



Parents' perspectives on the use of children's facial images for research and diagnosis: a survey

Lize Schoeman¹ · Engela M. Honey¹ · Helen Malherbe² · Vinet Coetzee¹

Received: 14 July 2022 / Accepted: 30 September 2022 / Published online: 10 October 2022
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Abstract

Computer-aided facial diagnostic tools are valuable emerging technologies for the early detection and initial diagnosis of congenital disorders. These tools require large datasets of facial photographs, especially of infants and children, to identify these disorders and improve classification accuracies. Researchers need to balance this need for larger datasets with patients' privacy rights, needs and preferences. This study aimed to investigate parents' views regarding the collection, storage, use and publication of their children's facial images for research and diagnostic purposes. A total of 151 parents of children with and without congenital disorders completed an online survey evaluating their views on the collection, storage, use and publication of children's facial images for research and diagnosis. Overall, 72.5% of parents would allow researchers to take facial photographs of their children, preferring the images to be stored in a secure database that is not available to the public. Parents of children with congenital disorders were more accepting of researchers taking facial photographs of their children, compared to parents of children without these conditions. Half of the respondents would allow facial photographs of their children to be published in academic journals, without their eyes covered, and this acceptance increased as the proportion of the child's face covered increased. Parents also indicated specific requirements to allow the use of these images in other similar research studies which need to be taken into consideration when planning studies that involve facial analysis research.

Keywords Facial analysis · Patient perspectives · Facial images · Survey · Facial photographs · Congenital disorders

Introduction

Photographs are a powerful tool in a clinician's toolbox that add to the care and treatment of patients. Clinical photographs can be used to educate clinicians as well as patients and families, form part of medical and academic publications, and can be incorporated into medical consultation and documentation as part of the care of the patient (Harting et al. 2015). Facial diagnostic tools are an emerging health technology that can be used to detect and diagnose a range of conditions and features. Such tools have been used to identify and quantify specific facial features and classify various genetic disorders, autism spectrum disorders, foetal alcohol spectrum disorder and schizophrenia with accuracies of up

to 91% (Hennessy et al. 2007; Fang et al. 2008; Hammond et al. 2008; Aldridge et al. 2011; Suttie et al. 2013; Ferry et al. 2014; Gurovich et al. 2019).

Congenital disorders (CDs) are defined as abnormalities in structure or function present from birth (World Health Organization 2006). Causes of CDs include genetic or partially genetic factors, environmental factors (post-conception) or a combination of these and unknown factors (Christianson et al. 2006). An estimated 30–40% of all CDs have associated craniofacial features and a facial phenotype (Hart and Hart 2009). Clinical facial photographs may therefore be used to screen, identify and assist in diagnosing patients with these conditions. Multiple computer-based techniques have been developed by researchers to identify CDs based on the associated typical facial gestalts in both two-dimensional (2D) or three-dimensional (3D) facial images (Loos et al. 2003; Hammond et al. 2004, 2005; Boehringer et al. 2006; Ferry et al. 2014). The commercially available web tool Face2Gene (<https://www.face2gene.com/>) (FDNA Inc.), driven by DeepGestalt, allows clinicians to evaluate the phenotype of their patients and rank likely diagnoses

✉ Lize Schoeman
lize.schoeman@me.com

¹ Department of Biochemistry, Genetics and Microbiology, University of Pretoria, Pretoria, South Africa

² Rare Diseases South Africa NPC, Sandton, South Africa

using the facial photograph and other relevant information provided (Gurovich et al. 2019). This application, together with other computer-aided diagnostic tools, is enabling earlier and more accurate diagnosis of CDs (Rai et al. 2015). Low-cost facial analysis technologies are particularly useful for diagnosing children born with CDs in low-resource settings, where there are competing health care priorities and inadequate capacity (Porras et al. 2021).

The use of facial photographs in a clinical medical setting is a sensitive issue. Since individuals are readily identifiable from their facial photographs, protecting and maintaining a person's privacy and identity when using facial images are challenging. Privacy regulations, such as the European Union (EU) General Data Protection Regulation (GDPR) (European Union 2016), the South African Protection of Personal Information Act (POPI Act) (Republic of South Africa 2013) and the Australian Privacy Act 1988 (Australian Government 1988), aim to safeguard people's personal information. In addition to complying with relevant privacy regulations when conducting facial research, the needs and preferences of the patients and family members asked to supply facial photographs for research and diagnosis purposes should also be sought and understood. Comprehending these preferences will help guide better practice for future research in this field.

Previous studies investigating patient perspectives on medical photography found that individuals generally prefer the use of non-identifiable over identifiable photographs for medical purposes (Lau et al. 2010; Adeyemo et al. 2013; Leger et al. 2014). While a positive outlook was reported on clinical photography (Leger et al. 2014), patients are less accepting of clinical photographs that include the head and face (Qudaisat et al. 2021). Parental preferences on the medical photography of their children are less well studied, with only one study investigating this topic in a dermatology context (Hacard et al. 2013). To our knowledge, no previous studies have examined parents' preferences on facial photography of their children. There is also a well-documented lack of research in populations residing outside Europe and the USA in general (Henrich et al. 2010).

This study aims to better understand parents living in Africa's preferences on the collection, storage, use and publication of their children's facial photographs for research and diagnosis.

Materials and methods

Ethics approval

This study received ethical approval from the Research Ethics Committees of the Faculty of Natural and Agricultural Sciences and the Faculty of Health Sciences at the

University of Pretoria (NAS117/2021). All procedures followed were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Survey

The developed Qualtrics (Qualtrics, Provo, UT) online survey consisted of a brief introduction and a minimum of 34 questions. This included questions on basic demographic and clinical information, and parental preferences on the collection, storage, use and publication of their children's facial images (Online Resource 1). A Likert scale was used to measure responses on scale of preference and was followed by additional open text questions on feelings underpinning these responses. Certain responses elicited additional questions. The survey was pre-tested on selected collaborating academics ($N=5$) and piloted on a convenience sample of parents ($N=10$). The survey was modified using feedback from the pilot groups before distribution to the larger target audience: parents of children with and without CDs. The term 'inborn conditions' (see survey section B Online Resource 1 for definition given to respondents) was used in the survey to optimise the understanding of participants, and in this context was considered equivalent to the definition of CDs. The survey was distributed electronically (via a URL) to acquaintances (i.e. colleagues and other parents the research group is in contact with) and disease-specific patient advocacy organisations. Snowball sampling was applied, where participants were requested to recruit other participants (Naderifar et al. 2017). The URL was also posted to disease-specific advocacy organisations' social media platforms (i.e. Facebook) for users to follow and complete. The survey was limited to English-speaking individuals. All responses to the survey were anonymous and no personal identifying information (i.e. names, ID numbers, email addresses, etc.) was collected.

