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REVIEW ARTICLE

Infant dental enucleation: A literature review on a traditional remedial practice in East Africa

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Abstract

Objective. To summarize and review the literature on infant dental enucleation, a traditional remedial procedure prevalent mainly in East Africa. ***Materials and methods.*** Literature searches were made electronically using general and specialized search engines. The main search was performed through a systematic strategy in PubMed, comprising tabulated keywords, search codes, and translated and transliterated terms. Criteria for the selection of studies were designed to provide a general understanding of the procedure. The findings were synthesized into two sections: a summary of the population prevalence studies, and a thematic literature review. ***Results.*** An overview of the known prevalence and clinical specifications was established. Insight was gained into the purpose of the procedure and the factors influencing its performance. Diverging suggestions were seen with regard to the rationale for use of infant dental enucleation between different populations. Moreover, reports of complications and consequences involving the general health and the dentoalveolar structures of patients were examined. ***Conclusions.*** Prevalence, clinical features and risks of infant dental enucleation point to a need for greater awareness of the procedure. Further, there is a need for specific guidelines regarding management of suspected cases within dental and healthcare communities.

Key Words: *African traditional medicine, attitude to health, cuspid, post-operative complications, tooth germ surgery*

Introduction

Theories of infant morbidity caused by teething have been observed to go back at least 5000 years [1]. Four symptoms of potentially fatal conditions have emerged as central—convulsions, diarrhea, fever and vomiting [1–4]. With child mortality rates and the opinions of the contemporaneous medical practitioners regarding teething it has been suggested that the link between teething and the risk of morbidity and death was easily established in the public mind [5,6]. As such, teething was documented as a cause of death in Europe and the US [3,6,7]. The theories were often accompanied by remedial suggestions, including use of animalistic and botanical substances, and contemporary therapies [1–3,5,6]. A new procedure was introduced in the 16th century, which comprised lancing of the gingiva covering unerupted tooth germs in infants [6]. Even though the procedure was not without controversy, it spread and became common within Western medicine [3,5]. Lack of gingival lancing was used as an explanation of high infant mortality. No records of lancing complications have been found. It is suggested that even though some doctors doubted the benefit of the method, the general opinion was that the procedure itself was not dangerous [6]. With the advancement of medical science the perceived need of lancing has been increasingly questioned by the scientific community [8].

Nonetheless, public perception of teething as an event associated with morbidity continues in other regions of the world, with serious consequences for the oral and general health of the child. Such is the case of infant dental enucleation (IDE), a traditional remedial procedure comprising the removal of unerupted dental germs in infants [9–13]. In 1969 the

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Table I. Keywords in columns used in the first stage of the search strategy to form phrases.*

Milk	Tooth	Bud	Extraction
Primary	Canine	Germ	Extirpation
Deciduous		Follicle	Enucleation
Oral			Excision
Dental			Gouging
			Mutilation

*All searches were conducted as described in the methods section and also by preceding each search phrase by the keywords *traditional* and *infant*, individually and simultaneously.

practice was brought to the attention of the scientific community through publication of a population study [9]. Since then, cases have been shown to exist in East Africa and among immigrants from the area [14]. A summary of population studies has been published in a clinical report by Johnston and Riordan [15] and the British oral health charity organization Dentaid (http://www.dentaid.org) has worked to raise awareness of the subject through journal publications [16,17] and through an active document on their website. Otherwise, little is known about the procedure and its background. In this study, by reviewing the literature on IDE, we aimed to summarize current knowledge about and enhance understanding of the practice.

Methods

Literature search strategy

All searches were conducted electronically through databases on the Internet. First, a broad pre-search was performed to gain insight into the names used for and the terms describing the procedure and the availability of information on the subject. The primary database used was PubMed. Secondary databases used in the pre-search were the specialized search engines Anthrosource, Google Scholar and Scirus. No generally accepted term could be found. Instead, the search showed different terms in use. This step facilitated the identification of commonly used keywords (MeSH) and subject terms. To conduct a systematic search in PubMed as comprehensively as possible, the strategy was divided into the two following steps. First, the identified keywords were organized into columns based on descriptive similarities (Table I). These were combined into phrases in the following manner: one keyword was chosen from each column to form a search phrase. The step was completed by additional searches using the following three search phrases coded by PubMed search codes: (dental OR oral) AND mutilation, (traditional AND infant) AND (tooth OR canine) AND (extraction OR enucleation OR excision OR extirpation OR gouging), (traditional OR infant) AND (tooth OR canine) AND (extraction OR enucleation OR excision OR extirpation OR gouging) and (oral OR dental) AND mutilation. Step 1 produced 682 references. In the second search step, we used identified, native, translated and transliterated terms occurring in the literature found in the first step (Table II). Where there was more than one word in the translated or transliterated name, the whole term was searched within quotation marks. The second step produced 335 references. Then, we merged the search results in PubMed to eliminate duplicate references. The resulting number of unique references was 1012. The search was done for all published literature up to May 2012.

Selection and evaluation of studies

Selection and evaluation were performed by one of the authors (JB). To assess the relevance of the studies, a general review of the references was made, based on the titles and the abstracts. Only studies in English with human subjects were selected (n = 575). Studies found that were unrelated to the subject were eliminated. In dubious cases, the author conducting the search obtained a copy of the original

Table II. Transliterated and translated terms used in the literature to describe IDE.

