

# Evaluation of Psychosocial Burden on Parents of Children with Anorectal Malformation.

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## Abstract

**Aim:** The aim of our study was to assess the quality of life and psychosocial burden on parents of anorectal malformation and to compare quality of life and psychosocial burden among parents between staged and definitive group.

**Materials and Methods:** Our study was a prospective study which was carried out on 30 consecutive patients of anorectal malformations. All patients were new cases under 12 years of age. The WHO-QOL questionnaire was administered to the accompanying parent at presentation, 2 weeks, 1 month and 3 months postoperatively and Zarit Burden Scale at 2 week, 1 month and 3 month postoperatively.

**Results:** Most of our parents experienced greater psychosocial burden and their quality of life worsened following surgery. Among all domains of quality of life, social relationship was affected most followed by physical and psychological. Environmental domain was least affected. Our study also compares the psychosocial burden and quality of life of caregiver between staged and definitive group and study revealed that there was greater psychosocial burden and poor quality of life in staged surgery group.

**Conclusion:** The results of our study should be taken into account in policy making to provide better and more specific supports and interventions for this group of diseases. More attention should be given to parents (and in particular mothers) needs. Social support and different coping strategies should be developed to respond positively to individual changing needs and in buffering parents from the stress of having a child with anorectal malformation.

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**Keywords:** Anorectal malformation, WHO-QOL, Zarit burden scale, psychosocial burden.

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## Introduction

Anorectal malformations include a wide spectrum of diseases. Defects range from the very minor and easily treated with an excellent functional prognosis, to those that are complex, difficult to manage, are often associated with other anomalies, and have a poor functional prognosis [1].

The incidence of anorectal malformation is one in 2500-5000 live births. Approximately 36.4% are isolated lesions and 63.6% are associated with other anomalies [2]. The male: female ratio associated with ARM is almost equal, with a 56:44 male: female ratio [3].

The aim of surgical treatment to restore anatomy as

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normal as possible is essential, post-operative care is essential for these patients, to reach if not continence, at least a socially acceptable cleanliness [4].

Anorectal malformations are commonly associated with VACTERL anomalies. The coexistence of VACTERL anomalies negatively affects not only the surgical outcome but also the bowel functioning [5].

Thinking about the serious consequences of congenital anomalies may induce a process of parental mourning. Abandoning expectations of a healthy child, parents must prepare themselves for raising a child being severely ill, either temporarily or life-long [6].

## Materials and Methods

This study was conducted in the Department of Pediatric surgery and Department of Psychiatry, Dr RML Hospital, New Delhi. It was a prospective study of 1.5 year duration. The instruments used in this study was

**Zarit Burden Interview [7]:** The interview schedule

contained 22 items, and for each of the 22 items, caregivers were asked to respond about the impact of the patient's illness on their life, by indicating how often they felt in a particular way ("never," "rarely," "sometimes," "quite frequently," or "nearly always").

**WHOQOL-BREF** [8]: The tool consists of 26 items and is used to measure quality of life. The items were rated on a 5-point scale, which gives a profile with 4 domain scores (physical health, psychological health, social relationship, and environment) and 2 individually scored items about an individual's overall perception of quality of life and health (Q1 and Q2). The 4 domains are scaled in positive direction, with a score range of 0-100 and 4-20; and higher score denoting higher quality of life.

This study was carried out on 30 consecutive patients of anorectal malformations. All patients were new cases under 12 years of age. The WHO-QOL questionnaire was administered to the accompanying parent at presentation, 2 weeks, 1 month and 3 months postoperatively and Zarit Burden Scale at 2 weeks, 1 month and 3 months postoperatively.

Total number of cases included in our study is 30, out of which 63.3% were female and 36.7% are male, 24 patients were less than 1 year (80%) while 6 patients were greater than 1 year at presentation. In our study, 22(73.3%) patients had LOW ARM, 3(10%) patients had INTERMEDIATE ARM and 5(16.6%) patients had HIGH ARM. In our study 20 patients had isolated ARM while 10 patients had associated malformation. The most common associated malformation was cardiac malformation.

Out of 30 caregiver included in this study 29(96.7%) were mother and 1(3.3%) father. We chose mother for our study whenever possible, because in our society rearing and caring of child is carried out mainly by the mother. In 1 case we included father for our study purpose because mother was not available at the time of presentation. Most of the caregiver (43.3%) were illiterate, 46.7% undergraduate and only 10% were graduate. Out of 30, 9 (30%) caregiver were from joint family and rest were from nuclear family. Minimum age of caregiver was 19 year, maximum age was 44 year and mean age was 26.27 year. The minimum monthly income

**Table-1: Frequency Table of Patients**

		Number	Percentage
<b>Sex</b>	Male	11	63.3%
	female	19	33.7 %
<b>Age</b>	Less than 1 year	24	80 %
	1 year and above	6	20 %
<b>Type of ARM</b>	Low	22	73.3 %
	intermediate	3	10 %
	high	5	16.7 %
<b>Isolated ARM</b>		20	66.6 %
<b>Arm with Associated Malformation</b>		10	33.4 %

