor for frena, may further complicate the classification of tongue-ties (Fig. 1).

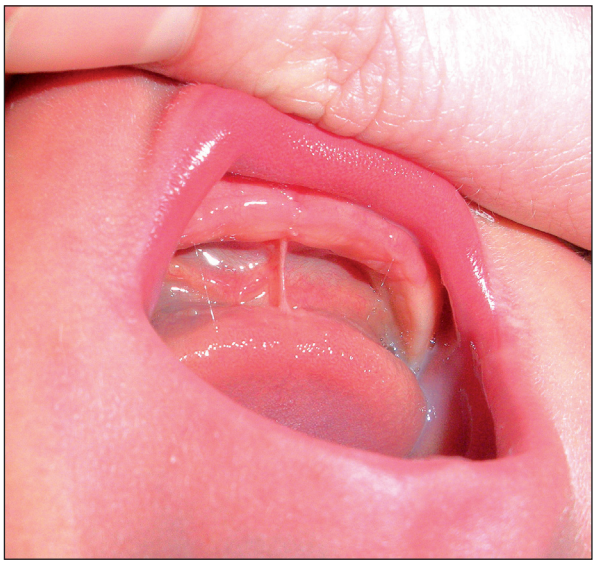

Fig. 1. Oral cavity in a neonate with tongue-tie and transverse mucosal ridge in the floor

[1] In Polish: *krótkie wędzidełko języka*; arch. *przyrośnięcie* or *przyrośnienie języka*, *ścięgno podjęzyczne, plewka*, *łyczko, węzeł językowy*.

[2] In Polish: *ankyloglosja*; arch. *nieruchawość języka.*

The concept of "posterior tongue-tie" appeared in a publication of the American Academy of Pediatrics in 2004 (Coryllos 2004) and Elizabeth V. Coryllos is acknowledged for its introduction[3]. This term is increasingly being used for certain tongue restrictions which have been assessed as negatively impacting breastfeeding, yet which are difficult to identify with commonly used criteria. In their illustrated "Classification of Ankyloglossia" tool, Coryllos and her colleagues were the first to formally describe these as tongues where sometimes the tip can be raised, but the medial part of the tongue remains tethered to the floor of the mouth, which interferes with the coordination of extracting milk from the breast and transferring the milk to oropharynx. We will set aside contemporary controversies regarding this real clinical topic, as the detailed discussion goes beyond the scope of our article. Knowledge of anatomy and mechanics of the human neonatal tongue is still fragmentary (Iskander 2009). However, the enduring discrepancies in anatomical description and terminology are important because they have made it more difficult, through the centuries, to formulate uniform criteria for surgical intervention in tongue-tie management. In consequence, it was hard to assess and compare the results of strategies chosen by single operators, be they attending women, barber-surgeons, midwives or medical doctors.

INCIDENCE OF TONGUE-TIE

Lingual frena were usually only noticed when associated with problems or when these consequences were anticipated by people aware of them and engaged in their prevention. In the past, the opinion prevailed among medical practitioners that tongue-tie is rare or even very rare (Lafontaine 1802; Malcz 1834). Influential early paediatricians, like Nils Rosén von Rosenstein (1706–1773) and David Francis Condie (1796–1875), confessed that they have never met a case of the frenum requiring incision although according to the nurses it was a common occurrence (Rosén von Rosenstein 1771; Condie 1858). It would appear that the physicians saw infants only after midwives had released the frenum. However, the opinion of the German respected midwife Justine Siegmundin was that the frequency was one in a thousand babies (Siegemund 1723), and this opinion was shared in the first paediatric textbook in Polish by Jakób Szymkiewicz (Szymkiewicz 1810). The prevalence was sometimes estimated as one in a hundred newborns (Steiner 1877) or one in ten children suspected by laypeople of having ankyloglossia (Hagen 1784). Earlier, a clinically oriented observer William Moss, a Liverpool physician, thought that 'more or less' one in 3–4 children were tongue-tied (Moss 1794). Recently, with the ongoing disagreement

[3] Genna C.W. Personal communication, 2 July 2022.

on unequivocal criteria, the prevalence of tongue-tie was given as from 0.02–10.7% (Power 2015), the latter figure having been calculated in the first small randomised controlled trial (RCT), in Southampton, in which the condition was consistently defined through use of clinical images used to train staff (Hogan 2005). However, when the criterion is an apparent tongue-tie-related articulatory defect, then speech pathologists might estimate the frequency as even higher in speakers of certain languages, including Polish.

Midwives and laity through the ages had their own, different assessments, which can be deduced from the prevalence of routine frenotomy performed in newborn babies soon after birth. With a few outstanding exceptions in which tongue-tie was consistently defined, midwives did not leave written opinions on prevalence, but we have many statements of their medical opponents in this respect, who criticized routine frenotomy performed by attending women, midwives and wet-nurses well into the twentieth century.