Transliterated terms used in the literature to describe IDE. Meaning and origin within parentheses	Translated terms used in the literature to describe IDE
Bimeno ya barefugie (refugee teeth; Congo) [35]	False tooth/teeth (Sudan, Tanzania and Uganda) [13,23,33]
Ebiino, ebinyo, bino (Uganda) [24]	Fox tooth/teeth (Somalia) [30]
Ebino, ibino (false teeth; Congo, Tanzania and Uganda) [23,33,35]	Killer tooth/teeth (Ethiopia and Sudan) [26,31]
Gidog (Uganda) [24]	Lugbara tooth/teeth (Sudan) [13]
Gira kwanya (that which is removed; Uganda) [46]	Nylon/plastic tooth/teeth (Kenya and Tanzania) [29,32]
Ikela/Icela (Uganda) [24]	Refugee tooth/teeth (Congo) [35]
Ilko dacowo (fox teeth; Somalia) [30]	Worm/maggot tooth/teeth (Kenya and Uganda) [29,55]
Lake jo marak (Uganda) [24]	
Lakijo marach (bad teeth; eastern Uganda) [46]	
Meno ya nailoni/meno ya plastiki (nylon/plastic teeth; Tanzania) [33]	
Telak (northern Uganda) [48]	
Two lak (Uganda) [24]	

article and/or consulted the other authors. At this stage 41 references remained. The reference lists of the studies that remained were examined for further and yet undiscovered studies. Due to the limited number of studies found on the subject, the inclusion criteria were set to provide an extensive understanding of IDE: Studies were included that used cohort, cross-sectional and case-report designs, as well as qualitative approaches. Also included were clinical reports, editorials and correspondence. Summaries, newsletters, studies with no new information (those that summarized or referred to previously found studies without providing any new data) and secondhand data presentations, such as oral communications, were excluded from the review. Exceptions to the criteria were made for three sources, due to the value of the information: a case report in Danish, with a Danish literature review, published in the journal of the Danish Dental Association (*Tandlaegernes nye landsforening*), showing prevalence of IDE in Denmark [18]; a case report in Norwegian, with a Norwegian literature review and an English summary, published in the monthly journal of the Norwegian Dental Association (*Den Norske tannlegeforening*), showing prevalence of IDE in Norway [19]; and descriptive conventional articles published in the biannual journal *Fédération Dentaire Internationale*, indicating prevalence of IDE in Rwanda [20], and highlighting IDE [21,22]. Overall in the systematic search, 38 IDE-related studies according to the criteria were found. Together with relevant studies and publications found in the pre-search stage and in the reference lists, a total of 57 publications were found to be relevant for this review.

Data synthesis

Due to the selection strategy, the studies chosen were heterogeneous. This was due not only to the purpose of the studies but also to their quality and form. Therefore, this review was formed in two sections, first, a summary of the population prevalence studies and, second, a thematic literature review.

Prevalence and clinical specifications

Besides being found in Uganda [9,23–25], IDE has been reported in Ethiopia [12,26–28], Kenya [11,29], Somalia [30], Sudan [13,31] and Tanzania [10,32,33]. A summary of population studies is seen in Table III, in which details regarding differences between settings, age and sex of the children and samples are presented. There are also reports indicating prevalence in northern Chad [34] and in the northeast of the Democratic Republic of the Congo (DR Congo) [35], as well as in Rwanda [20]. In addition, cases of previously performed IDE have been reported among East African immigrants in Australia [36], Denmark [18], France [34], New Zealand [37], Norway [19], Sweden [38] and the US [14,39,40] and also in populations in England [30] and Israel [41]. Due to recent geopolitical changes in Sudan and the division into two separate states, results from previous studies may apply differently. The population studies and case reports reviewed have either been conducted in or refer to individuals from the newly formed state of South Sudan.

Regarding age at which IDE is performed, Welbury et al. [26] reported a mean age of 6.5 months. Also, Iriso et al. [42] found modal age to be 5 months and Accorsi et al. [23] saw a median age of 6 months. The practice is contended to be common during the first year of life and incidents have been reported in children as young as 16 days, although tooth germ removals are unusual after the age of 24 months [23,26,27,42]. No differences have been reported between sex, either in Ethiopia [26] or in Tanzania [10]. Rodd and Davidson [30] reported a small but insignificant difference between Somali males and females.

The tooth most commonly subjected to the procedure (95–100% of cases) is the deciduous canine [10,31,32]. The mandibular canines account for the majority of the affected teeth in the primary dentition, although removal of deciduous maxillary canines also occurs [9,10,26,29,41]. Bimaxillary removals have also been reported. This was observed in 62% of the IDE-subjected children by Hiza and Kikwilu [10], and by Matee and van Palenstein Helderman [32], as they reported 79% to be bimaxillary affected compared to 16% in the mandible only. The removals appear to be mainly bilateral, as shown by case reports [14,34,38] and by findings of 100% in observed populations [10,32]. Still, unilateral cases are present [31,36]. Incidents of missing adjacent deciduous teeth have also been observed [10,32,41], speculated to be not procedural but rather iatrogenic, while intending canine enucleation [30,41].

The reason for the predilection of mandibular canines in particular is not fully understood. Suggestions have been made regarding the location and form of the follicles: the canines might be more palpable, and appear more prominent [42] and paler than in the maxilla in an infant with a dehydrating disease [23], which might make them easier to notice [10,41].

The purpose and process of IDE

The purpose of IDE is to prevent [43,44] and to treat various symptoms, manifestations and conditions [31,42]. Main instances are diarrhea, fever and vomiting [10,13,26,27,29,45]. In addition, the presence of more than one of the symptoms and reoccurrence and/or prolonging of a symptom or symptoms are factors suggested to elevate the risk of IDE

Table III. Summary of population studies conducted on the suggested prevalence of infant dental enucleation (IDE), by country.