Analyses

For the interpretation of the results, indicated responses on the Likert scale of 'Definitely yes' or 'Probably yes' were interpreted as agreement with the statement, 'Might or Might not' as no preference or unsure, and 'Probably not' or 'Definitely not' as disagreement with the statement. Statistical analyses, including descriptive statistics and logistic regression, were performed using SPSS for Macintosh version 27 (IBM SPSS Statistics). Text responses for specific questions were individually coded and analysed thematically using ATLAS.ti 9 software (ATLAS.ti Scientific Software Development GmbH), as described by Braun and Clarke (2006).

Results

A total of 193 survey responses were received. Two spam responses, 29 partial responses and 11 responses from individuals without children were excluded, leaving 151 completed survey responses for analysis.

Demographics

Most respondents were female, with a high level of education and a mean age of 43.7 years ($SD=8.977$, 10 respondents did not indicate; Table 1). The mean number of children per respondent was 2.06 ($SD=0.788$). See Table 1 for a summary of respondents' demographic information and survey sections A and B (Online Resource 1) for survey questions.

Inborn conditions/CDs

The 42 parents of children with a CD were asked further questions regarding the diagnosis, age of child at diagnosis and age of parent at diagnosis (see survey section B; Online Resource 1). A summary table of their responses can be seen in Table 2. Overall, 16.3% of mothers were of advanced maternal age (AMA; indicated with ^e) when their child with a CD was born (Table 2). Down syndrome was the most common diagnosis (34.8%, $n=16$). Overall, 39.1% ($n=18$) of children were diagnosed in the first month, 69.6% ($n=32$) in the first 2 years and 84.8% ($n=39$) in the first 6 years.

Facial analysis research

Collection of facial photographs

Parents were asked if they would allow researchers to take facial photos of their children (survey section C; Online Resource 1). Of the 149 respondents answering this question, 72.5% ($n=108$) indicated they would permit researchers to take facial photos of their child/children, while 6.0% were unsure, and 21.5% ($n=32$) would not (Fig. 1). Parents who would not allow researchers to take a photo of their children were not asked any further questions. The remaining results are reported for the remaining 119 completed surveys only.

Parents of children with CDs (28%, $n=42$) were significantly more likely to allow researchers to take a photo of their children than parents of children without a CD (logistic regression: $\chi^2(4)=8.370$, $p<0.004$; Fig. 1) and none would prevent ('Definitely not' allow) researchers taking such facial photos (Fig. 1). In comparison, 6.5% of respondents

without CD-affected children (72% and $n=108$) would 'Definitely not' allow facial photos to be taken of their children, and 20.6% would 'Probably not' allow it (Fig. 1).

Of those respondents who would allow photos to be taken, most (87.9%, $n=102$) indicated that they would allow researchers to take both 2D and 3D photographs. The analysis of the underlying feelings for these parental preferences on the taking/collection of their child/children's facial photos revealed four themes, three positive and one negative:

1. Contributing a photograph may help: Respondents ($n=28$) felt that they would contribute facial photographs to studies because they see value in the research, and indicated that these photos could help with the diagnosis of CDs.
2. Knowledge and awareness of CDs: Seven respondents indicated they would like to improve knowledge and awareness around CDs (both for their own and others' benefit) and could contribute to this by allowing photography of their affected children.
3. Conditions for contributing a photograph: Ten respondents indicated that they would allow researchers to take photos of their child/children if it would help their own children or others and have a positive contribution to research and if the photos are taken under strict controls.
4. Safety and protection of children: Respondents ($n=6$) were concerned about the safety of their children's identity and photographs if made publicly available or to individuals who may not handle the photographs respectfully.

Storage of facial photographs

Respondents were asked if they would allow researchers to store their child's facial photos, across four different scenarios (survey question 13; Online Resource 1). The greatest preference was seen for storage in a secure database only accessible by the original researchers (92.2%, $n=107$), followed by a secure database accessible by other pre-approved researchers (79.5%, $n=89$) and/or doctors, but not the general public (73.7%, $n=84$; Fig. 2). However, only 13.9% ($n=15$) would allow the storage of their children's facial photographs in a publicly available database (Fig. 2).

Three main themes were identified when respondents were asked why they feel that particular way about the storage of their children's facial images, specifically the following:

1. Relevance of access: Sixteen respondents felt that only relevant people (medical professionals and researchers) should be able to access the stored facial photographs, and not the general public.

Table 1 Profile of survey respondents

	Description	n (% of cases)
Sex/Gender	Female	132 (88.0)
	Male	18 (12.0)
Age of respondent (years) ^a	25–29	5 (3.5)
	30–34	24 (17.0)
	35–39	19 (13.5)
	40–44	22 (15.6)
	45–49	33 (23.4)
	50–54	25 (17.7)
	55–59	6 (4.3)
	60–64	6 (4.3)
	65–69	1 (0.7)
	Education	Grade 10
Grade 11		2 (1.3)
Grade 12		25 (16.7)
Higher certificate		12 (8.0)
Diploma		31 (20.7)
Bachelor's degree		27 (18.0)
Bachelors honours degree		29 (19.3)
Master's degree		14 (9.3)
Doctor's degree		9 (6.0)
Ethnic/Language grouping	Coloured	7 (4.7)
	Pedi	1 (0.7)
	South Sotho	1 (0.7)
	Tsonga	1 (0.7)
	Tswana	2 (1.3)
	Venda	1 (0.7)
	White	128 (85.9)
	Xhosa	2 (1.3)
	Zulu	2 (1.3)
	Foreign	1 (0.7)
	Other	2 (1.3)
Prefer not to say	1 (0.7)	
Location	Africa	
	- South Africa	117 (95.1)
	- DRC	1 (0.8)
	- Zimbabwe	1 (0.8)
	Other regions	4 (2.3)
Number of children	One	35 (23.3)
	Two	78 (52.0)
	Three	30 (20.0)
	Four	7 (4.7)
Age of children (years) ^b	0–4	53 (17.3)
	5–8	40 (13.1)
	9–14	64 (20.9)
	15–19 and older	149 (48.7)