THE SIGNIFICANCE OF THE LINGUAL FRENUM

Problems in breastfeeding and in speech development as negative consequences of tongue-tie have been recognized since ancient times. For the ancients – Celsus (Celsus 1953), Antyllus in the third / fourth century AC (Haeser 1876), Aëtius of Amida (Aëtius n.d.), Paulus of Aegina, Arabian scholars and also Avicenna (Paulus 1846) – hindered speech development was the main problem, although for Galenus Claudius of Pergamon the negative effect on a newborn's sucking skills seemed more important (Galenus 1821). These ancient authors were frequently quoted on tongue-tie by other authors until the nineteenth century. The significance of tongue-tie as an impediment to success in breastfeeding was not mentioned in old texts by the most illustrious early surgeons (Lanfranco da Milano, Ognibene Ferrari, Hieronymus Fabrizio d'Acquapendente, Wilhelm Fabry, Johannes Scultetus) but the surgical incision of 'the String' was accepted with reservations as prevention or treatment of speech disorders. Yet the prominent early French obstetricians – Jacques Guillemeau (1550–1613) and François Mauriceau (1637–1709) – were aware that "the String" can hinder latching on the breast and sucking effectively (Guillemeau 1635, Mauriceau 1673). It is not known whether the same opinion was universal among barber-surgeons (Read 1650).

In the eighteenth century, it was mostly surgeons who published prolifically on tongue-tie and wrote about its significance in disturbances of breastfeeding**,** to be resolved by incision of the abnormal frenum. Pierre Dionis, Jean-Louis Petit, Lorenz Heister, Jean-Charles Desessartz, and Carlo Spagnolini had the greatest

impact. Possible articulatory consequences of tongue-tie were left to be solved eventually later in childhood, and have remained a bone of contention even today among opponents of frenotomy. There were only occasional mentions of specific tongue-tie-related articulatory defects, for example, the defective pronunciation of the /l/ and /r/ phonemes (Hervás y Panduro 1789). In the Polish-Lithuanian Commonwealth, ankyloglossia had been briefly mentioned by single authors: Jan Piotr Różański, Johann Raphael Steidele, Jacques Ballexserd, Józef Rafał Czerwiakowski, Teofil Teodor Weichardt. In their usually short texts published in Polish they emphasized the most important aspects of tongue-tie management, specifically the indications for frenotomy and safety of this procedure. A uniform opinion on the negative effect of tongue-tie on breastfeeding could be observed in the Polish and Lithuanian lands until the mid-nineteenth century.

From the mid-nineteenth century opinions began to prevail in written European and American sources that there was no important clinical significance of tongue-tie. Consequently, the topic disappeared from many paediatric textbooks published in the next one hundred years, also in Poland, or it was mentioned briefly with the usual remark that the lingual frenum had no influence on breastfeeding and bottle feeding, and stating that frenotomy is unnecessary (Reuss 1922; Ostapiuk 2006, 39–40, 45–46; Obladen 2010, 87). These prevailing opinions corresponded with the decrease in the prevalence of breastfeeding until the 1970s and heavily influenced also the next fifty years during which programs of breastfeeding support were successfully organized worldwide. Yet bottle-fed infants with identified tongue-tie also may feed poorly prior to frenotomy (Hogan 2005).

With more women initiating breastfeeding in the last quarter of twentieth century, it was rediscovered that there are certain newborn babies who are unable to suck efficiently**,** some of them because of tongue-tie (*ibidem*). Nonetheless, divergent opinions on the significance of tongue-tie persisted, not only among different medical specialists who could be potentially involved in treating tongue-tie, but also among members of the same specialty (Bell 1809; Kupietzky 2005) and this continues today (Jin 2018). Since some children with tongue-tie breastfed well, this fact has been used to fuel the controversy. That created a lot of confusion regarding a possible influence of tongue-tie on impairing sucking of the breast in infancy, speech development and articulation, influencing also relations between these clinicians and the mothers of tongue-tied children. In the twenty-first century, research by ultrasonographers in Western Australia has provided dynamic images revealing differences in tongue function in relation to ankyloglossia (Geddes 2008; Garbin 2013). Further quotations of multiple recent investigations indicating the significance of tongue-tie are beyond the scope of this historical article. As there is no reason to suspect that the frequency and features of tongue-tie has changed

in humans during the last two millennia, it means that many children and mothers suffered because of differing medical opinions and ignorance.

THE DIAGNOSIS OF TONGUE-TIE

The importance of examination of the tongue in newborn babies was presented in verses on nursing and rearing children written in Latin by Scévole de Sainte-Marthe (1536–1623) (de Sainte-Marthe 1595):

> Do examine the tongue with particular care,
> since this precious gift
> the loving heavens have sent to the human race.
> How useful it is to the babes
> when they suck milky dew from the breasts[4].