Country	Study	Year of publication	Location and setting	Sample and age specifications	Prevalence specifications in populations
Ethiopia	Dagnew and Damena [12]	1990	Three rural communities	853 subjects, 0–5 years	70%
	Welbury et al. [26]	1993	Addis Ababa	2 groups, 2–18 years old: Primary (2–9.9 years*): 665 subjects Permanent (13–14 years*): 310 subjects	Primary group: 197 of 1330 canines affected, 15% Permanent group: 41 of 620 canines affected, 7%
	Asefa et al. [27]	1998	Jimma	1502 subjects, ≤ 1 year	38%
Kenya	Hassanali and Amwayi [11]	1988	Two rural schools	71 subjects, 5–7 years old	37%
	Hassanali et al. [29]	1995	Five rural health camps	2 groups, 6 months–7 years old: 6 months–2 years old: 95 subjects 3–7 years old: 111 subjects	6 months–2 years: 87% 3–7 years: 72%
Sudan	A/Wahab [13]	1987	Juba	80 subjects, infants	70%
	Baba and Kay [31]	1989	Juba	90 subjects, ≤ 16 months old	100%
Tanzania	Matee and van Palenstein Helderman [32]	1991	Eight rural communities scattered throughout the country	2 groups, 3–19 years old: Primary (3–5 years old): 1890 subjects Permanent (12–19 years old): 1377 subjects	Primary group: 10% (distribution 0–17% between the communities) Permanent group: 0.02% (IDE seen in only one community, 30/371 subjects = 8%)
	Hiza and Kikwilu [10]	1992	One rural community	262 subjects, ≤ 5 years old	37%
	Kikwilu and Hiza [33]	1997	Six rural communities scattered throughout the country	1052 subjects, ≤ 5 years old	31% (spread 0.5–60% among the communities)
Uganda	Pindborg [9]	1969	Four districts, rural dispensaries and urban and rural schools scattered throughout the country	1169 subjects, in seven age groups	IDE seen in only one district of 322 subjects, of which: 16%
	Accorsi et al. [23]	2003	Hospital serving rural and urban communities	2 groups, ≤ 4 years old: ≤ 1 year: 977 subjects 1–4 years: 1018 subjects	Total: 30%** ≤ 1 year: 25%** 1–4 years: 35%**
	Batwala et al. [25]	2007	Urban schools	437 subjects, 5–12 years old	2%
Israel	Holan and Mamber [41]	1994	Jewish Ethiopian immigrants	59 subjects, 3–12 years old	59%
England	Rodd and Davidson [30]	2000	Somali immigrants	260 subjects, 3–17 years old	32%

*The age specifications in the groups refer to clinical criteria used at certain ages in the study as indicative of either normal dentition or IDE-affected dentition. In the primary dentition, the following age intervals were used: the maxilla—2 to 9.9 years, the mandible—2–8.9 years. In the permanent dentition, 14 years of age was used for the maxilla, while the criterion for the mandible was set at 13 years of age.
**The data in the original study are presented partially together with another traditional practice, tea-tea. The percentage presented in this table is a recalculation of the data, representing the actual prevalence of IDE in patients with and without tea-tea.

[31,33,46]. The generality of this, however, has been challenged by findings indicating that IDE may also occur after one attack of ailment [13,33]. Other reasons described as inducing IDE are general ill health [13], persistent cough [27,47], oral infections [31], breast biting [46], gum itching [26], weight loss [26,33], unsatisfactory weight gain [46], refusal of suckling/eating and unexplainable crying [33].

The procedure is usually performed by local traditional practitioners [9,23,31,33,45], traditional birth attendants [43], elderly members of the community [29] or members of the family, such as the mother or grandmother [42,48,49]. Others noted to carry out IDE are school teachers [46], church employees [31] and even medical personnel such as assistants and doctors [13,45,50]. The initiative is taken by the parents [32,45] and might occur by recommendation of local practitioners, elderly family members or community acquaintances [31,33,35,45,48]. IDE can also be performed due to lack of treatment alternatives [13]. Even though the opinion of parents regarding IDE is thought to be crucial, procedures have occurred due to pressure from the social surrounding [13,46].

The procedure occurs under simple conditions using instruments such as knifes [23,31,45,47], razor blades [31,47], pliers [47], sharpened metal objects [13,26,31], nails [31], wires [47,48], botanic thorns [43] and finger nails [23,31]. The operative instruments are mainly not sterilized [47,48] and reuse of unclean instruments has been seen [23]. The tooth germs are commonly removed without the use of pain relief or sedation and no disinfection occurs [23,29,31,45,48]. However, cases have been reported of medication or injection with penicillin or other unknown substances during the removal process [26,46]. Among Maasai in Kenya, post-operative washing of the used equipment took place with a mixture of water, milk and soda ashes [43] and Mosha [45] reported cases of pre-heating of instruments. A look into equipment cleaning prior to IDE in Uganda showed that both reuse of instruments without cleaning and boiling or washing of instruments occurred. In 3% a new instrument was used [23].

Post-operative management has been observed to take place in some regions. Substances are rubbed or packed into the wounded area [33,51], such as unidentified herbs [29,47,48], ashes of herbs, lizard feces [31], honey [43], salt, sugar, sodium bicarbonate [29], charcoal powder [52] and local dressings [26]. The use of crushed medications such as antibiotics and sulfur capsules has also been reported [31], as well as the ordering of intake of herbs [46]. Nevertheless, some parents have recounted that, after the procedure, they were advised by the operator to take the child to a formal healthcare unit to be medicated [45,47].

Influential social factors

A number of factors have been considered to influence the existence and the prevalence of IDE. It is reported to be more common in rural communities [29,53] and among populations of low socioeconomic strata [31]. It is also suggested that the prevalence is lower in communities with higher literacy and level of education [53]. Support for the latter is seen in the study by A/Wahab [13], as he found 100% prevalence of IDE among children of illiterate mothers, compared to 76% of mothers with education of high school level or above. Similar results have been reported by Ahmed et al. [54]. These results, however, are contrasted with other reports finding no difference in the public belief, regardless of level of education [51,55]. In addition, it is suggested that social factors such as instability in politics, conflicts, lack of social security, isolation of rural communities, insufficient healthcare systems and decrease of health education contribute to the continuance of the procedure [42,51,53]. It is also argued that strong sociocultural cohesion and deep-rooted faith in traditional remedies, often in combination with disbelief in and failure of formal healthcare, might be an important preserver of IDE [26,31,43,45,46,56]. As reported in Tanzania, of 60% of the parents that had turned to hospitals for help prior to seeking traditional care, only 6% reported improvement [33]. Paradoxically, the majority of parents have perceived IDE as a successful treatment of their child's ailment [13,26,33].

