

The impact of hypodontia: a qualitative study on the experiences of patients

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SUMMARY Congenital absence of teeth affects 2–6 per cent of the population, but its impact on quality of life (QoL) is not fully understood. The symptoms of hypodontia can vary and therefore also the treatment and management of this condition. Determining and understanding the possible impact of hypodontia on patients could inform and improve the management of such patients.

A purposive sample of 10 participants aged 16–25 years (five males and five females) with mild, moderate, or severe hypodontia at various stages of treatment was recruited. The participants had previously completed a 49-item Oral Health Impact Profile (OHIP) questionnaire with summary scores ranging from 24 to 143. All subjects participated in semi-structured interviews, and these were transcribed and analysed using NVivo software.

The results show that the role of hypodontia patients in the treatment decision-making process changes significantly as they move from childhood to adulthood. Participants indicated dissatisfaction with the lack of communication with dental services throughout early adolescence as they became more cognizant of their condition which in turn led to concerns with regard to appearance which impacted on their psychosocial well-being.

Hypodontia patients expect improved communication with dental practitioners and services as they become more cognizant of their condition and wish to become more actively involved in the decision-making process regarding their current and future treatment.

Introduction

Hypodontia is a term used to describe the developmental absence of one or more teeth (Whittington and Durward, 1996), oligodontia where six or more teeth are missing, and anodontia the developmental absence all teeth (Silva Meza, 2003). The prevalence of hypodontia in the permanent dentition ranges from 2.3 to 11.3 per cent (Larmour *et al.*, 2005). The third molars and mandibular second premolars are the most frequently missing teeth and represent 40–50 per cent of the total number of developing missing teeth (Rose, 1966). The maxillary lateral incisors are the next in terms of frequency (25 per cent), followed by maxillary second premolars (20 per cent), and the mandibular central incisors (6.5 per cent; Jepson *et al.*, 2003).

The aetiology of hypodontia is unknown; however, it may occur due to genetic and environmental factors or without an hereditary history. Hypodontia may occur either in isolation or in association with syndromes, such as ectodermal dysplasia, Down syndrome, and conditions such as a cleft lip and palate. The aim of restorative treatment in the management of patients with congenitally missing teeth was to achieve and maintain a functional, healthy, and aesthetically acceptable dentition throughout the life of the individual. Several treatment options exist according to the degree of severity. This can vary from no treatment (Jepson *et al.*,

2003), orthodontically closing a space (Robertsson and Mohlin, 2000), orthodontically opening a space, or maintaining or redistributing tooth space in preparation for prosthetic replacement (Carter *et al.*, 2003).

There have been a number of studies reporting prevalence, probable aetiology, and various dental treatment modalities for this condition. In a study of 451 patients (Hobkirk *et al.*, 1994), it was found that dissatisfaction with appearance was a major concern for patients with congenital absence of teeth. They also commented on the potential impact of delayed referral for management on social and educational development of these patients. However, to date, there has been little investigation into the impact of hypodontia on the quality of life (QoL) of patients, particularly the social and emotional impact of the condition. Locker *et al.* (2005), in a study of oral health-related QoL evaluated by the Child Perception Questionnaire (CPQ) in children aged 11–14 years, found few differences between patients with orofacial conditions to those with normal dental caries. However, they did state that oligodontia has a greater impact on oral health QoL. Wong *et al.* (2006) in a study using the CPQ in Chinese children with severe hypodontia concluded that the impact of missing teeth on QoL was also high. Laing *et al.* (2010), who evaluated patients aged 11–16 years found no difference in the psychosocial status of subjects with

hypodontia but only in relation to some functional aspects compared with other patients with features of malocclusion. There are no studies to date that have explored QoL and the potential impact of hypodontia on adolescent or young adults. Furthermore, the subjective concerns of patients with hypodontia have not been evaluated using qualitative methodology.

The Republic of Ireland has a mandatory dental screening programme whereby children are screened at primary school between the ages of 7 and 10 years. Patients with hypodontia are generally diagnosed through this screening programme and are initially referred for orthodontic treatment followed, if necessary, by restorative treatment. The screening programme is targeted at early detection of dental caries, and detection of congenital absence of teeth is often an opportunistic finding. As a result, in some cases, diagnosis is not made until much later and this delay in diagnosis and management can result in some of the social embarrassment problems alluded to by Hobkirk *et al.* (1994). Understanding the impact of hypodontia on patients' QoL would provide an insight into the potential consequences of the condition in their day to day lives. Improving understanding of the impact of the condition would facilitate improved provision of oral health care and inform best practice guidelines.