Table 1 (continued)

	Description	<i>n</i> (% of cases)
Children with/without CDs ^c	No CDs	108 (72.0)
	With CDs	42 (28.0)
	Proportion of children with CDs:	
	- 1st born	19 (45.2)
	- 2nd born	15 (35.7)
	- 3rd born	9 (21.4)
	- 4th born	3 (7.1)

DRC, Democratic Republic of Congo.

^aAge ranges of respondents organised according to Statistics South Africa (2021) Statistical release P0302. Mid-year population estimates. Pretoria: Stats SA.

^bAge ranges of children adapted from Statistics South Africa (2021) Statistical release P0302. Mid-year population estimates. Pretoria: Stats SA to accommodate survey options.

^cIndicates the birth order of the children born with CDs.

2. Protection of their child/children: Protection of their children's identity, safety and privacy are important to respondents ($n = 18$).
3. Potential misuse of the photographs: Six respondents indicated nonspecific concerns around the respectful treatment of their children's images if made available to the general public.

Use of facial photographs

When asked if they would allow researchers to use their children's facial images in different instances, 95.6% ($n = 110$) of respondents would allow these images to be used for the original study they signed up for; 83.2% ($n = 94$) would allow their use for other, similar research studies with ethical approval, and 58.2% ($n = 64$) would allow them to be used for any research study that received ethical approval (Fig. 3).

Parents indicated the following prior conditions of use of their child/children's facial photos in other similar research studies:

1. Protection of children's personal information: Anonymity and maintaining the privacy of the images are important. The protection of their child/children's identities, personal information and confidentiality was a prerequisite for eighteen parents prior to considering the use of their child/children's facial images in other similar research studies.
2. Information regarding the study and re-use of photographs: Parents ($n = 12$) want to be informed when and if the images are to be used again for a research study, or wanted more information about the study in which the images will be used.
3. Contractual agreement between the researchers and parent: Respondents ($n = 3$) felt that researchers should sign

- a contract containing the uses of the images provided. This contract could indicate that the images will not be made publicly available and be used to ensure the ethical use and distribution of the images in another similar research study.
4. Approval for reuse of the image: Sixteen respondents indicated that they would like to provide consent again before the facial images are used in another similar research study. One respondent even indicated that if the child becomes of age when the new study requires their images, researchers should seek consent from the child before using the images again.
5. Availability and access to photographs: Respondents indicated various conditions on access to the photographs for reuse. Parents ($n = 12$) do not want the images to be available to the public and they should be securely stored and only accessible by medical professionals.
6. Ethical issues: Seven respondents indicated that the research study should be ethically approved and that both the research and handling of images should be conducted ethically and with respect.

Publication of facial photographs

Respondents were asked if they would allow researchers to publish their child's facial images in academic journals in different forms (see questions 18–21; Online Resource 1). Half (51.2%, $n = 60$) of respondents indicated that they would allow their child's facial photos to be published as individual images, without their eyes covered. Acceptance of publishing their children's photos grew as the child's identity becomes more protected, reaching 53.9% ($n = 62$) if their eyes are covered, 60.0% ($n = 69$) if large parts of their face are covered and 66.1% ($n = 78$) as a composite image.

Table 2 Children's diagnosis, age at diagnosis and parent age at birth of child

Diagnosis	All CD children ($n=46$), n (% of cases)	Age of child at diagnosis	Age of mother at birth of child ^d
18q deletion syndrome	1 (2.2)	2–3 months	41 ^e
22q11.2 deletion syndrome	2 (4.3)	3–4 years	25
		3–4 years	25
ADHD and cognitive delay	1 (2.2)	7–8 years	33
Angelman syndrome	1 (2.2)	5–6 years	(42)
Autism ODD, epileptic convulsions	1 (2.2)	5–6 years	27
Cerebral palsy	1 (2.2)	8–9 months	34
Charcot Marie Tooth	1 (2.2)	9–10 years	36
Chromosome 15 duplication	1 (2.2)	1–2 years	38 ^e
Cleft lip and palate	1 (2.2)	0–1 month	33
Cleft palate	2 (4.3)	8–9 months	24
		0–1 months	33
Congenital hyperinsulinism	1 (2.2)	4–5 months	34
Congenital trigger finger	1 (2.2)	1–2 years	31
Down syndrome	13 (28.3)	0–1 month	35
		0–1 month	31
		0–1 month	28
		2–3 months	28
		0–1 month	32
		0–1 month	34
		0–1 month	31
		0–1 month	(35)
		0–1 month	27
		0–1 month	32
		0–1 month	37 ^e
		0–1 month	40 ^e
		^a	43 ^e
	1 (2.2) ^b	0–1 month	45 ^e
	2 (4.3) ^c	0–1 month	41 ^e
		2–3 months	26
Fragile X	1 (2.2)	^a	20
Idiopathic scoliosis	1 (2.2)	9–10 years	36
Muscular myopathy	1 (2.2)	0–1 month	27
Myelomeningocele spina bifida	1 (2.2)	^a	35
Prader-Willi syndrome	3 (6.5)	8–9 months	23
		3–4 years	36
		2–3 months	36
Rett syndrome	5 (10.9)	1–2 years	30
		3–4 years	34
		1–2 years	25
		5–6 years	19
		^a	30
Tetralogy of Fallot and 22q11.2 deletion syndrome	1 (2.2)	0–1 month	27
Trisomy 18	1 (2.2)	0–1 month	34
Unknown	1 (2.2)	1–2 years	(28)
Woolly hair syndrome	1 (2.2)	1–2 years	36

^aRespondent did not provide an answer.