Traditional health views may be maintained within immigrant communities and they might be important in the case of IDE [56]. Through information from Jewish Ethiopian parents, IDE was suggested to be practiced in the Ethiopian community in Israel [41]. Additionally, Rodd and Davidson [30] reported that 22% of the children with IDE features were born in the UK to immigrant parents.

Public perception and justification

Public knowledge and opinion about IDE in East Africa offer a variety of explanations for its rationale. Some think the canines are 'maggots' or not real—nylon/plastic/false—teeth, while others claim that all children are born with them [46,47,57]. Public association between diarrhea and 'false' teeth has also been reported in Kenya and in Uganda [51,58]. Other presumed initiators of morbid canines included witchcraft, difficulty breathing and even bus rides [51,55]. In some regions of Uganda there is a belief in contagiousness of the canines. There are reports of traditional practitioners prescribing herbs or smoking of eggshells to pregnant women, as preventive measures [46,47]. A dissimilar explanation has been offered by Maasai adults in Kenya. There, the motive was given in

Table IV. Reported dental features and in occurring cases prevalence data, suggested to be complications of infant dental enucleation (IDE).

Dental hard tissue complications	Dental eruption complications
Hypoplasia of the deciduous canine [9,26] or adjacent deciduous tooth 19 canines affected in 59 children [41] Found in 35% of the children [30]	Absence of permanent canine [26,32,41]
Hypoplasia of the succedaneous canine or adjacent permanent tooth [30,32,50]	Absence of deciduous [10,26] or permanent adjacent tooth [14,50] Of all IDE-affected teeth: 11% mandibular lateral incisors, 36% mandibular canines, 10% mandibular premolars and 14% maxillary canines [32] Mandibular permanent lateral incisor in 5% of children [30]
Malformation of deciduous or permanent canine [9,14,32,41]	Retention of deciduous canine [9] or adjacent tooth [24,30,41] Lateral mandibular incisor in 12% of children [30]
Malformation of permanent adjacent tooth [9,32]; crown disfigurements [36]; split-shaped [38], peg-shaped [34], shovel-shaped [9] or stump-shaped tooth [32]	Displacement and impaction of permanent canine [41]
Dilaceration of deciduous canine [41] or permanent adjacent tooth [36]	Ectopic eruption of deciduous or permanent canine [26]
Supernumerary deciduous [37] or unidentified tooth [50]	Eruption deviance of adjacent permanent tooth [14,30]
Development of odontoma [38,39] or gemination-like tooth [32,37]	Early eruption of permanent canine [9]
	Transposition of permanent canine with adjacent permanent tooth [9]

reference to the dentition of bovine calves regarding absence of canines and of diarrhea and fever, in the animal, that is applied to humans [29]. Overall, even though some parents perceive risks and complications with the practice, the opinions point towards blaming the bodily symptoms and the underlying disease for negative outcomes of IDE [47].

Risks and consequences of IDE

The procedure has been seen to pose risks and cause general, as well as dentoalveolar, complications. Regarding the general risks, awareness has been raised of the risk of blood-borne infections, in particular HIV [42,47,57,59]. Also, it is argued that IDE could lead to proper diagnostic and necessary treatment being ignored, postponed or given in a belated stage and thereby subjecting the child to risks [23,31,33]. As such, the rate of general complications caused by IDE might consequently be under-reported [23,42].

IDE has been seen to coincide with confirmed conditions and diseases. In northern Uganda, most hospital-admitted cases of IDE suffered from malaria, pneumonia or gastroenteritis [23]. A similar overview in southern Sudan revealed that 97% of the children were dehydrated and 11% had malaria [31]. Cases with malnutrition have also been seen [47]. Taking account of underlying diseases, it is believed that general complications are results of accumulated strain on the infant's health [53]. The most common ones observed have been septicemia and severe anemia [23,42], a pattern also seen by Accorsi et al. [23]. Also, there have been reports of lethal outcome due to IDE complications. In Uganda, fatality rates of 21% and 22% have been observed in hospitalized children [23,42]. Mortality cases have also been seen in Ethiopia [27], Sudan [13] and Tanzania [45]. Other reported complications are fever, other infections [29], tetanus [13] and meningitis [42]. In addition, infants subjected to IDE were observed to have significantly lower weight gain during a 4-month follow-up [27]. However, a different study found the impact of IDE on the infant weight to be inconclusive, referring to future focussed studies on the subject [60].

In the dentoalveolar area, complications have been seen that involve the dental hard tissues, the eruption path and order and the periodontal tissues. Reported features and, in occurring cases, prevalence data suggested to be IDE complications are presented in Table IV. Also, Figures 1,2,3 show examples of clinical manifestations of IDE. A contrasting addition to specifications on missing permanent teeth has been reported by Bataringaya et al. [24]. In their study, missing permanent maxillary canines among 14-year-old Ugandans were 3-times more common compared to permanent mandibular canines. The exceptional findings were speculated to be a probable reflection of the long-term effect of IDE. Other dentoalveolar complications are osteomyelitis [42], intra-oral scar tissue [14], inter-dental space closure [30], marginal bone defect [40] and loss of vitality with periapical infection in permanent canines [37,38] as well as in adjacent incisors [30,50]. There are indications of IDE having impact on the occlusion, as midline deviances toward the side of unilaterally enucleated teeth and reduced mandibular dimensions have been observed [41,61].