The aims of the present study were to determine, using qualitative methods, the impact of congenital absence of teeth on functional and psychosocial well-being and to identify factors considered important to patients which should be incorporated into health status measures for population studies and clinical trials.

Subjects and methods

Ethical approval for the study protocol was granted by the clinical research ethics committee of the Cork Teaching Hospitals.

Qualitative methods are increasingly used in the area of dental public health research. By undertaking qualitative research, it is possible to explore social phenomena which is beneficial as it enables researchers and practitioners to gain an insight into patients' perspectives on oral health care practices (Silverman, 2000). There are a variety of qualitative methods that can be utilized; however, for the purpose of this study, a thematic analysis was adopted. A thematic analysis is an inductive method whereby key themes are identified and interpreted (Braun and Clarke, 2006).

Recruitment

As the aim of this research was to evaluate the experiences of patients with hypodontia, a sample with the condition was recruited from those attending the Cork Dental School and Hospital, Ireland, for the treatment of hypodontia.

This centre is the only teaching hospital outside of Dublin and receives referrals for secondary and tertiary level dental care from a catchment area with a population of approximately 1 million. The recruitment strategy was to have a sample consisting of patients, both male and female at various stages of treatment with either mild, moderate, or severe hypodontia. Since the participants had previously completed a 49-item Oral Health Impact Profile (OHIP) questionnaire, this was also incorporated into the sampling strategy. Qualitative research is non-probabilistic and therefore, small sample sizes are common. Sample recruitment in qualitative research is dependent on the concept of saturation whereby data are collected until saturation is met. Saturation is met when no new themes are emerging from the data. Guest *et al.* (2006) outlined that data saturation occurs after 12 interviews. However, this varies across studies and key themes can emerge after the completion of as few as six interviews.

Sample

A sample of 10 participants provided informed written consent to participate in the study (for participants aged under 18 years, this was also agreed to and signed by the participant's parent). The subjects were made aware that participation was voluntary and written and oral information regarding the study was provided. Patients were interviewed using a topic guide with themes based around patient QoL. The guide was updated after each interview until saturation was reached. After completion of the interview, each participant was assigned a pseudo name to protect their anonymity and a subject code number. All participants were Irish and were undergoing treatment which varied according to the severity of the hypodontia. The participants ranged from 16 to 25 years of age of whom five were male and five were female. The summary scores for the previously completed a 49-item OHIP questionnaire ranged from 24 to 143. Characteristics of the sample are outlined in Table 1.

Data collection

Semi-structured interviews were conducted by one author (SM). These interviews were digitally recorded and professionally transcribed. Interviews were conducted either in a room in the dental hospital or in a location convenient to the participant. On completion of the interview, a voucher to the value of €30 was given to participants as an acknowledgement of her/his contribution. A constant comparative method was utilized whereby data collection and analysis proceeded concurrently until saturation was met, meaning no new themes arose from the participant's responses. As the themes which emerged from the data were, adequately explained by the existing data extra data collection was not required (Silverman, 2000).

Table 1 Overview of the sample.

Participants	Age (years)	Oral Health Impact Profile summary score	Number of missing teeth	Position of missing teeth	Time delay to orthodontic consultation after diagnosis (years)	Time delay to prosthetic consultation after orthodontic (years)	Time delay to restorative treatment after consultation (years)
Males	1	32	6	Antero-posterior	1	4	4
	2	24	10	Antero-posterior	4	6	4
	3	24	8	Anterior	0	4	4
	4	66	5	Antero-posterior	0	8	3
Females	5	61	9	Antero-posterior	0	7	6
	6	27	9	Antero-posterior	1	2	4
	7	76	4	Antero-posterior	3	4	1
	8	106	6	Posterior	N/A	4	1
	9	143	2	Anterior	3	3	1
	10	57	19	Antero-posterior	7	4	1

Data analysis

The overall objective of this study was to identify, interpret, and understand the experiences of patients with hypodontia; therefore, an interpretive methodology was utilized. Once the transcribed data were available, a thematic analysis was undertaken. Transcripts were imported into a qualitative data analysis software package (NVivo 7; QSR International, Melbourne, Australia) and data were sorted as the transcripts were coded on a line by line basis. NVivo software is a package which imports and stores data relating to qualitative studies such as transcripts, audio recordings, and relevant literature. This software allows qualitative data to be managed by sorting and organizing the data into the various categories and themes. A coding framework was developed around the topic guide and the key themes were identified as they emerged as the data were analysed.