^bDown syndrome with sensory processing disorder.

^cMosaic Down syndrome.

^dAge of father indicated in brackets.

^eAdvanced Maternal Age (AMA). Woman ≥ 37 with conception is considered AMA in South Africa (Department of Health, Republic of South Africa 2015).

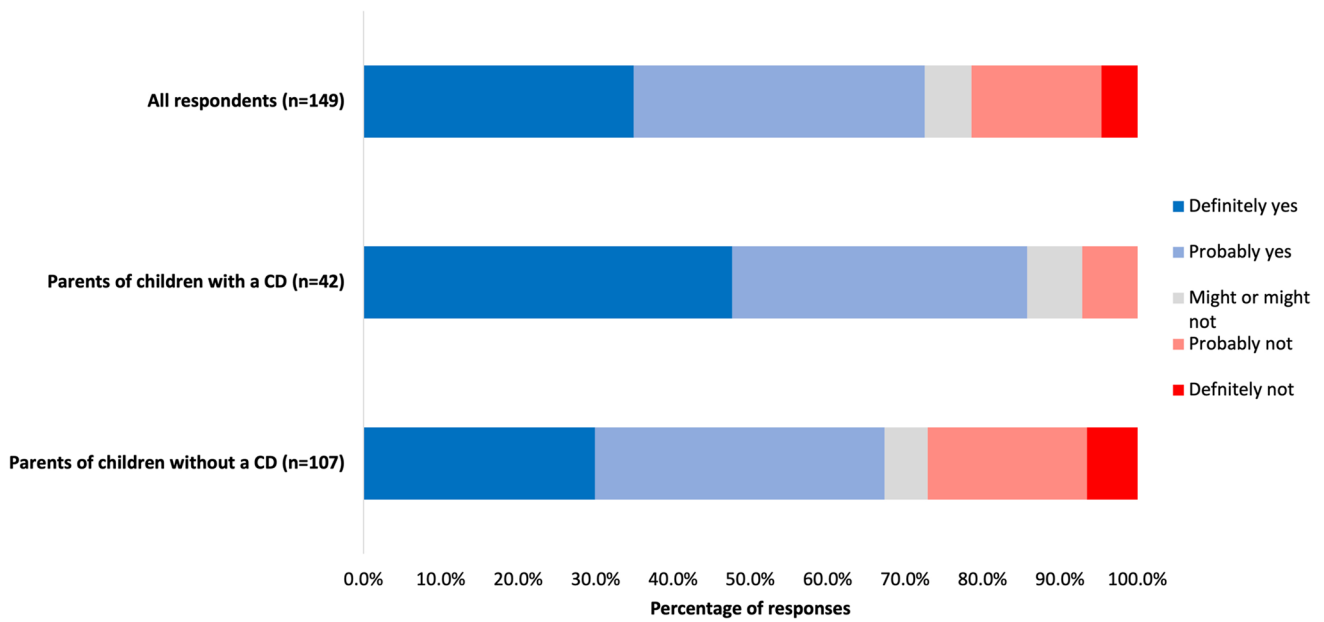


Fig. 1 Parents' attitudes towards the collection of their children's facial images for research and diagnosis

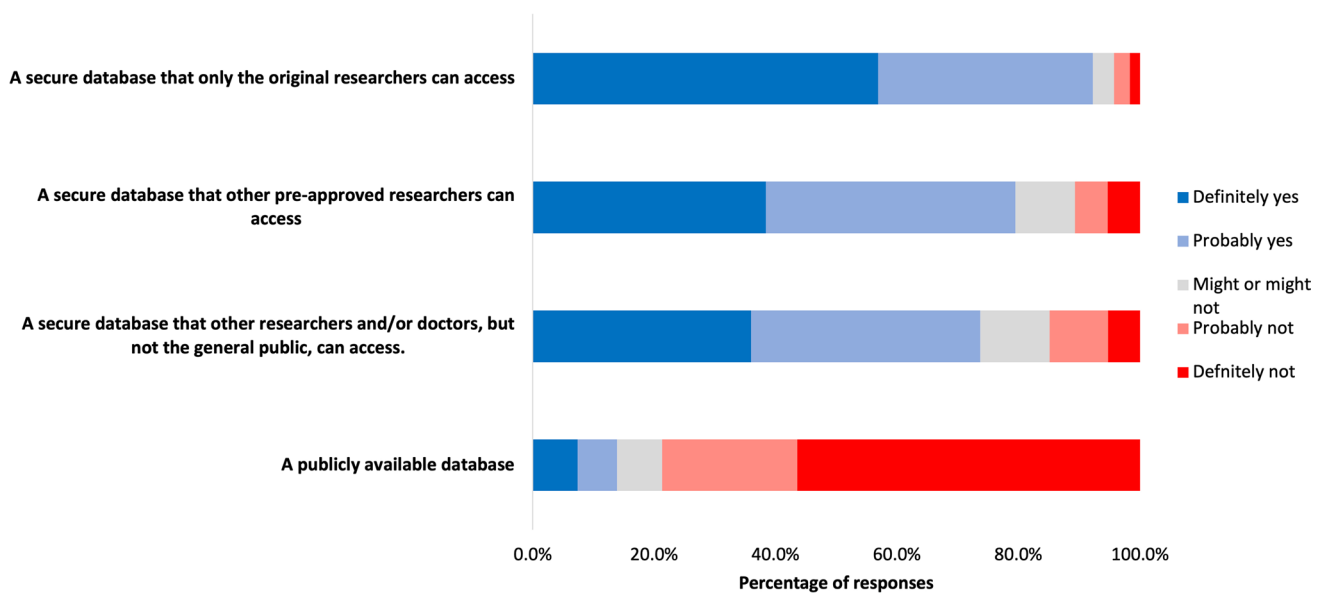


Fig. 2 Parents' attitudes towards the storage of their children's facial photographs

Respondents were then asked how they would allow their child/children's images to be presented on different platforms, and could select more than one option (Online Resource 2). There were similar preferences for newspapers, magazines and websites; thus, these scenarios are presented as an average. Respondents preferred that their children's facial images be presented at scientific conferences (29.8%), with a somewhat lower preference indicated for newspapers, magazines and websites (21.0%), and a strong dislike for publication on social media

(53.4%, $n = 62$). At scientific conferences, greatest preference was for their faces uncovered and unblended (40.5%, $n = 47$). However, in newspapers, magazines and websites, some preference was seen for use of a composite image (24.5%) over use of a facial image with covered/uncovered eyes or large sections of the face covered (16.3–21.9%). Amongst the small percentage of respondents that would allow their child's image to be presented on social media, most preferred that it be presented as an individual image with uncovered eyes (14.7%, $n = 17$).