Public interventions and outcomes

The procedure is illegal in Sudan [13] and in various Ugandan areas [55]. Authoritative interventions such as political initiatives, police action and public educational programs have been implemented in Tanzania and in Uganda [33,55]. Further, the charity organization Dentaid has attempted to address the issue through activities and campaigns inside and outside Africa [21,62,63]. Comparable ongoing governmental projects are known in Kenya [22] and the

Figure 1. Clinical photo of a 6-year-old Somali girl missing both mandibular deciduous canines. According to her father, the "germs" were removed because of bodily sickness when the girl was 6 months old. Photo by Jir Barzangi.

procedure has been highlighted by the World Medical Association [64]. To our knowledge, no official follow-ups of the interventions have been published. Instead, in Tanzania, Kikwilu and Hiza have presented data and speculated about the long-term effects of previous interventions. Regions where IDE was first reported and undergone educational programs had very few cases of IDE, while areas where IDE was reported later had the highest prevalence [33]. In contrast, in northern Uganda it was revealed that, despite health education campaigns, the practice had not been abandoned. Instead, the infants were taken to health care facilities to prevent complications [48]. In a multi-step program aimed at exploring and altering public opinion in Uganda, attitudes and effects of interventions were studied. Educational workshops were staged and, in a follow-up, the group manifested change of knowledge and attitude: they exhibited adequate knowledge of IDE and about treatment alternatives for sick children. Further, hospital admissions and fatal cases due to post-IDE complications decreased, while infant complaint cases increased, indicating a rise in search of formal health care [55].

The role of traditional practitioners

The role of traditional practitioners in the local culture is important, as they are held in high regard by communities [33,43]. Even though some perform the procedure free of charge, many charge relatively high fees [13,33,48,55]. They share the same sociocultural background and values as the community and they are often the first point of healthcare contact [65]. Thus, due to the trust the public has in their skills, their role might be of crucial importance [33,43,47]. As such, a lack of programs designed with the intention of co-operating with and educating the traditional practitioners is considered a matter of concern [24,47]. The profundity of their position in the community is partially demonstrated by the Maasai in Kenya. Rejection of traditions concerning practitioners can lead to threats, expulsion from the community and even death [43]. Therefore, continuous change in public opinion without the engagement of traditional practitioners has been considered to be not realistic [24]. Results from the known attempts of this sort of intervention have been ambiguous. Many traditional practitioners are unwilling to change, sometimes in spite of health education [43]. Their practice usually is a source of social status and economic gain, which, together with fear of legal reprisals, are likely explanatory factors in the reluctance [43,55].

On the other hand, progress has also been indicated by Kikwilu and Hiza [33], as they reported that a transition had occurred in the traditional treatment.

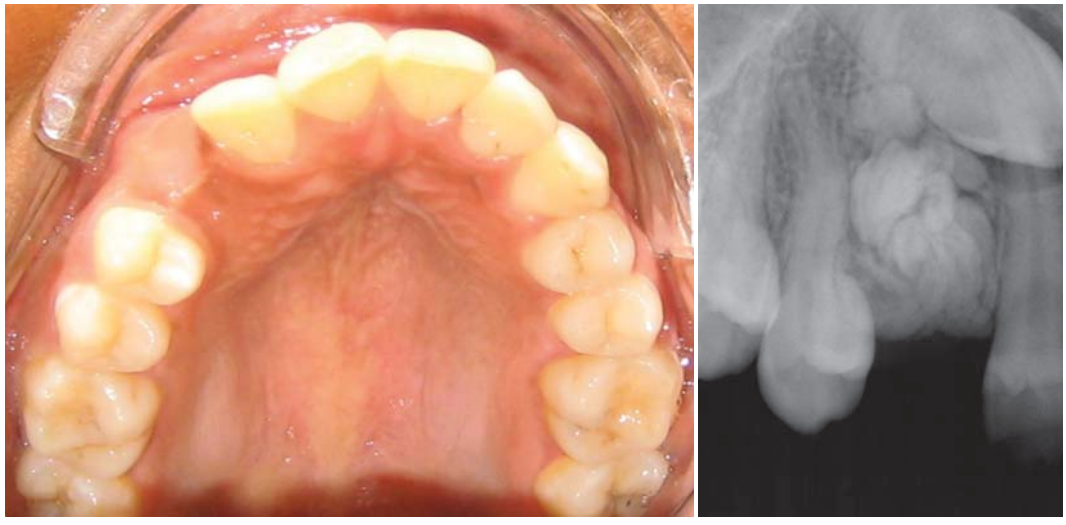

Figure 2. Clinical photo and an intra-oral x-ray of a 17-year-old Somali girl suspected to have been subjected to IDE at a young age. An odontoma was discovered to be obstructing the eruption of tooth no. 13. Photos and x-ray by Terttu Kinisjärvi.

Figure 3. Clinical photo and an intra-oral x-ray of an 8-year-old boy from Somalia. Tooth no. 82 has an enamel hypoplasia and it is persisting due to deviating eruption path of tooth no. 42. Also, tooth no. 43 has erupted early and it manifests hypoplasia. Photo and x-ray by Charlotta Jensen.

Instead of IDE, the practitioners favored the non-invasive method of rubbing of herbs. There is also a note about positive results from an education program in Uganda. However, statistics or details of the results have not been presented in the study, referring rather to a future publication [66].

Discussion and conclusions

In this review, we have presented information on a traditional remedial procedure that is justified through traditional motivations. It is performed on children at a young age and has harmful general health and dentoalveolar consequences. The summary of prevalence studies shows variation between and within the examined countries. Some socioeconomic and cultural factors that may influence the presence and practice of IDE have been exposed.