Results

Analysis of the data indicated three key themes in relation to patients' experiences of both living with and being treated for hypodontia. Standard practice in qualitative research is to present direct quotations from subject interviews to illustrate these themes. Appendix 1 contains direct extracts from the subject interviews in this study and the comments attributed to a participant by number within the following text.

Theme 1: Transition from childhood to adulthood

All participants were initially diagnosed while attending primary school, but their treatment did not commence until mid to late adolescence. This highlights how the patients' expectations and participation in the process of their treatment changed from initial diagnosis to current treatment. At the stage where participants were initially diagnosed, they would have been in primary school, aged between 7 and 10 years of age and their parents were responsible for decisions on treatment at the time of the diagnosis. Participants highlighted how their comprehension of the condition and the implication for the future treatment which they would have to undergo was limited when initially diagnosed (participants 7 and 4).

Each of the participants indicated that as they made the transition from childhood to adulthood, they had a much better understanding of what their condition was and what was involved in their treatment. This was reinforced by the fact that the treatment did not begin until they reached late teens or early adulthood. The participants were more inclined to take responsibility over the choices of their treatment with the parents influence declining compared with when they were initially diagnosed. It is important to note that no participant felt that they would have changed any of the treatment their parents chose for them when they were children (participants 5 and 8).

Theme 2: Waiting time for treatment

All the participants were receiving treatment through the Irish public dental service and were managed by clinicians working in different centres. At present, there is no integrated or formalized service for management of patients with congenital anomalies of teeth in the south-west of Ireland. Accordingly, patients move from one waiting list (e.g. for orthodontic treatment) to another (for restorative dental care) and experience lengthy delays in completing their treatment. Participants outlined very strong feelings of frustration with the delay between initial diagnosis and initial treatment. Participants clearly stated that throughout that time they felt forgotten. Many felt that improved communication during that time would have eased this frustration (participants 2, 6, and 7).

Once initial treatment began, each participant spoke of their satisfaction that the treatment had begun and how expectations rose and assumptions were made about the length of time it would take to complete treatment. Frustration once again arose due to the complications involved in completing treatment and the length of time that it took (participants 10, 4, and 9).

Theme 3: Importance of aesthetics

As mentioned before, patients are initially diagnosed in primary school at around 9–10 years of age. Their treatment typically did not begin until mid to late teenage years. These are formative years where a person's perception of their appearance can have an impact on how they adapt to their social settings. Patients stated that as they became older they increasingly became more aware of their appearance due to the development of their teeth compared with those of their peers (participants 5, 1, 9, 7, and 4). Participants modified their behaviour to conceal their teeth as they felt they were not aesthetically pleasing and felt uncomfortable in social surroundings.

The primary motivation for both current and future treatment was that of aesthetics. The participants felt that the treatment would provide the 'cosmetic look' which in turn would result in feeling less self conscious and more comfortable in their social surroundings. Data were analysed to assess the participants' satisfaction or dissatisfaction with their treatment. Overall, all participants were happy with their treatment so far in respect to appearance and durability. These were the primary two factors in the success of the treatment. Dissatisfaction arose with regard to time delays and also slight discomfort; however, these factors were not as influential on the participants as that of appearance and durability (participants 10, 3, and 6).

Discussion

The aim of this study was to explore the experiences and perceptions of patients with hypodontia. Qualitative analysis

enables researchers and dental practitioners to broaden their understanding of the impact this condition can have on the patient and that in turn can influence future policy and practices in oral health care. There are some limitations which are important to highlight. Qualitative studies are not generalizable to the overall population and are context specific. However, qualitative methods are valid in order to provide insight into attitudes and opinions of patients. These themes, which are highlighted from qualitative analysis, can then be incorporated into questionnaire development which will allow for further enquiry (Neergaard *et al.*, 2009). To date, this appears to be the first study to use qualitative methods to examine the impact of hypodontia on patients with the condition.