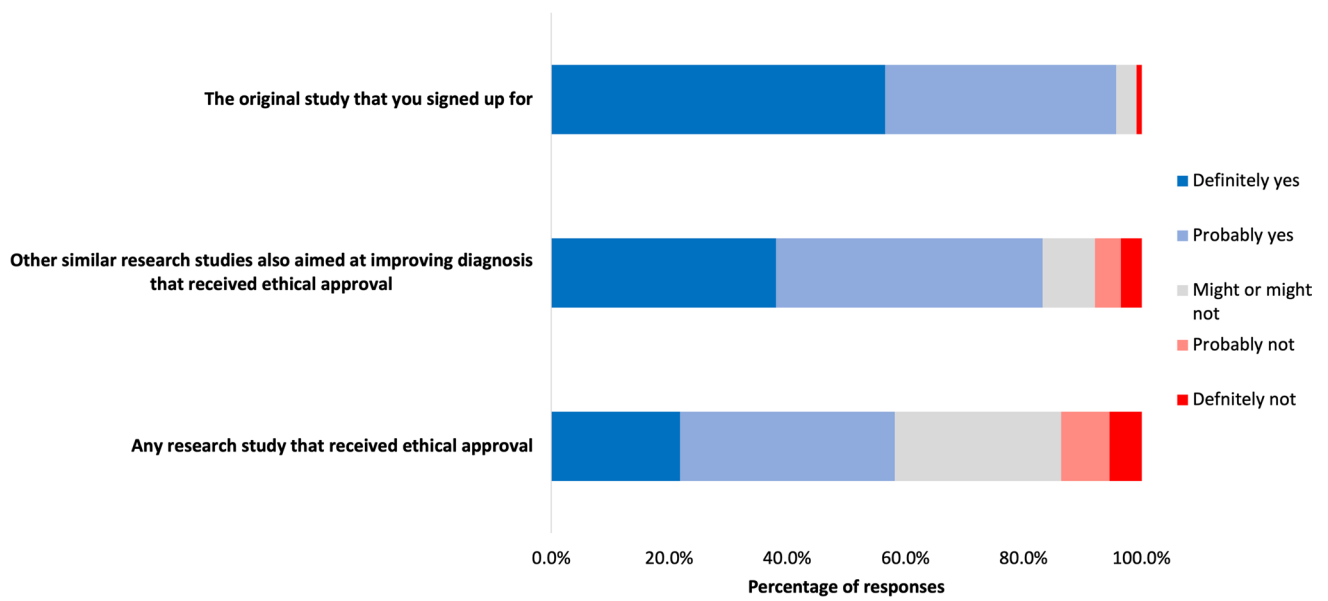


Fig. 3 Parents' attitudes towards the use of their children's facial photographs

The difference in attitudes towards the publication of facial images for parents of children with and without CDs was also examined. Respondents were divided into two groups: parents of children with CDs (hereafter the 'CD' group; Fig. 4) and parents of children without CDs (hereafter the 'no CD' group; Fig. 5). Most notably, 55.3% of parents

in the CD group would allow their child's facial images to be presented at scientific conferences without their eyes covered (Fig. 4), compared to only 32.5% of parents in the no CD group (Fig. 5). Parents in the CD group are more likely to share their child's facial photo on social media than parents in the no CD group (Fig. 4 and Fig. 5).