The literature search provided studies that varied in design and quality. Even though the inclusion criteria used for study selection were designed to be extensive, the number of studies with original data and/or information was limited. Further, the studies were mostly of observational and descriptive design. Although we have applied an integrative concept to the thematic part of this literature review, data evaluation has been intended to provide an extensive overview on IDE. Due to the heterogeneity in design and quality and to the limitations of studies, we have concluded that it was not possible to conduct a systematic review. As there is little known about the prevalence and complications of IDE outside East Africa, we estimated that it is a mainly unknown phenomenon amongst dental and health practitioners in Western countries. Therefore, we found it necessary to highlight available information through this literature review and we hereby discuss important aspects of our findings.

As IDE is believed to be ongoing in East Africa, there is a need of increased awareness about the practice. In reference to the presumed breadth of the phenomenon in its original setting, many areas appear to be unexplored. To our knowledge, no studies have been published on IDE occurring in Somalia, Chad or Rwanda. Simultaneously, it is suggested that native populations may have been influenced by migrants, regarding the benefit of IDE as a procedure [29,31,48]. We, therefore, estimate a risk of IDE being prevalent in regions and countries not discussed in the literature and of a currently ongoing process of spreading. These factors call for further studies of the extent of IDE. Nevertheless, we also see a need for updated sufficient data on IDE, particularly in Ethiopia, Tanzania and Sudan, as the data in the studies, with one exception, date before the year 2000. To our knowledge, no newer studies have been published.

Needless to say, there is a vital need for population studies amongst immigrants. As far as we know, only two such studies have been conducted [30,41]. During recent decades there have been migrations of East African refugees into Western countries. Considering Sweden, in particular, according to Swedish population statistics from 2011, over 85,000 residents in Sweden either were born or have both parents born in the countries Chad, DR Congo, Ethiopia, Kenya, Rwanda, Somalia, Sudan, Tanzania or Uganda [67]. Due to the size of the population of immigrants and the challenges IDE might constitute, it is important to seek knowledge regarding its prevalence. Nevertheless, in reference to the results by Holan and Mamber [41] and Rodd and Davidson [30], the risk of IDE being practiced on children born to immigrant parents cannot be excluded and this requires further examination. There is a call for diplomacy in the approach to the parents with regard to IDE, as

some might show hesitation to speak of the issue [30]. A diplomatic approach is also recommended to help form culturally sensitive health programs and to achieve successful results [30,56].

Additionally, we see a need for further exploration of the rationale of the procedure and the consequences of it. In the context of IDE, the tooth germs are regarded by many as infectious or foreign harmful bodies and associated with a risk of fatality [26,31,57]. Even though there are aspects of IDE that are commonly influential regardless of the nationality or geographical localization, the literature still points to diverging suggestions regarding the applicability of the rationale. As some have presented trends of change in the public, others emphasize the cultural/traditional connection as a major obstacle to change. To us, the literature indicates the nationality and/or ethnicity as important factors. Thus, we acknowledge the possibility of different rationales in different populations, and we stress a need for further qualitative, in particular, anthropological [15], studies researching these aspects. Such studies might clarify potential differences in the justification processes and thereby aid in forming strategies suited for local conditions in the attempts to counteract the procedure.

As to the consequences of IDE, to our knowledge, only one population study [41] has accompanied the clinical examination of the subjects with necessary x-rays. Knowledge is limited on the prevalence and extent of complications; in particular, the formation of odontomas or even yet undiscovered consequences. Further, considering the development of the dentition, additional risks arise that are poorly examined. Early loss of deciduous lower canines has been associated with retrusion of permanent incisors [68], anterior crowding in the permanent dentition [69] and inter-dental and arch perimeter reduction [70,71]. Moreover, more patients with early loss of deciduous canines require orthodontic treatment [69]. Additionally, there are potential psychological aspects of IDE in reference to the pain the child is exposed to. With regard to the complexity of factors influencing pain perception, studies have argued that exposure to pain or painful stress, during infancy might cause long-term tactile hypersensitivity and increased anxiety behavior [72,73]. These aspects demonstrate a need for systematic follow-up studies of patients subjected to IDE.

The absence of a conventional term for the practice is another matter. Considering the literature presented in this review, we find the previously used terms to be incomplete and/or unspecific with regard to its performance, the setting and how it is justified. A number of terms reflect local views, which may not apply in other areas. Also, the presence of other types of traditional dental and oral procedures existing in Africa, besides IDE, emphasizes the need to distinguish the practice. One dental example found in Sudan is *haifat*, which comprises puncturing dental follicles with hot needles for the same purpose as IDE [74]. As for oral procedures, known examples are traditional uvulectomy and tonsillectomy [28]. We also see difficulty in the use of transliterated names, as these might be subjected to spelling differences. This might already be present. As examples, due to similarities between the terms *Lake jo marak* [24] and *Lakijo marach* [46] or *Two lak* [24] and *Telak* [48], we hypothesize that they are the same. Thus, while we acknowledge IDE to be a severe form of mutilation, we see a need for a term to define and to describe the practice in its common basic form. Also, a general term might help in making information more accessible. Hence, we hereby propose the descriptive term *infant dental enucleation*, as used in this study.

In conclusion, the prevalence of IDE and the risks of it point to a need for more studies and increased awareness of the procedure. Also, the notion of its existence, with the risk of it still being practiced, among immigrants, emphasizes the need of specific guidelines of management within dental and health communities encountering East African immigrants.

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References

[1] Radbill SX. Teething as a medical problem. Changing viewpoints through the centuries. Clin Pediatr (Phila) 1965; 4:556–9.
[2] Rendle-Short J. The history of teething in infancy. Proc R Soc Med 1955;48:132–8.
[3] McIntyre GT, McIntyre GM. Teething troubles? Br Dent J 2002;192:251–5.
[4] Swann IL. Teething complications, a persisting misconception. Postgrad Med J 1979;55:24–5.
[5] Ashley MP. It's only teething. A report of the myths and modern approaches to teething. Br Dent J 2001;191:4–8.
[6] Dally A. The lancet and the gum-lancet: 400 years of teething babies. Lancet 1996;348:1710–11.
[7] Gibbons HL, Hebdon CK. Teething as a cause of death. A historical review. West J Med 1991;155:658–9.
[8] Markman L. Teething: facts and fiction. Pediatr Rev 2009;30: 56–64.
[9] Pindborg JJ. Dental mutilation and associated abnormalities in Uganda. Am J Phys Anthropol 1969;31:383–9.
[10] Hiza JF, Kikwilu EN. Missing primary teeth due to tooth bud extraction in a remote village in Tanzania. Int J Paediatr Dent 1992;2:31–4.