Examination of the tongue at birth was seldom advised in old writings (Wedel 1717; Perzyna 1790; Czerwiakowski 1804; Dziarkowski 1819). The above-mentioned opinions on the definitions and prevalence of tongue-tie in newborns demonstrate that the authors differed substantially in their definitions of the physical features of tongue-tie, and hence in their classification systems regarding diagnosis and management. The main features of a tongue-tie were well summarized in Poland by Juljan Moszyński, with a focus on function or its lack: 'A baby cannot suck breasts well, he does not put out his tongue, he can not raise his tongue upward; when lifting up the tongue with the end of a teaspoon one sees a bridle underneath, reaching almost to the tip of the tongue; on top, along the tongue, a groove is made when opening the mouth.' (Moszyński 1855). Non-specific behavioural symptoms were added sometimes: the infant does not latch on well, looses the nipple and expresses 'impatience and annoyance' (Nicolas 1788), the newborn 'coughs since he does not swallow because of tongue torpidity' (Seeman 1843), 'he can not cry, only squeaks' (Mianowski 1818), and a clucking noise can be heard on sucking (Dewees 1834). More accurate descriptions of the behaviour at breast of a baby with a sucking problem appeared late in the twentieth century.

In the past, the sublingual area was usually inspected in infants only visually during natural movements and with the help of a spatula, flat spoon-handle or special forks and spatulas which were useful also for eventual frenotomy (see below). An examination of the sublingual area with a finger (Ballexserd 1774; Lafontaine 1802; Czerwiakowski 1804; Szymkiewicz 1810; Orkisz 1835) or a test of lingual movements by an inserted finger were seldom recommended (de la Faye 1771; Petit 1774, 264) until recent times. Squeezing the infant's nose was

[4] Another English translation by H.W. Tytler in 1797 does not faithfully render this passage of the poem.

advised in order to achieve his opening the mouth. The discovery that certain symptoms (nipple pain) and signs (characteristic changes in the nipples) in the breastfeeding mother could be caused by tongue-tie in the suckling was an achievement of contemporary clinicians engaged in promotion of breastfeeding. In recent times identifying a clinically significant tongue-tie should be no problem for an educated lactation consultant or physician with due care.

For comparative studies of treatment of tongue-tie the need for precise diagnostic criteria has been called for by many contemporary authors who desire agreement on such criteria. Several scoring tools exist nowadays to guide identification of who would, and who would not, benefit from tongue-tie release, that is, to prevent under- and over-diagnosis (Hazelbaker Assessment Tool for Lingual Frenulum Function, Lingual Frenulum Protocol with Scores for Infants, Bristol Tongue Assessment Tool, Tongue-tie and Breastfed Babies). Their usefulness for clinical practice, teaching and research is still subject to critical evaluation, and discussing their applicability or reliability is beyond the scope of our article.

INDICATIONS FOR THE MANAGEMENT OF TONGUE-TIE

We shall focus here mainly on the opinions and recommendations from the Polish sources but exactly the same opinions and recommendations can be found in publications written in other languages in corresponding times. The main indication for the surgical intervention in infants was well presented by the English surgeon Samuel Cooper in the early nineteenth century: 'It is highly necessary for every surgeon to know that in infants an incapacity to suck in consequence of the evidently immoveable [sic] state of the tongue caused by the above defects [malformed frenum] forms the only just ground for dividing the frænum.' (Cooper 1813). The Polish texts were generally short, so the authors did not discuss the problems of tongue-tie in detail, but usually emphasized only what was decisive in their opinion. Incision of the lingual frenum – frenotomy – should be performed when the frenum 'puts the sucking in the hindrance' (Malcz 1834), 'if the infant does not protrude the end of the tongue to the lower lip, and if the end of the tongue makes a pronounced notch at the movements' (Litzmann 1879) or 'only when the frenum is not only short, but thick and fleshy, tied to the tip of the tongue' (Erlichówna 1930, 716). The most frequently quoted author in support of such indications in earlier world literature seems to have been the French surgeon Jean-Louis Petit (1674–1750) whose writings on tongue-tie remained the most comprehensive available until the end of the nineteenth century. He observed critically that half of the newborn babies sent for frenotomy had no indications to perform it (Petit 1774, 263). Other sensible texts were

published throughout the nineteenth century, although of a limited international impact; we should indicate here at least these written by James Blundell (1791–1878) (Blundell 1834) and William Fairlie Clarke (1833–1884) (Clarke 1873). Both these authors also described the potential developmental consequences of tongue-tie. Blundell wrote: 'If a child be suffered to grow to the age of eight or ten years before the tied tongue be liberated, it may never afterwards acquire a free use of the organ, an occurrence indicating criminal neglect.'. A possible causal connection of tongue-tie with articulatory defects was considered already by the ancient authors, as noted above, but the developmental aspect of tongue-tie and preventive effects of early frenotomy were subsequently seldom mentioned in this respect (in Poland only by Hipolit Terlecki (Terlecki 1835). Writing in 1876, the authoritative Austrian paediatrician Andreas Hüttenbrenner was convinced that the frenum had no influence on the development of the tongue (Hüttenbrenner 1876), as was Władysław Ołtuszewski, the first Polish specialist in disorders of speech (Ołtuszewski 1901). The opinions of Polish speech pathologists and ENT specialists in the twentieth century were mostly similar and were described in greater detail by Barbara Ostapiuk (Ostapiuk 2006, 39–40, 45–46).