The findings highlighted key issues which are of importance to oral health practice with regard to patients with hypodontia. A great deal of work has been carried out in recent years examining and evaluating patients' satisfaction and dissatisfaction with oral health practices and services. All participants involved in the study illustrated satisfaction with the outcomes of the treatment they received. Previous anxiety over the appearance of each participant's mouth or teeth was considerably reduced once treatment was completed or near completion. It has been well documented that a patient's perception of their appearance and its deviation from the norm, or minimum standard of acceptability (Davis *et al.*, 1998; Trulsson *et al.*, 2002) is a major influencing factor in the decision to undergo orthodontic and restorative dental treatment. Davis *et al.* (1998) stated that 'it also indicated that patients tended to feel, not that their appearance attained some ideal of beauty, but rather it was normalized and no longer needed to be a source of concern'. The findings of this study mirror those of Davis *et al.* (1998) whereby patients indicated that cosmetic appearance was the most influential factor in the choice of current or future dental treatment.

Patients who suffer from hypodontia have a number of needs with regard to treatment as the symptoms of their condition can vary considerably. In most cases, they will need both orthodontic and restorative treatment. Recent trends illustrate that patients are becoming more aware, and expectations of treatment and service provision have risen dramatically. Patients are more involved in the decision-making process in particular as their rights are coming to the fore in dental practice (Cronin *et al.*, 2009). The findings of this study illustrate that hypodontia patients are a unique group whose role in decision making, as well as their relationship with the dental practitioners involved, changes dramatically from initial diagnosis over the course of the treatment.

Each participant was initially diagnosed in primary school during routine screening for dental disease. The benefits of such a programme have been illustrated elsewhere with many countries implementing such programmes (Oliver *et al.*, 1997). However, it is obvious from the findings that

comprehension of their condition and the necessary treatment are not clear to these patients between the ages of 7 and 10 years. It was clear that during the stages of early adolescence, patients became more aware and concerned about their condition. The lack of understanding of the treatment that they were to undertake from the various disciplines was the main source of dissatisfaction. Consideration needs to be given with respect to the implications that this has. Patients illustrated strong feelings of social discomfort and a lack of understanding of the process which was to be undertaken in the coming years. The results suggest the importance of improving communication with patients with hypodontia throughout their early teenage years; perhaps the introduction of a programme or clinic designated to these patients would alleviate this problem. The participants illustrated that during their early teenage years, they did not understand that the delay in treatment was not due their condition and a necessity for development before treatment was appropriate but rather a belief that they were forgotten and left on a waiting list. The introduction of a programme dedicated to such patients would allow for practitioners to communicate with subjects as they reach an age where they are more cognizant of their condition so that they can then be informed once again of what the condition is, as well its implications and the best course of action with regard to their treatment. Oliver *et al.* (1997) stated that the combined clinic, including orthodontic and restorative consultants and practitioners, provides high quality care for the patient in a resource-efficient manner. Hobkirk *et al.* (1994) highlighted that there are two core standards which need to be met with regard to care for patients with ectodermal dysplasia and hypodontia whereby all patients should have access to informed health care professionals and that these health care providers work closely with patients. A combined clinic would allow for increased communication and co-operation not only among practitioners but also with patients and perhaps ease some of the anxiety and frustration illustrated by this group. Although context specific, this study illustrates how important it is for patients with congenital absence of teeth to be able to access appropriate care in a timely fashion. In addition to a dedicated multidisciplinary referral clinic, it will be important that community-based screening programmes are modified to include detection of congenital anomalies as part of their remit. Successful implementation of such a programme would reduce the incidence of delayed diagnosis and hopefully overcome some of the problems alluded to by the patients in this research.

Conclusions

The findings of this study show that patients have difficulty understanding their condition and its implication for treatment at initial diagnosis. Time delays between diagnosis,

orthodontic, or restorative consultation and the initiation of treatment were of particular concern. As treatment was initiated and completed, psychosocial impacts with regard to anxiety concerning facial aesthetics reduced. The results have implications for the service provision to patients who have hypodontia and how they are managed from diagnosis to completion of treatment. Improved services need to be developed which are patient focused and will reduce the frustration of patients while waiting for treatment to begin.