[11] Hassanali J, Amwayi P. Report on two aspects of the Maasai dentition. East Afr Med J 1988;65:798–803.
[12] Dagnew MB, Damena M. Traditional child health practices in communities in north-west Ethiopia. Trop Doct 1990;20: 40–1.
[13] A/Wahab M. Traditional practice as a cause of infant morbidity and mortality in Juba area (Sudan). Ann Trop Paediatr 1987;7:18–21.
[14] Edwards PC, Levering N, Wetzel E, Saini T. Extirpation of the primary canine tooth follicles: a form of infant oral mutilation. J Am Dent Assoc 2008;139:442–50.
[15] Johnston NL, Riordan PJ. Tooth follicle extirpation and uvulectomy. Aust Dent J 2005;50:267–72.
[16] Campaign aims to create awareness of infant oral mutilation. Br Dent J 2008;205:588.
[17] Longhurst R. Infant oral mutilation. Br Dent J 2010;209: 591–2.
[18] Lauritzen G, Poulsen O. [Forekomsten af tanddannelsesforstyrrelser efter tidlig 'germektomi' af temporære hjornetænder Disturbances in the creation of teeth after early 'germectomy' of temporary canine teeth] [Danish]. Tandlaegernes Tidsskr 1999;14:8–12.
[19] Espelid I, Agnalt R. Fjerning av tannanlegg i afrikansk medisin. Litteraturoversikt med kasuistikk Removal of tooth buds by traditional healers in Africa. Literature review and case report] [Norwegian]. Nor Tannlegeforen Tid 2009;119:294–7.
[20] Benzian H. World dental development fund Rwanda project visit report. Dev Dent 2003;3:21–3.
[21] Wordley J. Infant oral mutilation. Dev Dent 2003;3:19–20.
[22] Hassanali J, Wanzala P. Enucleation of deciduous canine tooth buds in infants in sub-saharan Africa. Dev Dent 2008;9:8–10.
[23] Accorsi S, Fabiani M, Ferrarese N, Iriso R, Lukwiya M, Declich S. The burden of traditional practices, ebino and tea-tea, on child health in northern Uganda. Soc Sci Med 2003;57:2183–91.
[24] Bataringaya A, Ferguson M, Lalloo R. The impact of ebinyo, a form of dental mutilation, on the malocclusion status in Uganda. Community Dent Health 2005;22:146–50.
[25] Batwala V, Mulogo EM, Arubaku W. Oral health status of school children in Mbarara. Uganda. Afr Health Sci 2007;7: 233–8.
[26] Welbury RR, Nunn JH, Gordon PH, Green-Abate C. "Killer" canine removal and its sequelae in Addis Ababa. Quintessence Int 1993;24:323–7.
[27] Asefa M, Hewison J, Drewett R. Traditional nutritional and surgical practices and their effects on the growth of infants in south-west Ethiopia. Paediatr Perinat Epidemiol 1998;12: 182–98.
[28] Jeppsson A, Tesfu M, Persson LÅ. Health care providers' perceptions on harmful traditional health practices in Ethiopia. Ethiop J Health Dev 2003;17:35–44.
[29] Hassanali J, Amwayi P, Muriithi A. Removal of deciduous canine tooth buds in Kenyan rural Maasai. East Afr Med J 1995;72:207–9.
[30] Rodd HD, Davidson LE. 'Ilko dacowo:' canine enucleation and dental sequelae in Somali children. Int J Paediatr Dent 2000;10:290–7.
[31] Baba SP, Kay EJ. The mythology of the killer deciduous canine tooth in southern Sudan. J Pedod 1989;14:48–9.
[32] Matee MI, van Palenstein Helderman WH. Extraction of 'nylon' teeth and associated abnormalities in Tanzanian children. Afr Dent J 1991;5:21–5.
[33] Kikwilu EN, Hiza JF. Tooth bud extraction and rubbing of herbs by traditional healers in Tanzania: prevalence, and sociological and environmental factors influencing the practices. Int J Paediatr Dent 1997;7:19–24.
[34] Khonsari RH, Corre P, Perrin JP, Piot B. Orthodontic consequences of ritual dental mutilations in northern Tchad. J Oral Maxillofac Surg 2009;67:902–5.
[35] Sabuni L. Dilemma with the local perception of causes of illnesses in central Africa: muted concept but prevalent in everyday life. Qual Health Res 2007;17:1280–91.
[36] Amailuk P, Grubor D. Erupted compound odontoma: case report of a 15-year-old Sudanese boy with a history of traditional dental mutilation. Br Dent J 2008;204:11–14.
[37] de Beavis FO, Foster AC, Fuge KN, Whyman RA. Infant oral mutilation: a New Zealand case series. N Z Dent J 2011;107: 57–9.
[38] Erlandsson AL, Backman B. A case of dental mutilation. ASDC J Dent Child 1999;66:278–9.
[39] Graham EA, Domoto PK, Lynch H, Egbert MA. Dental injuries due to African traditional therapies for diarrhea. West J Med 2000;173:135–7.
[40] da Fonseca MA, Hoge C. Extraction of "killer" teeth: the case of two Somali siblings. J Dent Child (Chic) 2007;74:143–6.
[41] Holan G, Mamber E. Extraction of primary canine tooth buds: prevalence and associated dental abnormalities in a group of Ethiopian Jewish children. Int J Paediatr Dent 1994;4:25–30.
[42] Iriso R, Accorsi S, Akena S, Amone J, Fabiani M, Ferrarese N, et al. 'Killer' canines: the morbidity and mortality of ebino in northern Uganda. Trop Med Int Health 2000;5: 706–10.
[43] Mutai J, Muniu E, Sawe J, Hassanali J, Kibet P, Wanzala P. Socio-cultural practices of deciduous canine tooth bud removal among Maasai children. Int Dent J 2010;60:94–8.
[44] Woodruff A, El Suni A, Kaku M, Adamson E, Maughan T, Bundru N. Infants in Juba, southern Sudan: the first six months of life. Lancet 1983;2:262–4.
[45] Mosha HJ. Dental mutilation and associated abnormalities in Tanzania. Odontostomatol Trop 1983;6:215–19.
[46] Mogensen HO. False teeth and real suffering: the social course of 'germectomy' in eastern Uganda. Cult Med Psychiatr 2000;24:331–51.
[47] Stephen S. False teeth still a public health problem among children in Kanungu district - South Western Uganda 2006. Educ Res Rev 2006;1:156–61.
[48] Stefanini A. Influence of health education on local beliefs: incomplete success or partial failure. Trop Doct 1987;17:132–4.
[49] Halestrap DJ. Indigenous dental practice in Uganda. Br Dent J 1971;131:463–6.
[50] Dewhurst S, Mason C. Traditional tooth bud gouging in a Ugandan family: a report involving three sisters. Int J Paediatr Dent 2001;11:292–7.
[51] Nuwaha F, Okware J, Hanningtone T, Charles M. False teeth "Ebiino" and Millet disease "Oburo" in Bushenyi district of Uganda. Afr Health Sci 2007;7:25–32.
[52] Abusinna I. Lugbara teeth germectomy of canines for the newborn babies. A magico-religious phenomena in some African tribes. Egypt Dent J 1979;25:209–14.
[53] Accorsi S, Ferrarese N, Stefanini A, Iriso R. Cultural beliefs and traditional health practices in the context of war, population displacement and social disruption: the examples of "ebino" and "tea-tea" in northern Uganda. In Liamputtong P, editor. Childrearing and infant care issues: a cross-cultural perspective. New York: Nova Science; 2007. p 63–77.
[54] Ahmed I, Eltom A, Karrar Z, Gibril A. Knowledge, attitudes and practices of mothers regarding diarrhoea among children in a Sudanese rural community. East Afr Med J 1994;71:716–19.
[55] Jamieson LM. Using qualitative methodology to elucidate themes for a traditional tooth gauging education tool for use in a remote Ugandan community. Health Educ Res 2006;21:477–87.
[56] Hodes R. Cross-cultural medicine and diverse health beliefs. Ethiopians abroad. West J Med 1997;166:29–36.
[57] Weiss B. Plastic teeth extraction: the iconography of Haya gastro-sexual afflicition. Am Ethnol 1992;19:538–52.

