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Citation:

Rees, J and Muskett, TA and Enderby, P and Stackhouse, J (2016) Implications of Untreated Cleft Palate in the Developing World: Adaptation of an Outcome Measure. *Folia Phoniatrica et Logopaedica*. ISSN 1021-7762 DOI: <https://doi.org/10.1159/000440836>

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Document Version:

Article (Accepted Version)

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## **Implications of Untreated Cleft Palate in the Developing World: Adaptation of an Outcome Measure**

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Running title: Outcome Measure for use with untreated CP

## **Abstract**

*Objective:* To identify the implications of living with untreated cleft palate (CP) and develop an outcome measure which reflects the broad impact of surgery [1] but is sensitive to the physical impact, speech changes and psycho-social implications of late intervention.

*Design, participants, setting:* Focus groups and individual interviews were used to gather information on the implications of living with untreated CP and the impact of subsequent surgery. Participants included 11 individuals attending a cleft department in India whose cleft had persisted into adolescence or adulthood, and 16 of their family members. Findings were used to assess whether The Therapy Outcome Measure (TOM) [1] could capture the implications of untreated CP and the impact of surgery beyond that of speech alone.

*Results:* Findings indicated that implications of living with untreated CP revolved around difficulties with communication, reduced autonomy and nasal regurgitation. These issues are encapsulated within the third and fourth domains of the TOM but there is a need for an adapted version (TOM-clp) to use in the developing world incorporating areas specific to cleft palate.

*Conclusion:* The TOM has potential as a global tool for measuring the broad impact including the psychosocial benefit for attending to untreated CP.

Oral health is acknowledged by the World Health Organisation (WHO) as a fundamental human right [2] yet over a million individuals with cleft lip and palate (CLP) in the developing world remain untreated into adulthood [3]. Typically, this is attributed to an overall lack of resources and inadequate awareness of treatment opportunities [4][5][6]. However, another consideration is that there is a lack of appreciation regarding the broad impact of untreated clefts which may be restricting funding for older patients.

The accepted global gold-standard for managing CLP demands early intervention by a multidisciplinary team in fully equipped specialist centres of excellence. This policy is based on considerable data showing that timely closure of the lip and palate at a young age is associated with the best aesthetic and speech outcomes [7]. Thus, service provision based on this policy will naturally direct resources towards treatment of younger rather than older patients.

Little has been published regarding the impact of untreated adult CP in the developing world, and even less relating to the outcome of intervention for this cohort. Whilst Murthy et al 2010 [6] demonstrated improved intelligibility following late palate repair and a number of papers allude anecdotally to improved articulation and nasality [8][9], the majority of published research indicates limited benefit, particularly in relation to speech [10][7][11][12][13]. Since Global healthcare strategies rely on available evidence to ensure that funds are appropriately directed, the published data ostensibly substantiate the decision to direct funding towards young patients rather than an older cohort. Consequently, cleft charities such as *Smile Train* and *Operation Smile* prioritise treatment for children under six years old leaving little resource for management of older patients [14]. However, there are significant limitations in much of the research in this field which decrease reliance on their findings.

A common feature of many studies in this field is that they rely on detailed phonetic analysis of cleft speech characteristics including, articulation, hypernasality, nasal emission and velopharyngeal function, to ascertain the parameters of change [15][16][17]. This generates two fundamental concerns when considering untreated CP in the developing world. First, detailed speech analysis requires the presence of highly specialised SLTs who are preferably fluent speakers of the relevant language. In practical terms, this is rarely available which makes the replication of such studies impractical. Second, by focussing almost entirely on the impairment and detailed speech characteristics, there is no accounting for the psychosocial impact or the effect on personal well-being that the intervention may afford even where cleft speech characteristics still persist. These may include improved ability to communicate and participate in social events, and take advantage of educational or occupational opportunities [18].

The narrow focus and arguable limitations of previous research indicate that more work is required to develop robust evidence that will enable an informed decision to be made regarding investment in services for adults with untreated CP. However this necessitates the selection and implementation of an outcome measure which is appropriately designed to capture such data.