The incision of the frenum was contested, especially if performed routinely and by midwives and common people, from the seventeenth century until the early years following World War II. Explicit statements that there is no indication for frenotomy in newborns were sometimes enunciated from the beginning of nineteenth century (Denman 1805) and widely promulgated from the middle of the twentieth century until the present. Routine frenotomy was usually considered a wrongful practice (Berger de Lonchamps 1789), folk superstition (Lafontaine 1802; Dziarkowski 1819) and contrary to common sense (Malcz 1834). The fact that certain babies with tongue-tie suckle without difficulty, although articulatory problems may appear in the future, was rarely emphasized (Dziarkowski 1819). Other arguments could be questioned on ethical grounds: it could be performed because other doctors do it, or because it will be done anyway by an ignorant person (Denman 1805; Hüttenbrenner 1876; Kossakowski 1949), or a mock procedure should be performed because 'Rural women even when there is no tongue-tie in their children, they are convinced it is present' (Simon 1860). The attitude of Alfred Vogel (1829–1890), a distinguished German paediatrician, was indulgent:

> The frænum is divided hundreds of times where it is once really indicated. But as this operation when performed by a steady hand is totally devoid of harm it is therefore not necessary to look so strictly for the indications especially if any comfort can thereby be conferred upon the patients. (Vogel 1870)

WHO IS TO INTERVENE IN TONGUE-TIE?

A common sense medicine since ancient times allowed attending women and midwives to intervene at birth in a case of suspected tongue-tie. In the opinion of de Sainte-Marthe, a midwife was allowed to perform frenotomy in the absence of a surgeon (de Sainte-Marthe 1595). The influential physician Jean Astruc (1684–1766) was probably the last author who accepted that 'The midwives prudently anticipate the physicians or surgeon's advice in this case [of vicious conformations];' (Astruc 1746). However, voices criticizing this practice by women, particularly when performed on newborn infants routinely and with a "dirty fingernail", were raised in Poland at least since the eighteenth century (Berger de Lonchamps 1789; Struve 1797; Lafontaine 1802), but it is unclear whether this censure was based on eyewitness experiences or caused by frictions among emerging medical specialists. Most of the medical authors agreed that frenotomy should be performed by a surgeon (Struve 1797; Lafontaine 1802; Orkisz 1835) or barber-surgeon (Ballexserd 1774; Steidele 1787; Perzyna 1792; Czerwiakowski 1804; Maleszewski 1848) and, in the nineteenth century, also by a physician (Malcz 1834; Pfau 1838; Fijałkowski 1842; Seemann 1843). They advised that whoever was it should be skilled and prudent (Steidele 1787, 393). Nineteenth-century opinions further eroded the active role of midwives and attending women regarding the frenum in neonates, leaving them with a screening role only (Litzmann 1879). However, in the early twenty-first century, there are signs of a changing attitude: namely, the training and endorsement of selected staff midwives who have also achieved the International Board Certified Lactation Consultant credential (IBCLC), to perform straightforward frenotomies, within the hospital (Hogan 2005, Amir 2016,), and a British paediatric hospital's endorsement of a senior ENT nurse to perform frenotomies (Rose 2015). Frenotomy and frenectomy are included in dental practice in certain countries (Kupietzky 2005), including Poland.

INTERVENTION

For a long time, the most common method of treatment of tongue-tie in newborns was probably rupture of the frenum with a finger (Fonklofen 1724) or a fingernail, the method already recommended by Galen (Galenus 1821). Midwives left one of the fingernails uncut especially for that purpose which was generally accepted (Wittich, 1596) but also had fought against with disgust as unhygienic and improper by Girolamo Fabrizio d'Acquapendente (1537–1619) (d'Acquapendente 1620). Subsequently, that procedure was condemned by numerous authors, although it was still in dispute in the Polish lands even