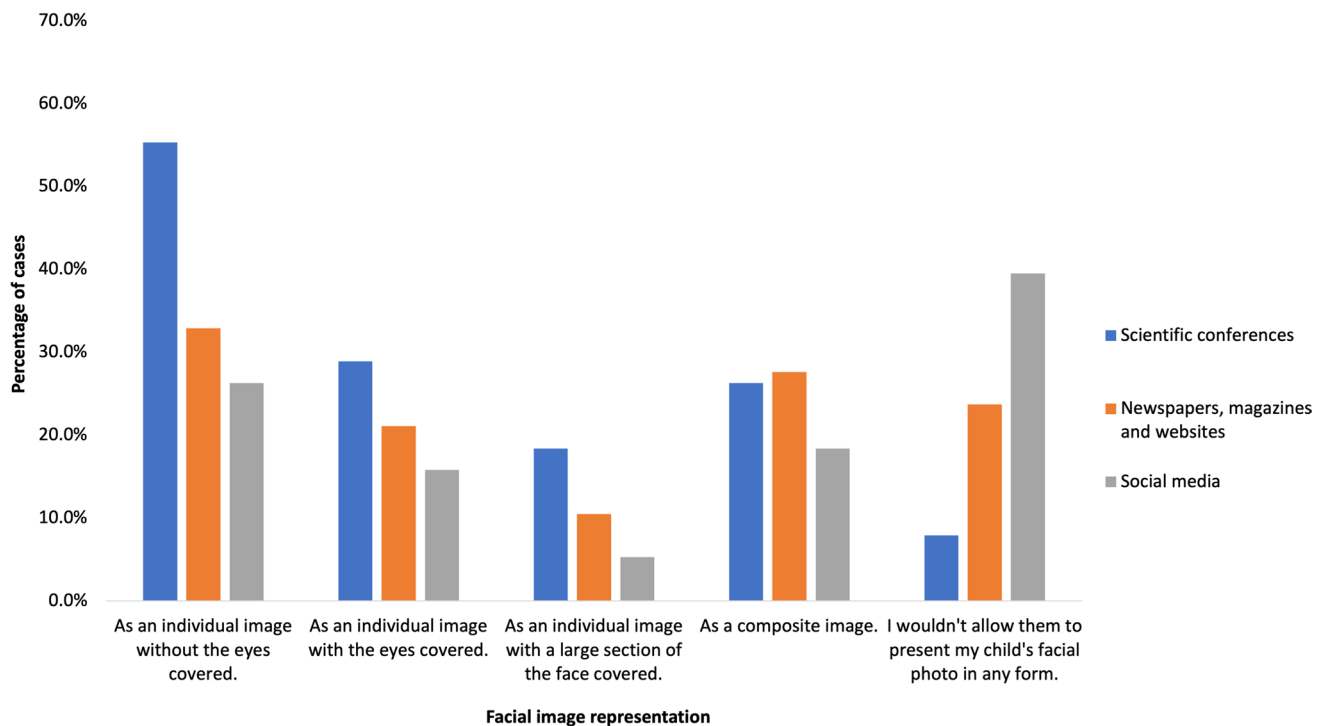


Fig. 4 Attitudes towards publication of facial images of parents with children with CDs

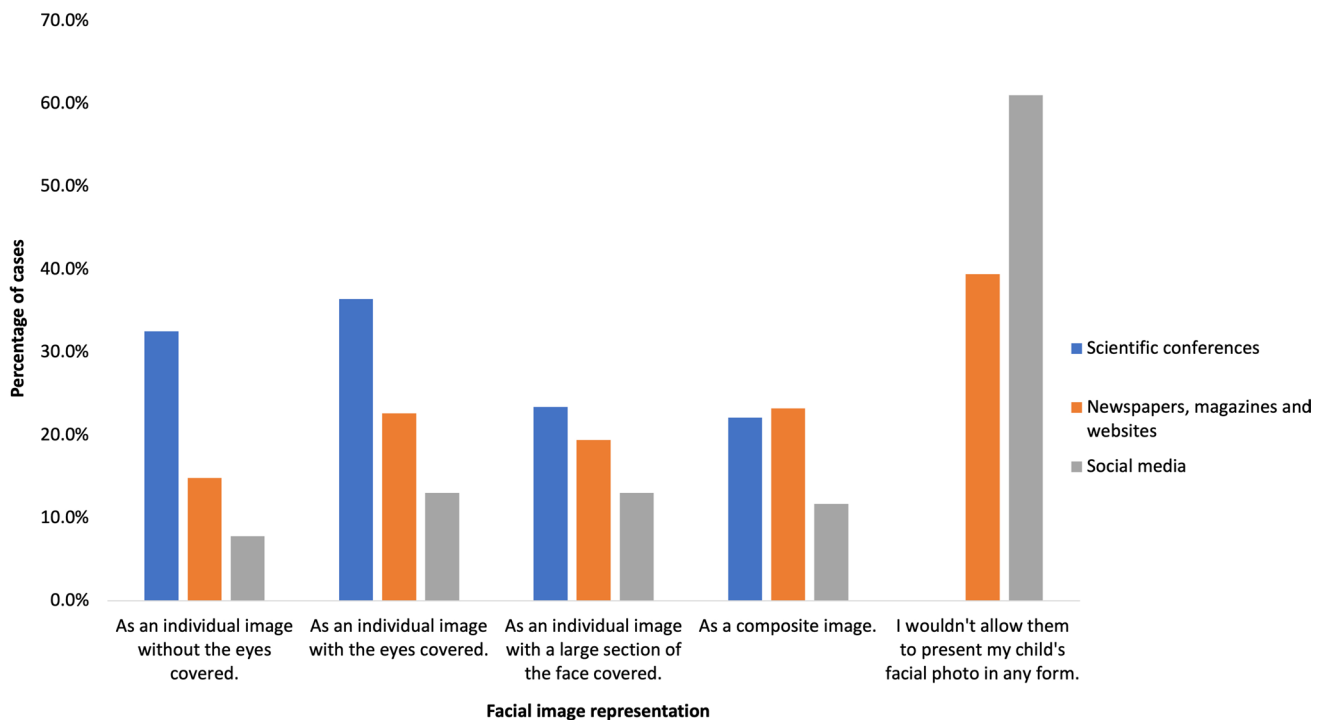


Fig. 5 Attitudes towards publication of facial images of parents with children without CDs

Five main underlying themes emerged in response to feelings underpinning these preferences:

1. **Protection of children and safety:** Respondents ($n = 13$) indicated the need to protect their children's privacy, safety and identity, especially in the context of images being published/available to the public.
2. **Negative consequences of public images:** Respondents ($n = 14$) are concerned about negative consequences for their child/children if the images are placed on publicly available platforms. Perceived risks included stigma, bullying and child trafficking.
3. **Social media concerns:** Six respondents indicated a negative view about social media and were concerned about the accessibility of the photos by the general public via these public platforms.
4. **Publication versus research use:** Four respondents felt that publishing facial images of their child/children was not aligned to the original research purpose of the study.
5. **Child's own decision:** Five respondents felt that ultimately, it should be the child/children's own decision around the publication of their images and consent/assent should be sought directly from the child at an appropriate age.

Photos versus delineation points

An alternative to storing actual facial photos is to store the information as data points representing the structure of the face, making them less easily identifiable (see question 27; Online Resource 1). When asked if respondents would feel differently about how their child's facial image is stored, used and published if the information was recorded as points and not as actual photos, 46.6% ($n = 55$) preferred points over actual photos. A further 39.8% ($n = 47$) showed no preference, and 13.6% ($n = 16$) preferred actual photos over points.

Attitudinal differences for different groupings

Babies vs older children

Following a short introduction (Online Resource 1, question 29a), respondents were asked if they would prefer the use of facial photographs of babies rather than older children for research and diagnosis. Over half of respondents (52.5%, $n = 61$) preferred the use of babies' facial photographs over older children, 31.9% ($n = 37$) were unsure and 15.5% ($n = 18$) did not prefer the use of facial photographs of babies

over older children. Those preferring the use of baby photographs felt that their use in facial analysis research would limit recognition later in life and protect their identity due to babies' features changing and maturing over time (see Online Resource 3 for full thematic analysis).