The most important factors when choosing an outcome measure are that it should make an accurate representation of the clinical scenario, and be practical for the clinical setting [19][20][21]. A plethora of outcome measures have been devised for use with craniofacial anomalies, assessing such features as aesthetics and function [22][23], detailed speech characteristics [24][25][17], and parity of service quality [26][27]. However, whilst these approaches have advanced cleft service provision significantly, they pay little attention to the psycho-social and quality of life (QoL) issues relating to CLP, which can have disproportionate psychosocial affects through marginalisation and exclusion [28][29][30][31][32][33][34][35]. Furthermore, any measure must be internationally acceptable, culturally sensitive, cross language barriers, and be simple to administer at minimal cost without the need for highly trained specialist services.

There is not currently an outcome measure specifically designed for adults with CP, treated or otherwise. However the TOM [1], may meet these criteria. The TOM comprises four domains; Impairment, Activity, Participation and Well-being. Each aspect is assessed independently but work together to provide a holistic reflection of an individual's circumstances, from both a clinical and a functional perspective, whilst also accounting for social and personal concerns. The TOM's basic construct lies in the *WHO-International Classification of Impairments, Disabilities and Handicaps* (WHO-ICIDH)[36], a framework developed and updated to the *WHO International classification of Functional Disability and Health* (WHO-ICF) [37] to reflect how health and disability have an impact on peoples' lives [38]. This foundation, the fact that it is simple to use, is not language specific and does not require specialist equipment or resources to implement, makes it particularly appropriate for an international healthcare context.

This study examines the appropriateness of the TOM for adults with late or unrepaired cleft palate and targets the following as yet unaddressed questions:

1. What are the psycho-social and QoL implications of living with CP into adulthood?
2. What is the psycho-social and QoL impact of receiving intervention for untreated CP?
3. How can the TOM be adapted for collecting data which reflects QoL issues affecting cleft palate patients in the developing world?

## **Method**

### ***Design***

In order to address the first two research questions, guided semi-structured interviews and focus groups were conducted with individuals who had experienced untreated CP beyond 13 years of age. This process was undertaken over a period of two weeks in a busy cleft department in Karnataka State, India. Focus groups were used as an effective and efficient means of collecting a substantial amount of qualitative data within a short period of time [39]. Additionally, individual interviews provided a route for the inclusion of those who were uncomfortable in group situations or could not attend focus group sessions.

This research was designed to adapt an outcome measure specifically tailored to collect information regarding the personal perceptions of individuals' experience of late or non-repair of cleft palate in adults. Tightly matching a control group is challenging for this sort of study and resource limitations were also an issue for us. Moreover, in the absence of an exploratory initial awareness of key individual and social issues for this population, it was difficult to ascertain which groups might be the most appropriate to use as a comparison, particularly given the cross-cultural context.

### ***Participants***

Primary participants were identified from two sources: i) Purposeful consecutive convenience sampling was used to identify all individuals with untreated CP over 13 years of age, attending the department during the 2 week data collection period and ii) departmental attendance records to include patients who had presented between 2006 and 2012 with untreated CP over 13 years of age, and subsequently treated. These individuals were telephoned and invited to participate whilst attending a routine review appointment. Each primary participant was encouraged to include family members and friends to participate in the interview sessions.

Patient demographics are summarised in table 1. Seven primary participants were recruited opportunistically at clinic appointments and four individuals were approached directly, giving a total of eleven participants. Age from 13 to 25 (mean age 18.9). All of the participants had received at least some level of education but only three were still attending school.

#### ***Table 1: Summary of participant demographics***

Seven of the participants had undergone lip repair in childhood but not received timely treatment for their cleft palate and four had isolated CP which was not treated in childhood. Of the eleven participants four were seeking treatment for a failed repair following treatment in a different unit and three had recently undergone successful late repair at the unit.

Family members and friends were recruited through the primary participants as detailed above. Sixteen friends and family members attended the focus groups and interviews, each of the primary participants having at least one individual accompanying them during the session.

### ***Consent***

Although written consent forms and information sheets had been produced in English and Hindi, a decision was made to undertake verbal consent through the interpreter in the preferred local language, Kannada or Marathi. This negated the requirement for good literacy and ensured that participants had a good understanding of the project and ensure fully informed consent. Where participants were under sixteen years of age consent was obtained from the participant as well as the parent / legal guardian.