in the nineteenth century (Malcz 1834). Instead of the fingernail, a small, sharp-edged coin was used in Italy (Vittori 1557; Ferrari 1577), a clean pfenning in the German lands (Wittich 1596) while in England it was a groat (Pechey 1697) which corresponds in its function to a Polish 'trojak' of King Stefan Batory. Galen also mentioned a ligature of the frenum (Galenus 1821), the method recommended especially for 'fleshy' frena (Avicenna 1583, Guillemeau 1635). William Smellie recommended cauterization with lunar caustic (silver nitrate) or Roman vitriol (iron sulphate II) for a 'fleshy' frenum, in addition to manual stretching (Smellie 1766). Generally, an incision of the frenum was recommended, and performed with a lancet, a small curved bistoury or – as favoured by most surgeons – blunt-ended scissors, possibly bent laterally. A golden instrument could be heated for the procedure (Lanfranco da Milano 1519). Special instruments for frenotomy were devised by certain ingenious doctors – Jean Petit (Petit 1774, 278), the tongue-tied polymath Thomas Young (Moss 1794) and others – in order to diminish the risk of excessive incision but never became popular. According to older authors, the incision could be replaced by lubrication of the frenum with a liniment (Sharp 1671; Culpeper 1755).

As depicted in classical textbooks on surgery, two main methods were employed for performing frenotomy: a baby was usually well wrapped to prevent movement, and either the tip of the tongue was pulled out with two fingers and held towards the palate with the frenum stretched, or the tongue was held upwards by means of a spatula or, better, by a two-toed blunted fork (Scultetus 1666, 67; Heister 1743) or a grooved director (*sonde cannelée*), whose handhold with a middle opening is used to lift up the tongue (Croissant de Garengeot 1723, 387; Petit 1774, 277; see Fig. 2 and 3). The latter method allows a good visualization of the stretched frenum, which diminishes the risk of excessive incision. The open wound, usually of a diamond shape, was commonly washed with wine or with vinegar diluted with water, and treated with rose honey (*Rhodomel*), individually prescribed in barley water (Astruc 1746; Anon. 1774; Szymkiewicz 1806), or with a small quantity of alum and red ochre powder (*styptica* for prevention of bleeding) (Zwinger 1722), and then healed without any further application. Nowadays the wound itself is generally left without any treatment, "leaving the healing to the infant's saliva and the nurse's milk" (Bacheracht 1750).

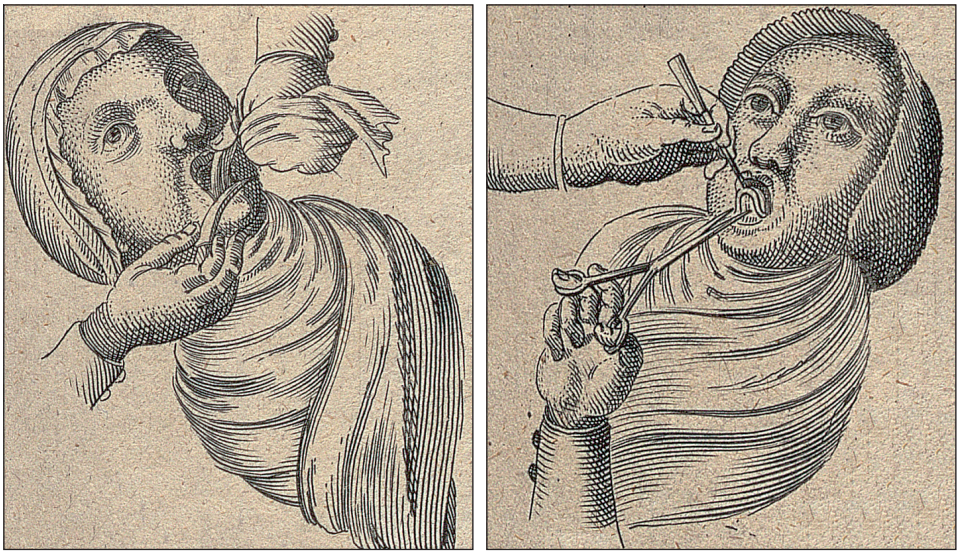

Fig. 2. Two methods of frenotomy according to Lorenz Heister after Johannes Scultetus – (Heister 1743, pl. XXI, fig. 1 and 2). Courtesy of the Jagiellonian Library, Cracow.

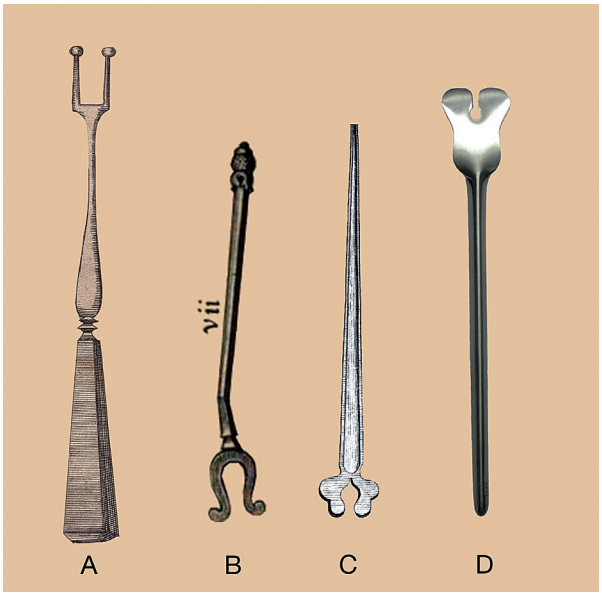

Fig. 3. Instruments used for frenum inspection and frenotomy: A. Fork depicted by Lorenz Heister (Heister 1743, pl. XXI, fig. 3); B. Fork depicted by Johannes Scultetus (Scultetus 1666, pl. XI, fig. VII); C. sonde cannelée depicted by René-Jacques Croissant de Garengeot (Croissant de Garengeot 1723, 120); D. Modern grooved director. In public domain, free access