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Appendix 1 Verbatim extracts from subject interviews to illustrate themes

Theme 1: Transition from childhood to adulthood

Participant 7: ‘... well when you’re eight or nine you don’t really know what, you know you’re there with your parents and they make the decisions and you go along with them ...’

Participant 4: ‘... the dentist was talking to my mother and the ones [teeth] that hadn’t come through and I was in the room but he wasn’t speaking directly to me because I was a child, but it kind of didn’t really sink in. I didn’t understand the gravity of what missing teeth meant ... although my comprehension of it now is, it couldn’t compare to what I would have had at eight or nine’.

Participant 5: ‘... now I am kind of much more independent about coming here and decisions I make ...’

Participant 8: ‘She [mother] never really comes in with me. The first day she kind of came in ... they [dentist] were getting the colour [pigment of denture] done again today. I was like, ‘Oh I like this one.’ And they asked my Mom and they went with the one that she liked. ... It’s a bit frustrating because it’s going into your mouth. You kind of want you to like your own ... I’d rather go in on my own ...’

Theme 2: Waiting time for treatment

Participant 2: ‘... And I’d come here [Cork Dental Hospital] and they did a few moulds and I didn’t hear anything more about it for years, you know what I mean. ... then eventually came round to getting into the system and going ahead but I, by the time I started proper treatment I’d have been in transition year [16 years old] or so ...’

Participant 7: ‘... but I suppose the years went by and I suppose I wanted treatment but I didn’t know if I was still on the waiting list and I think that my mother rang about it a few times just to check am I on the waiting list. And I got verification and I just kind of sat it out and waited’.

Participant 6: ‘Oh well there was a big gap, after we did the all the X-rays, and they said that they would call me for an appointment, and it was two years and we hadn’t heard anything from them and then Mum rang them and said ‘What’s going on? You know you told us that she has a problem and you haven’t come back to us at all’.

Participant 10: ‘I remember I asked them years ago, do you know would it be done by, I think I said my 21st. You know this is when I first started with the braces and everything. And they started laughing like ‘of course it will be done!’ and like you know its nowhere near ...’

Participant 4: ‘... Part of the problem, I mean I would get an appointment and it would be a year before I’d get another one’.

Participant 9: ‘I just, I don’t know, I didn’t think that it was going to go on as long as it was. I thought maybe a year top, but you know. It’s not really, it goes on longer than that’.

Theme 3: Importance of aesthetics

Participant 5: ‘... I suppose when you come to that get to that age where you are kind of self consciousness or self awareness of 13. And a certain kind of disparity between me and my peers ... I don’t have teeth and they have teeth! And I thought like, ‘wait a minute, what’s going on here?’ and it was only at that stage really that it became I would exactly say a major issue ... it was quite a concern for me and it bothered me a lot. I was really self conscious ...’

Participant 1: ‘I knew my teeth were slightly odd. People used to say, you know ‘What’s wrong with your teeth?’ Or ‘Did you break them?’ And I used to say, ‘No I inherited them from my granddad’. So yeah, I, I know myself but I wasn’t too annoyed ...’

Participant 9: ‘I would kind of put my hand up to my mouth. I kind of still do that sometimes but not a lot, not as much as I used to’.

Participant 7: ‘Well, well I was always self conscious about my teeth. When I was very young ... I would always put my hand in front of my mouth when I smiled or laughed or anything like that. And as I get older I kind of got out of that habit but I was always very conscious of them.

Participant 4: ‘Well I wasn’t too pushed when I was young, but obviously in teenage years, everybody becomes self conscious of how they look and stuff, so because I was kind of growing older I became conscious of it and when I smiled I would bring my top lip over my teeth and when I was talking to you I did that and it was kind of funny after the braces, I had braces about three years at this stage, and I would still bring my top lip over when I smiled. It was a weird habit that I had and one of my friend’s mother didn’t realise I had braces at all—three years—because I would kind of hide it that way from habit’.

Participant 10: ‘It’s more the appearance thing, the missing teeth and things like that. That’s kind of what, being a bit vain, that’s what kind of would spur you on into action really’.

Participant 3: ‘I was so delighted with it [bridge] really to be honest. And I got it just before Christmas so it was a good Christmas present’.

Participant 6: ‘I was very proud of the bridge though. I was pretty happy with it’.

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