Participants were assured that their decision over whether to participate would not affect their on-going treatment in the department and that they could withdraw at any time without consequence. Since this research was undertaken during routine care appointments participants did not incur additional costs and thus compensation was not offered for participation.

### ***Development of an interview guide***

A guide for use in the focus groups and interviews was developed from a thematic literature review aimed at identifying the key concepts and concerns relating to untreated CP conducted specifically for this study and documented in previous research [40]. A number of search engines including MEDLINE, CINAHL, OVID were employed in order to obtain as comprehensive a picture as possible, making use of search strategies including key word and snowballing.

The resulting 22 papers were read and notes made on the themes presented in each. By iteratively comparing, aggregating, and refining information across the studies, four key themes were identified at saturation: (i) problems with communication; (ii) impact on integration and inclusion in the community; (iii) interference with activities of daily living; (iv) restriction on educational and employment opportunities. These themes were used as the foundation for developing an interview guide for use in the individual interviews and focus groups [40].

### **Data collection procedure**

One focus group (consisting of 4 individuals with CP and 5 family members) and seven individual interviews with a single CP participant each with at least one accompanying friend or family (total 7 participants with CP and 11 family members) were employed. A quiet side office was chosen as a location in order to minimise disturbance and to provide a relaxing environment. Sessions were chaired by a multilingual, non-clinical member of the cleft team who also acted as an interpreter and moderator. This individual was the cleft centre appointment coordinator and had long term experience of working with individuals with severe clefts. Consequently he was comfortable and skilled at understanding these individuals who may otherwise have been difficult to understand. A facility for writing down answers was available in the event that there were any problems with intelligibility. The same procedure was followed for the focus group and the individual interview sessions with the first author in attendance throughout.

Participants sat in a small circle and were encouraged to talk freely about their personal experience of CP. Participants who had already received surgery were asked to consider the impact of their cleft on their lives prior to intervention and comment on changes following surgery. The moderator facilitated the conversation, using the predetermined interview guide, to ensure that discussions covered the themes arising from the literature review. The moderator translated and summarised the participants comments at intervals to allow the researcher to monitor how the session was proceeding and to interject where clarification was required. These comments were recorded for later transcription and analysis but no attempt was made to analyse speech patterns for purposes of this study.

As each session proceeded the researcher began to generate provisional key themes based on real time analysis of the interview data [41]. At the close of conversation these initial themes were immediately reviewed with the participants to verify whether this initial summary was a true reflection of the discussion and to make additional comments. This process often prompted the delivery of additional information which would have been missed in a formal interview or questionnaire. This cycle of events was continued to saturation point where there was confidence that no new themes would emerge [39]. Each session lasted no longer than 30 minutes.

This interview and summary technique was chosen to allow a degree of validation of the information presented without requiring the participants to return for a second session. This was considered important in order to minimise disruption and expense for those involved. A debrief was included at the end of each session in order to address any sensitive issues which had arisen during the discussion. Participants were thanked for their participation and ensured that members of the cleft team would keep them updated with the outcome of the research at future appointments.

### **Modifications to the original methodology made during data collection**

Initially it was planned to record and transcribe the sessions with the express consent of the participants involved. However, since several different languages were involved it was felt that translation and transcription of the entire process would have proved overly complicated so only a translated summary of the responses was recorded as the session progressed.

### **Thematic analysis of focus groups and individual interviews**

The data generated through these procedures were thematically analysed. This phenomenological analysis approach was chosen as a means of generating qualitative data which would reflect real life experiences of living with untreated CP [42]. The recorded translations from the mediator were transcribed verbatim into a word document by the researcher. Each transcript was read through several times in order to familiarize the researcher with the interviews. The transcripts were analysed using an inductive approach [43] to identify comments which captured important features of the ways in which individuals were affected by their CP. Care was taken to identify not only the obvious themes which were predominant throughout the data set, but also less prevalent expressions which nonetheless detailed an issue of relevance to the research questions.