Children with CDs vs without CDs

Respondents were asked if there should be different rules for the collection, storage, use and publication of facial photos of children with versus without CDs. The majority (60.4%, $n = 70$) indicated that there should not be different rules, 9.5% ($n = 11$) had no preference and 30.2% ($n = 53$) preferred different rules for these two groupings. None of the respondents indicating there should not be different rules offered further explanation. Those who indicated there should be different rules suggested that children with CDs should be better protected and that their images required more privacy and stricter rules for use (see Online Resource 3 for full thematic analysis).

Children vs adults

When asked if there should be different rules for the collection, storage, use and publication of the facial photos of children versus adults, half of respondents (51.7%, $n = 61$) indicated that this should be the case, 6.8% ($n = 8$) were unsure and 41.5% ($n = 49$) indicated that the rules should be the same. Overall, respondents felt that facial photographs of children should be more protected and their identities and privacy should be better safeguarded, as a vulnerable group (see Online Resource 3 for full thematic analysis).

Discussion

In this study, we evaluated parents' views on the collection, storage, use and publication of their children's facial images for research and diagnosis. Overall, the majority (72.5%) of survey respondents would allow researchers to take facial photographs of their child/children. A significant difference in preference for allowing collection of facial photos was seen between respondents of children with a child affected by a CD versus those with unaffected children, where parents of affected children are more willing to allow facial image collection. Respondents felt that photographs should be stored on a secure database, only accessible by relevant parties. Parents indicated that ethical approval of the study is important and were more comfortable allowing publication of their children's facial photo the greater their identity is protected.

The positive preference towards sharing images is encouraging since numerous images of children, with and without

CDs, are required to train computer algorithms to detect and accurately identify typical facial features of these disorders. These results are similar to those of a study in Nigeria, where 72% of patients aged ≥ 16 years attending maxillofacial and plastic surgery clinics agreed to having their photos taken for medical reasons (Adeyemo et al. 2013).

Taking and collection of photographs

Although less than a third of respondents (28%, $n = 42$) had a child/children with a CD, their responses were significantly more positive (85.7%) to the collection of their photographs compared to parents of unaffected children (67.3%). None of those with affected children would prevent ('Definitely not') researchers from taking facial photos of their children. This may be attributed to their direct, personal experience of the diagnostic journey of their affected children. One respondent indicated that their child was first suspected to have a condition at a very young age, based on the facial features identified by doctors, which is the ideal scenario to enable early treatment and intervention to mitigate mortality and disability. These results are encouraging, indicating that parents of affected children are willing to contribute and appreciate the benefit of this research. However, it also serves to highlight that the value of sharing photographs is perceived differently when there is less personal benefit to the child and family.

Storage and access

Respondents' willingness to allow the storage of their child/children's facial photographs in databases decreased as the number of individuals who have access to the database increased. This suggests that while respondents may consider contributing facial images to assist research and diagnosis, there is a strong desire for security and privacy of the images and to protect the identity of the children. Respondents indicated greater trust of researchers and doctors than of the general public. This aligns with previous research that found patients preferred medical photos be stored in departmental records/devices (Hacard et al. 2013; Wang et al. 2017; Qudaisat et al. 2021), with only the relevant doctors and personnel involved in the patients' care (e.g. treating physician) having access to the photographs (Lau et al. 2010; Adeyemo et al. 2013).

Ethics

Over half of the respondents (58.2%) would allow their children's facial images to be used in any research study with ethical approval. Here, ethical approval refers to approval from the relevant institution's ethics board. This may be interpreted as individuals valuing the ethics approval above

the aim and potential benefits of the study itself, and may lack a comprehensive understanding of the ethics process. When asked about specific requirements for the reuse of images in other similar research studies (i.e. for potential incorporation into consent forms), parents indicated they would require repeat/additional consent for each instance of photograph reuse. They would also want specific information on who is granted access and the purpose of this access. If such guidelines are implemented, this may increase the number of parents agreeing to photograph collection and use.

Publication of photographs

While preferences for publication of facial photographs were found to be similar between parents of children with CDs and parents of unaffected children, they varied proportionally (Fig. 4 and Fig. 5). More parents of affected children would allow publication of photographs at scientific conferences without the child's eyes covered (55.3%) than those of unaffected children (32.5%). Parents of affected children could be more willing to allow uncovered publication at scientific conferences, possibly due to their own experience of doctors and researchers reaching a diagnosis based on the typical facial features of their child, as one respondent indicated earlier. Additionally, parents of children with CDs are more likely to allow social media publication of their children's facial images than parents of unaffected children, likely because social media can be such a powerful platform to raise awareness of CDs.

Academic journals

Respondents were generally accepting of publication of their child's facial images in academic journals in different forms ($\geq 51.2\%$). Interestingly, half of the respondents (51.2%) would agree to their child's facial images being published as fully identifiable individual images (without their eyes covered) in an academic journal. This may have been primed by the explanation accompanying the question (see question 18, Online Resource 1). Such trust towards researchers is consistent with the previous observation of limiting access to their child/children's images to only researchers and treating physicians.

The increasing acceptance of academic publications with the greater degree of redaction (i.e. the proportion of the face covered) rose to 66.1% acceptance when published as part of a composite image (completely de-identified). This is higher than the 46.5% acceptance observed in a study by Hacard et al. (2013) which focused on both adult patients and parents of paediatric patients—the latter group being much less accepting of anonymised photos published in scientific articles, compared to adult patients (67.4%), likely

because parents want to protect their children's identity and privacy. Other researches indicate 55% and 40% of individuals will allow identifiable images published in journals and websites respectively (Lau et al. 2010), which is higher than reported in our study (21.5% uncovered faces in newspapers, magazines and websites). We also observed a lower rate of acceptance than Wang et al. (2017), who reported 37.1% of dermatological patients would allow photos to be used on medical websites—with the critical difference that these pictures may not always be facial images. Facial images are fully identifiable, whereas dermatological photos may be of certain areas only, protecting an individual's identity more and potentially increasing the willingness of the patient to allow publication of such photos.