[58] Othero DM, Orago AS, Groenewegen T, Kaseje DO, Otengah PA. Home management of diarrhea among under-fives in a rural community in Kenya: household perceptions and practices. East Afr J Public Health 2008;5:142–6.
[59] Chindia M. Traditional dental practices. East Afr Med J 1995; 72:205–6.
[60] Gray S, Akol HA, Sundal M. Longitudinal weight gain of immunized infants and toddlers in Moroto District, Uganda (Karamoja subegion). Am J Hum Biol 2010;22:111–23.
[61] Hassanali J, Odhiambo JW. Analysis of dental casts of 6-8- and 12-year-old Kenyan children. Eur J Orthod 2000;22: 135–42.
[62] Longhurst R. IOM awareness. Br Dent J 2010;208:198.
[63] Gollings J. Cultural awareness. Br Dent J 2011;210:501–2.
[64] Odigwe C. World medical association 'appalled' at oral mutilation in parts of Africa. BMJ 2004;328:976.
[65] Kubukeli P. Traditional healing practice using medicinal herbs. Lancet 1999;354:SIV24.
[66] Kirunda W. 'Ebino' (false teeth): how the problem was tackled in Tororo. Trop Doct 1999;29:190.
[67] Population statistics 2011 [Internet]. Stockholm, Sweden: Statistics Sweden; 2012. cited 2012 Sep 12 p Available online at http://www.scb.se/Pages/ProductTables____25795.aspx, acessed 12 september 2012.
[68] Sayin MO, Turkkahraman H. Effects of lower primary canine extraction on the mandibular dentition. Angle Orthod 2006; 76:31–5.
[69] Miyamoto W, Chung CS, Yee PK. Effect of premature loss of deciduous canines and molars on malocclusion of the permanent dentition. J Dent Res 1976;55:584–90.
[70] Kau CH, Durning P, Richmond S, Miotti FA, Harzer W. Extractions as a form of interception in the developing dentition: a randomized controlled trial. J Orthod 2004;31: 107–14.
[71] Lindsten R, Ogaard B, Larsson E. Anterior space relations and lower incisor alignment in 9-year-old children born in the 1960s and 1980s. Angle Orthod 2001;71:36–43.
[72] Peters JW, Schouw R, Anand KJ, van Dijk M, Duivenvoorden HJ, Tibboel D. Does neonatal surgery lead to increased pain sensitivity in later childhood? Pain 2005;114: 444–54.
[73] Grunau RE, Tu MT. Long-term consequences of pain in human neonates. In Anand KJ, Stevens B, Mcgrath P, editors. Pain in neonates and infants. 3rd ed: London, Great Britain: Elsevier; 2007. p 45–55.
[74] Rasmussen P, Elhassan E, Raadal M. Enamel defects in primary canines related to traditional treatment of teething problems in Sudan. Int J Paediatr Dent 1992;2:151–5.