Themes relating to the psychosocial impact of CP and consequent effects of treatment were identified and grouped into initial themed categories. Participant contributions were then re-assessed and tabulated into higher order core categories under headings relating to each of the research questions according to their semantic content. In view of the small size of the data set this process was undertaken manually rather than using electronic software. In order that comments could easily be related back to specific individuals each participant was allocated a unique identification number and colour code which was used throughout the analysis, enabling cross referencing and linkage to the original raw data.



Comments were re-reviewed, clustered and scrutinized to ensure that the resulting themes were truly representative of the raw data. Themes were then cross referenced to evaluate modes of data collection and against the provisional real-time analysis and compared to assess the level of corroboration.

### ***Ethical considerations***

All study protocols and materials were reviewed and approved by the Department of Human Communication Sciences Research Ethics Committee within the University of Sheffield. Local governance procedures were followed throughout this research.

## **Results**

### ***Thematic analysis of focus groups and individual interviews***

Initial analysis highlighted ten general consequences of untreated CP suggested by the participants and/or a family member or friend (Fig 1)

*Fig 1: Themed consequences proposed by primary participants and family and friends.*

The ten consequences identified were categorised into three overarching core themes into which these issues could be allocated:

- Impact on communication
- Effect on integration and general activities
- Limitation on aspirations, access to education, employment opportunities and marriage prospects.

Each theme will now be explicated, with relevant illustrative comments from the raw data included for reference. The comments are quoted verbatim as translated by the interpreter hence appear in the third person. (P) representing comments from primary participants and (F) those of a friend or family member.

### **Impact on communication:**

Poor communication was reported to be the overriding problem for ten (92%) of the participants. This was true not only when mixing with strangers but also with family members:

“She won’t mix with the strangers because speech is not good. And her mother is also facing the problems some difficulties words when she speaks that she can’t understand” (F)

This problem was reported equally between primary participants and family and friends and drew by far the greatest number of comments overall.

Inability to communicate fluently was also the reason given for seven of the eleven (63%) complaining of teasing and bullying which made them unwilling to talk to strangers:

“He won’t speak with the strangers.....because he feels that they won’t understand his language. Maybe they laugh at him so he won’t mingle or go with the strangers” (F)

Whereas an unwillingness to talk to strangers was seen by family and friends as noteworthy problem (9 out of 12 comments) bullying was considered more of a problem by the primary participants (9 out of 11 comments)

"Good friends at school but also suffered teasing" (P)

"Others used to tease him so he didn’t go to school" (P)

Similar comments indicated that poor communication made them poorly accepted and reluctant to attend school on a regular basis:

“Regarding in the school when he speaks to his friends or classmates they won’t understand him and he took fights with them”(F)

This issue was seen as an equal problem for all participants.

### **Effect on integration and general activities**

All participants expressed equal concern that they would not be understood and half of them required assistance shopping:

“Usually she used to take the help from her husband in the market or somewhere else if she wants to tell anything buy in the market she cannot convey that message to that person” (P).

Seven participants (63%) indicated they could not be understood on the telephone which reduced their independence but was more prevalent amongst primary participants than family and friends:

“On the phone he tries to speak if the person understands his language he continues it otherwise he gives to his friend” (P)

Inhibition whilst socializing was seen as a problem equally between all participants, with eight of the participants (73%) experiencing difficulties due to poor speech and nasal regurgitation. One participant's family made her keep completely silent in public owing to the embarrassment her condition:

"Hiding always hiding even with the functions with the functions she's hiding" (F)

"In public actually when she is attending any functions and all she only complains that when she drinks water, water it comes from the nose that's her only complaint is that. If it's with her family she'll do it" (P).

In general, nasal regurgitation was considered a greater impediment by primary participants than their family and friends.

### **Limitation on aspirations, access to education, employment opportunities and marriage prospects:**

Limitation on aspirations, access to education, employment opportunities and marriage prospects were considered a difficulty by all participants equally.

Three participants (27%) reported that they left their education as a result of difficulty with communication and teasing:

"They can't understand so the previous school they told his parents you just take him out because we are not understanding the language" (P).

Four participants (36%) specifically referred to poor job prospects as a result of their CP:

"He told that he went to the army selection; he got selected for all this other things other than the medical examination, was turned down because of speech" (P).