Scientific conferences

Respondents were most accepting of presenting their child's facial images without their eyes covered at scientific conferences than for any other scenario, supporting the earlier observation of greater trust of researchers and physicians. The use of such complete, unredacted facial images may contribute greatly to teaching and learning, since covering the eyes or large parts of the face can be limiting and of less use, particularly for dysmorphic facial features of some conditions.

Social media

While social media platforms are used widely to raise awareness related to CDs, over half of respondents (53.4%) indicated they would not want their child's facial photo shared in any form on social media. As noted previously, parents of unaffected children were less likely to share on social media (61.0% would not allow) compared with respondents with affected children (39.5% would not allow). Apart from reasons previously mentioned, reluctance to publish on social media is likely due to a strong desire to protect their children's identity from anyone who can access social media. When asked why respondents felt that way about the publication of their images, one parent indicated: 'I think unfortunately in this day and age having your child's photo everywhere and being shown around is worrying due to things like child trafficking'. This area could be explored further in future.

Other feedbacks

Parents have concerns for the consequences of publication of their children's photos on public platforms. Some respondents indicated that if images are for research purposes their publication is irrelevant. This suggests that there may be a gap in understanding and defining research and the role

of academic publication. It highlights the need for a clear explanation in the recruitment and consenting process prior to collection of facial images.

Extraction of data from facial images

The use of the extraction of landmark points from a facial photograph, or numerical data, enables the de-identification of facial images. Since the original facial image can be destroyed after landmark extraction with only the numerical data being stored, this provides substantially more privacy than actual photographs and offers greater protection of the child's identity and personal information. Surprisingly, only 46.6% of respondents indicated that they would prefer the information be stored as points, half of these felt 'Strongly' about this preference and 39.8% indicated that they would not feel differently. While the literature indicates that patients undergoing medical photography prefer that non-identifiable photographs be used for all purposes (Lau et al. 2010; Adeyemo et al. 2013; Leger et al. 2014), storing photos as numerical data limits the future value of these images.

Different rules for different patient groupings

Respondents agreed that the rules related to the collection, storage, use and publication of facial images should not differ whether the child is affected or unaffected by a CD. Although, parents' preferences for the collection and publication of their children's facial images differed depending if they had a child affected by a CD or an unaffected child, as discussed earlier. Half (51.7%) indicated that there should be a difference in the rules for children and adults due to issues related to consent, i.e. adults consent directly versus proxy consent by parents. As per ethical guidelines, the cognitive ability and capacity of the adult respondent is also a key issue.

Strengths and limitations

This study was timeously undertaken during the early implementation of the POPI Act (Republic of South Africa 2013) in South Africa and may help in developing POPI compliant guidelines reflecting people's views and preferences in the future.

Respondents to this survey were limited to those with online access only and so those without smartphones or computer internet access were excluded. This restricted study participation to individuals in higher living standards measurements (LSMs), and cannot be considered to be representative of the general population. The survey was only available in English and in none of the other 12 South African official languages (including sign language)

or official languages of other nationalities. A limitation of the study is that male and non-white respondents were underrepresented in the study, due to difficulty recruiting these respondents. That said, no significant differences were observed between all respondents' and only mother's views (see Online Resource 4), indicating that males' and females' views likely do not differ on this topic. No significant differences of the views of all respondents and only Caucasian respondents were seen (see Online Resource 4), indicating Caucasians' and non-white respondents' views likely do not differ strongly. Future studies could aim to recruit more male and non-white respondents and investigate these relationships in more detail. Finally, since the survey was compiled and distributed online, with no opportunity for clarification or assistance, this may have resulted in some respondents misunderstanding or misinterpreting a question.

Conclusion

The study suggests that parents (mostly white, female participants with a mean age of 43.7 years and a high level of education) are willing to contribute to facial research by allowing researchers to take facial images of their children. Parents of children with CDs are significantly more likely to allow researchers to collect facial images than parents of unaffected children. Parents prefer that these images are stored in secure databases inaccessible by the general public, and are generally accepting of the reuse of these images for similar research studies, under certain conditions. Preferences for publication of these facial images are not clear cut, as respondents value the privacy of their children's images but are more likely to recognise the benefits if they are personally affected (have a child with a CD). This may be addressed by developing detailed consent forms prior to the collection of images to aid the management of expectations. Consent forms may include details on the database used for storage of the images, conditions for reuse (if parents opt to give consent for reuse), platforms and photograph formats for potential publication.

Future research could investigate parental preferences of a broader demographic, and aim to set up detailed consent forms based on results from this study and that of a broader range of respondents. Reasons for publication preferences across different platforms may also be explored in future to aid in understanding.

Supplementary Information The online version contains supplementary material available at <https://doi.org/10.1007/s12687-022-00612-0>.

Acknowledgements Lize Schoeman was supported by the University of Pretoria Postgraduate Masters Research Bursary.

Author contribution Conceptualization: Lize Schoeman and Vinet Coetzee; methodology: all authors contributed; formal analysis and investigation: Lize Schoeman, Vinet Coetzee and Helen Malherbe; writing—original draft preparation: Lize Schoeman; writing—review and editing: all authors; research funding acquisition: N/A; resources: Vinet Coetzee and Helen Malherbe; supervision: Vinet Coetzee, Helen Malherbe and Engela Honey.

Data availability The datasets generated and/or analysed during the current study are available from the corresponding author on reasonable request.

Declarations

Competing interests The authors declare no competing interests.

Ethics approval This study received ethical approval from the Research Ethics Committees of the Faculty of Natural and Agricultural Sciences and the Faculty of Health Sciences at the University of Pretoria (NAS117/2021). All procedures followed were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Consent to participate All participants gave informed consent before participating by selecting ‘Yes’ to an informed consent question at the beginning of the survey.

Conflict of interest The authors declare no competing interests.

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