After the advent of anaesthesia the procedure in newborns usually continued to be done without anaesthesia. In recent years, surface anaesthesia is sometimes used although frenotomy without analgesia was approved in individual recommendations and institutional or national guidelines (NICE 2005).

In olden times, the physicians who advocated frenotomy for tongue-tied newborns were aware that not performing this operation could bring even a threat of death to the infant because of failure in breastfeeding (d'Acquapendente 1620; Szymkiewicz 1810). However, there were always proponents of a passive attitude to tongue-tie in newborns and children. Such an attitude was obvious for those who claimed that there are no short- or long-term adverse effects of tongue-tie. Others thought that the problem resolves with age, believing that the frenum stretches itself due to constant tongue movements. It may occur over a longer time that a membraneous frenum might stretch and not hinder speech development. This process is claimed to be enhanced by manual therapy offered at present by physiotherapists, osteopaths and chiropractors. Such management, as an alternative to frenotomy, was rather unprecedented in history.

THE AFTERMATH OF FRENOTOMY

Frenotomy in newborns and infants was regarded as a highly effective procedure: 'After cutting such fibres the child immediately begins to suck' (Szymkiewicz 1806). The effectiveness regarding sucking has been generally observed by the nurses and, today, by lactation consultants, and was confirmed by numerous investigations in the last twenty years (Żukowska 2019), although much of the current literature has a relatively low Level of Evidence from the perspective of evidence-based medicine (EBM) (CADTH 2016; O'Shea 2017, 17–18).

Historically, frenotomy was also considered safe, although many authors usually emphasized the necessity to perform this seemingly trivial operation by a skillful person and with due care. The most serious complication of unskillfully performed surgery was severe bleeding after inadvertently cutting the sublingual vessels in situations like that described by Hendrick van Roonhuizen (van Roonhuizen 1674), Pierre Dionis (Dionis 1708), François Mauriceau (Mauriceau 1738), Jean Petit (Petit 1774, 282) and others (Roche 1828; Eberle 1833; Butlin 1900, 31–32). To mitigate that dramatic risk, a clear view of the frenum, when being cut, was essential (Petit 1774, 275–276), the incision should not be performed late in the day (van Roonhuizen 1674), 'Sharp-pointed instruments ought never to be used for the purpose [of frenotomy]' (Eberle 1833) and the baby should be observed for some time after the procedure regarding bleeding from the wound. The same single cases of death following a haemorrhage were subsequently

repeated by other authors as a warning. Some of the cases might have indicated an inborn disorder of blood coagulation as the primary cause (Rieken 1829; Obladen 2010, 86). In the present century, protocols for tongue-tie release typically include that the infant has received vitamin K.

Inexpert hands performing frenotomy may also cause trauma to salivary submandibular and sublingual ducts (Zwinger 1722; Callisen 1790). Ranula in a newborn is very rare nowadays, being mostly of congenital origin, but that anomaly of non-uniform origin was frequently described by authors in past centuries in relation to tongue-tie in infants. The likelihood that an unskillful manipulation during routine frenotomies in the past could have lead to a minor trauma of delicate salivary ducts and sometimes subsequently contributed to ranula seems not insignificant (Wedel 1717; Obladen 2010, 86). Yet an even more serious trauma to the sublingual area could happen 'in the birth, from the accoucheur's helping the head along with his finger in the child's mouth.' (Smellie 1766). Two other complications mentioned by authors of the past were inflammation of the wound (d'Acquapendente 1620; Smellie 1766; Berger de Lonchamps 1789; Czerwiakowski 1804) and collapse of the tongue backwards (*linguæ revolutio ad fauces*) from excessive undercutting that could lead to suffocation (Petit 1774, 267; Wendt 1835; Butlin 1900, 33–37). In the latter case, not seen since those reports, a finger should be put into the mouth to restore the right position of the tongue, or even a surgical fixation of the tongue could be necessary. In the opinion of Petit, the most serious risks happened to 'bold' operators after an excessive incision, while 'timid' operators did not undercut enough (Petit 1774, 282). Frenotomy in the present-day conditions of neonatal wards and outpatients clinics has generally been confirmed as safe when performed by trained operators (Sioda 2012, 122; Mettias 2013; CADTH 2016, 13; Srinivasan 2018), yet reports on usually minor complications remind us to exercise due diligence. Severe injuries to the tongue at frenotomy, requiring surgical repair, also happened occasionally in the twentieth century (Kossakowski 1949).