Two participants specifically suggested that their marriage prospects were limited because of their cleft, although it was not ascertained whether this related to communication difficulties, nasal regurgitation or the stigma of having a cleft:

"His mother is telling, if he has cleft palate and all affects whether he gets married or no, whether he get girl or no" (F).

Consideration should be made that three of the participants were under sixteen and still at school. None of these individuals commented on the potential limitations on employment opportunities or marriage which might indicate that they had not yet considered these as an issue of concern at this stage.

### ***Psycho-social impact of receiving intervention***

All of the participants who had already received treatment for their CP claimed that surgery had markedly improved their ability to communicate, eliminated their nasal regurgitation, made them

more willing to participate in daily activities and improved their well-being through reduced anxiety, general inclusion and acceptance within their society:

“Before operation not really takes any liquid, comes down his nose but now is not after the surgery it’s not” (P).

“He’s telling when he meets the strangers he is not getting any problems to me he was telling he is very comfortable with the strangers to speaking only” (post-surgery) (P).

### **Reflection of the three core themes in relation to the domains of the “Therapy Outcome Measure**

Once the three core themes had been identified, each was assessed and considered regarding to its bearing to each of the TOM domains. The TOM is divided into four fundamental domains: Impairment, Activity, Participation and Well-being the first 3 of these reflecting the WHO-ICF and the fourth being an additional field.

The first domain of the TOM is intended to describe the severity of the impairment, the second to reflect functional capacity whilst the third and fourth domains relate to the psychosocial and QoL aspects of the condition. A summary of the TOM is presented here but a full account of its structure and function can be found in *Therapy Outcome Measures for Rehabilitation Professionals* [1].

**Impact on communication:** The inability to communicate was considered relevant to three of the four domains. The difficulty an individual has in producing words that can be understood is considered in domain 2 as the activity of communication and may be different according to whether an individual is in a familiar or unfamiliar environment. Poor communication will also affect an individual's level of participation in relation to their willingness to take part in social activities which in turn may affect their level of autonomy and integration thus considered in domain 3. Poor communication can also lead to anxiety and frustration thus affecting well-being and emotional status which is addressed by the fourth domain. However, it is important to consider that the domains are not linearly related i.e. a mild impairment may not necessarily lead to a mild activity restriction or have limited impact on social participation or wellbeing.

**Effect on integration and general activities:** Participants complained that their struggle to make themselves understood meant that they required assistance using the telephone and shopping independently which were considered under activities, as was the degree of nasal regurgitation when eating and drinking. In turn these factors affected individuals’ willingness to socialise and may have an impact on participation and wellbeing.

**Limitation on aspirations, access to education, employment opportunities and marriage prospects:** All participants mentioned their frustration and anxiety at not being able to reach their full potential as a result of their untreated CP. Thus the limitation in participation is reflected by the third domain which in turn may affect their well-being which is reflected in the fourth domain. It is

accepted that, since there was not a comparison group it is not possible to be certain whether these limitations are greater than for the general population or for adults with early repaired cleft.

The results were consolidated into a table representing the second, third and fourth domains of the TOM. This research addressed the psychosocial aspects of untreated CP so the results do not reflect the first domain which involves only details of the impairment it is however included here for completeness. Since the aim of the TOM-clp is that it should be appropriate for use by non-professionals, explicit examples were included, in order to explain the nature of each domain for simplicity of use in the field (see Table 2).

Table 2: Domain inclusions to adapt the generic TOM to the TOM-clp

## **Discussion**

This project allows an important insight into perceived psychosocial impact that untreated CP has on the lives of the participants and their families. The focus group sessions and informal interviews proved a valuable process for generating information and the importance of including family members and friends was borne out by the different perception they had of the consequences of CP in comparison with the individual who had the cleft. It was notable that whilst the individuals with the cleft were aware of the more personal psychosocial aspect such as bullying, nasal regurgitation and lack of autonomy using the telephone, family and friends were aware of the way in which CP prevented aspects of socialisation such as communicating with strangers.

In agreement with previous work [44] the issue of poor communication was the overriding complaint which pervaded the discussions. The frustration of not being understood had an impact on almost every aspect of daily living including education, shopping, socialising and using the telephone the consequence of which often resulted in failure to reach educational potential, marginalisation, under-employment and poor marriage prospects. This is interesting not only from a psycho-social and well-being perspective but also from the financial implications, since a lack of independence may lead an individual to become a life-long dependant on other family members rather than reaching personal autonomy and becoming a potential provider [45][46].

Whilst the impact of poor communication corresponded with the findings of other researchers [47], the reported impact of surgical intervention did not. A significant proportion of the literature suggests that surgical intervention without the additional input from SLT services does not improve speech quality [12], however all of the post-operative participants in this study indicated a significant improvement in their ability to communicate despite confirming that they had not received SLT services. Whether this represents a real improvement in their speech or enhanced confidence and willingness to speak needs to be established. Thus, although this is a small sample, it does substantiate the importance of developing an outcome measure to evaluate an individual's improvement in communication skills rather than an academic perspective using speech analysis.

Nasal regurgitation is barely mentioned in the literature as an issue for individuals with untreated CP however it was mentioned as a point of embarrassment by seven (64%) of the participants in this study. All the participants who had received surgery claimed that this problem had been eradicated post-surgery thus eliminating the associated social issue and improving QoL. These findings indicate that nasal regurgitation should be given more attention than is currently the case when considering surgery for adult patients. It is therefore considered an important issue to include in the adapted TOM.

The results from these discussion sessions indicated that untreated CP has a considerable negative impact on the lives of these individuals at a fundamental level. Although only four of the participants were post-surgery, their anecdotal remarks indicate that surgery had markedly improved their well-being. If this is indeed representative of the psychosocial outcome of intervention for this cohort then the need for further qualitative data and an appropriate outcome measure to collect is paramount.

The aim of this research was to identify the psycho-social implications of living with an untreated cleft palate and assess whether a scale within the TOM [1] can be adapted for use as an appropriate tool to gather data which reflects changes following late intervention for this cohort. The focus groups and interviews established which factors have an impact on the lives of individuals with untreated CP and to what extent surgical intervention played a part in addressing these problems. It became apparent that the concerns and implications of living with untreated CP focused around psycho-social problems such as difficulties with communication and reduced autonomy in relation to education and activities of daily living, as well as the embarrassment of nasal regurgitation. None of these factors have been adequately considered in previous research and yet they are of paramount importance in the everyday lives of those affected. This confirms the need for an appropriate outcome measure which will reflect the psychosocial impact of untreated CP as well as the physical impairment.

The core concerns and sentiments alluded to by the participants revolved around aspects of limitation in activity, negative impact on socialisation, participation and well-being. These aspects appear to fit succinctly into the second, third and fourth domains of the TOM, indicating that adapted scale would make an appropriate tool to reflect these broader impacts of CP in the developing world.

Although the study involved only eleven participants with experience of CP and sixteen family and friends, it embraced individuals from a range of socioeconomic backgrounds. While the size of the data set is limited, the results reflect the key concerns highlighted in other studies in India [48] and South America [44]. It is therefore possible that from a cultural viewpoint the TOM-clp should be transferable to other continents and cultures and would be broadly applicable to other developing nations on a more global scale.

## **Conclusion**

Untreated CP is prevalent throughout much of the developing world yet little is known about how it affects individuals at a personal and functional level. This study has not only contributed to our understanding of the impact of untreated CP in adult life but also shows how an existing outcome measure can be used and adapted to reflect the impact of the impairment on daily life. This study has focused on the QoL and psychosocial impact of untreated CP in relation to functional capacity, however the TOM-clp can also be used to reflect the structural severity and functional consequence of the impairment as well as an aesthetic component in relation to cleft lip.

In order to establish the reliability and validity of the TOM-clp it needs to be trialled on a larger number of participants across all four domains. It is the aim of the TOM-clp that it is simple to use and can be implemented reliably by non-specialist staff so future pilot studies will therefore be required to examine inter-rater and intra-rater agreement across different cultural divides. This research is on-going but initial results suggest a positive outlook for the development of an appropriate outcome measure for this sparsely researched field.

## **Acknowledgement**

The authors gratefully acknowledge the contribution of the plastic surgery unit, KLE University, Belgaum, Karnataka, India.

## **Disclosure statement**

The authors declare that they have no conflict of interest

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