Recurrence of ankyloglossia after frenotomy is relatively common as a result of recrudescence of the incised frenum with formation of the cicatrix, a problem that was noticed in the old texts on frenotomy. A method of prevention of consecutive frenotomies by a regular massage with a finger of the deep sublingual area was recommended incidentally (Guillemeau 1635; Fabricius von Hilden 1646; Mauriceau 1673; Zwinger 1722; Anon. 1772; de la Faye 1771; Callisen 1790), and this practice has re-emerged in the twenty-first century, becoming a current controversy as to the effectiveness, method and duration of treatment.

THE BASIC CAUSES OF HISTORICAL CONTROVERSIES REGARDING THE LINGUAL FRENUM

It is hard to find any better example, than that of the lingual frenum, of medical disorientation persisting until contemporary times despite the fact that the presenting situation and its potential after-effects were always easy to observe without any expert knowledge and instruments. It is also hard to find another medical problem regarding which the ancient authors and lay people were nearer the truth as to the existence of a physical problem, and many highly educated medical specialists in the twentieth century were over-cautious. Certainly, the foundational causes of controversies described above depended on the historical epoch – controversies had arisen mostly between representatives of different epochs, because what was common and agreed upon at one time, might not have been accepted at another time due to changed conditions of life and work. In the meantime, fluctuating knowledge of human health shifted the attention of physicians and scholars to other themes and activities, also changing their insight into the potential problems created by tongue-tie. Tongue-tie could be considered sometimes as more or sometimes as less an important medical problem depending on the actual attitude towards the treatment of infants with breastfeeding problems and the treatment of children with speech disorders. For the mothers and nurses, these problems were always important. Numerous authors who lived and worked in different times, and are quoted in this article, confirm a very strong pressure exerted on them by the nurses and mothers regarding a real or imagined tongue-tie in their infants.

For many centuries until the nineteenth century newborn babies were under the care of midwives and attending women who were allowed to use only their hands in their work, and no instruments except urinary catheters. To call a barber-surgeon or surgeon to perform frenotomy was a significant trouble and expense, and so it is no wonder that midwives used their fingernails to rupture the frenum. The emergence, especially in the second half of the eighteenth century, of new medical professions like man-midwives (accouchers) and specialist physicians, who no longer avoided performing minor surgical procedures, changed the situation. The problem of tongue-tied newborn babies, hitherto dealt with rather routinely, came under their scrutiny at least in the affluent social groups under their care. Thereafter, and throughout the nineteenth century, the basic causes of controversies over tongue-tie gradually shifted from the general level of domestic medicine to the level of individual contacts between medical professionals and their patients.

In these individual contacts, the transfer of knowledge was unidirectional until the advent of the internet era: from the medical professionals to the patients,

and in the case of tongue-tie, mostly to the infants' mothers. Such relations meant the importance of personal characteristics of providers of knowledge. They were exponents of certain concrete opinions, sensible or unreasonable – as we have demonstrated above – which more or less influenced the mothers, the families and, possibly, also other medical professionals. This arose from their possession of positive cognitive traits, like sagacity, openness to new information, the sense of observation, and negative traits, like lack of knowledge and ignorance of that lack, or a narrow-minded approach to medical quandaries. In the case of tongue-tie, the catalogue of wrongful attitudes to the problem may begin with the attitude of "much hack of nothing", a narrow, fragmentary approach to the problem, and not considering developmental aspects of individual human existence and the complexities of anatomy.

The lack of observational ability among scholars – 'he looks, but does not see' or 'perceives wrongfully' – was already emphasized by Albrecht von Haller (1708–1777) (Sontag 1983). We may only speculate whether character traits such as lack of humility, false ambition and shame that prevent weighing the attitude against the facts, could also play a role in the writings and recommendations of certain authors. We have encountered in the world literature prior to the current century only three avowals to changing from opposing frenotomy to acknowledging it has its place. Duncan C.L. Fitzwilliams (1878–1954), a London surgeon, confessed: '[...] I worked among children after being brought up among those who denied the existence of tongue-tie. I soon found out my mistake.' (Fitzwilliams 1927, 37). The second was uttered by the noted Polish paediatric surgeon Jan Kossakowski (1900–1997), who changed his opinion from negative to positive after twice having to suture the bleeding tongue after a botched frenotomy – and realizing it was generally impossible to convince mothers that frenotomy was unnecessary (Kossakowski 1949). The third was the English paediatrician I.M. Cullum's change of opinion after he performed frenotomies on two infants who struggled to extract milk from the bottle, but thereafter fed well (Cullum 1959). All the above-mentioned cognitive traits seem to have been of universal significance in ancient times and today. Without appropriate support from the medical professionals consulted, the mothers of tongue-tied babies have either found somebody else who did perform frenotomy, or, in more recent times, turned to bottle feeding.

An unproven but possible source of misconceptions regarding the significance of a restrictive lingual frenum is the confusion created by imprecise terms for abnormal frena in classical Greek, Latin and modern European languages alike. The observation by Władysław Biegański (1857–1917), a Polish physician and philosopher, applies here very well: 'History teaches us repeatedly that a well-chosen term of the disease has a great influence on the further direction

of investigations; in fact, the term crystallizes in itself the entire content of a given concept.' (Biegański 1900). The Polish language, as in writings by Jakób Szymkiewicz**,** had for tongue-tie a pertinent term which means a weakened mobility of the tongue[5] but that term was not promoted and medically accepted, and today sounds archaic.

Currently, a serious quandary among medical professionals exists in accepting the true consequences of tongue-tie – especially sucking dysfunction in infancy and subsequent articulatory dysfunction in childhood and adulthood, all with subsequent problems – because of the lack of studies meeting the requirements of Level of Evidence 1 in EBM (CADTH 2016; O'Shea 2017, 17–18). It should be noted that there are intrinsic difficulties in designing and conducting double blind investigations on tongue-tie in children because of ethical and practical barriers. There is also a lack of uniformity in technique and in instruments used because of preference. All these hindrances are obvious to professionals with convictions based on everyday experience to whom withholding frenotomy from a group of newborn infants chosen as controls seems strongly unfair on ethical grounds, knowing the possible life-long sequelae in individual cases. We should mention here at least the known consequences of artificial feeding in infancy (diseases of civilization, occlusal defects). Jack Donati-Bourne et al. demonstrated that delay beyond 4-weeks from referral to assessment of neonatal tongue-tie is more likely to be associated with abandonment of breastfeeding (Donati-Bourne 2015). It strongly suggests that there may be a critical time for frenotomy which should be followed to increase the chance of success in breastfeeding. If frenotomy is offered to control cases not earlier than after the conclusion of the study, the delay is necessary for the proper assessment of results what preserves the potentially harmful condition in this group and may be significant in susceptible infants (Todd 2015).

As an improved tongue mobility after tongue-tie division in children does not guarantee an intrinsic improvement in pronunciation quality (Ostapiuk 2013[6], Wang 2022[7]), a hypothesis arises that the timing of tongue-tie division in the course of ontogenetic speech/articulation development is a critical factor. Early intervention for the prevention of speech disorders is emphasized widely in speech pathology publications. The theory, standing behind the supposition of a beneficial impact of early tongue-tie division in the period of acquisition of phonetic-phonological competence in the child, is based on the critical period

[5] This is: *nieruchawość języka* (Szymkiewicz 1810).

[6] The investigated group consisted of children older than 2 years and adults to the age of 29 years, all speaking Polish.

[7] The review concerned studies of study groups composed almost entirely of English-speaking individuals.

concept, natural plasticity of the brain in the early stages of development, and an early opening of developmental perspectives.

In the first monograph on the diseases of the tongue in 1873, W.F. Clarke explicitly warned against delay with regard to articulation:

> [The surgeon] should not operate [on tongue-tie] unless he sees good reason. But if the child be really suffering from this congenital imperfection, it ought certainly to be rectified in infancy. If this is not done, it may be long before he is able to speak at all, or he may acquire a defective mode of utterance which no subsequent operation can serve wholly to remove. [...] If, unfortunately, a person having such a defective formation of the mouth is allowed to grow up unrelieved, an operation may be undertaken at any time in order to give the organ more mobility, and much may be done by instruction to induce a clearer and more distinct articulation ; but the cure will never be so complete as if the operation had been performed in infancy. (Clarke 1873, 48).

CONCLUSION

We have traced trends in opinions about tongue-tie across two millennia, up to the sudden resurgence since 1990 of interest in the potentially negative effects of a restrictive frenum on tongue function for breastfeeding and later articulation, in particular. This includes differences of opinion, between and within different professions, over doing nothing *versus* performing the minor procedure of dividing the frenum, and the potential developmental sequelae of delay. The problem of tongue-tie in newborns should not simply be dismissed on the grounds that it cannot be investigated according to current EBM standards of evidence. An attentive reader may notice that every strategy in tongue-tie management, which has been proposed or promoted nowadays, already had its precursor in the past times who had given his opinion founded on his professional experience and sometimes supported by chosen authorities from his past. It seems that closing eternal controversies regarding tongue-tie will be difficult unless different Hierarchy of Evidence pyramids are agreed on to incorporate traditionally acquired knowledge into modern evidence-based medicine, specifically to enable clinicians to meet the needs of tongue-tied human individuals better.

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