**Table-2: Frequency Table of Caregivers**

		Number	Percentage
<b>Relationship</b>	Mother	29	96.7 %
	Father	1	3.3 %
<b>Age (Year)</b>	Minimum	19	
	Maximum	44	
	Mean	26.27	
<b>Literacy</b>	Illiterate	13	43.3 %
	Undergraduate	14	46.7 %
	Graduate	3	10 %
<b>Type of Family</b>	Nuclear	21	70 %
	Joint	9	30 %
<b>Income (Monthly)</b>	Minimum	6000	
	Maximum	40000	
	Mean	17400	

of caregiver was 6000 INR, maximum 40000 INR and means 17400 INR. The cause of high mean monthly income in our study is 3-4 caregiver had monthly income greater than 25000 INR.

**Results**

**Postoperative Outcomes**

We performed staged surgery in 17 patients while definitive in 13 cases. Staged surgeries (divided sigmoid colostomy followed by posterior/anterior sagittal anorectoplasty followed by stoma closure) were performed in intermediate, high and those cases of low ARM in which general condition of patient was not good enough for definitive surgery.

Post operative complications are peristomal skin excoriation, stomal retraction and peritonitis, Wound dehiscence, retraction of pulled through segment and prolapse of rectal mucosa. Those complications that required surgical intervention were categorized into major complication and those complications that did not required surgical intervention were categorized into minor complication.

Out of total 13 patients, who undergone definitive surgery, 8 patients develop surgical complications. Out of 8 patients, one patient required surgical intervention, which was a case of perineal canal while rest was managed conservatively. Out of total 17 patients who underwent staged surgery, 14 patients developed surgical complications in which only 3 patient's required surgical intervention while rest were managed conservatively. Most common complication in staged group was stoma related complication that is peristomal skin excoriation

which was managed conservatively. The complications which required surgical intervention was stoma retraction there was no significant difference in complication between staged and definitive surgery.

**Functional Outcomes**

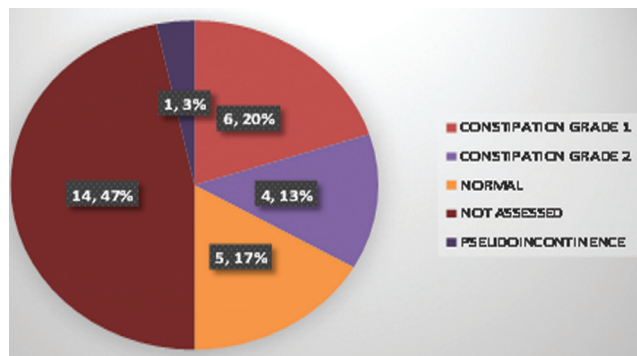


Figure 1: Functional outcomes after complete repair

The functional outcomes were evaluated in only those cases where complete repair had been done within study period. Out of 30 patients PENA'S SCORE were assessed in only 16 patients, of which 5 patients had normal bowel function while 6 patients had grade 1, 4 patients had grade 2 constipation and 1 patient had pseudo-incontinence. The patients in which bowel function was found be normal were cases of perineal canal and vestibular fistula. The function of bowel were not assessed in 14 cases as patients were on colostomy and definitive repair had not be done.

**Four Domains of Quality of Life**

The effects of postoperative outcome on parents were assessed by WHO-QOL questionnaire at admission and

**Table 3: Post operative complications**

Surgery Performed	Complication			Total
	Major	Minor	No	
Definitive	1	7	5	13
Staged	3	11	3	17
<b>Total</b>	<b>4</b>	<b>18</b>	<b>8</b>	<b>30</b>

p-value = 0.389, df=2, Chi-square value = 1.89

**Table 4: Psychosocial Burden on Parents between Staged and Definitive Group**

Zarit Burden Score	STAGED		Effect Size	DEFINITIVE		Effect Size
	Mean	SD		Mean	SD	
2 Week	52.65	9.57		36.15	13.40	
1 Month	54.24	10.92	0.16	34.15	11.36	-0.149
3 Month	52.73	12.17	-1.57	29.46	12.22	-0.41

at 2 week, 1 month and 3 month after surgery. We derived four domain scores from the WHOQOL-BREF. The four domain scores denote an individual's perception of quality of life in each particular domain. The four domain scores were converted into transformed scores on 0-100 scale using WHOQOL-BREF guidelines. Higher transformed scores on each of the domains indicate higher quality of life in that particular area. The effect size calculated between baseline and 2 weeks, 2 weeks and 1 month and 1 month and 3 months.

The mean physical health transformed score was 62.1 at time of admission (base line) which changed over 3 month period (at 2 weeks, 1 month and 3 months mean score was 48.2, 47.4 and 48.9) . The changes mainly noticed in satisfaction with sleep, daily work capacity and energy level. The effect size between base line and 2 week was -1.32, between 2 week and 1 month -0.079 and between 1month and 3 month 0.139.

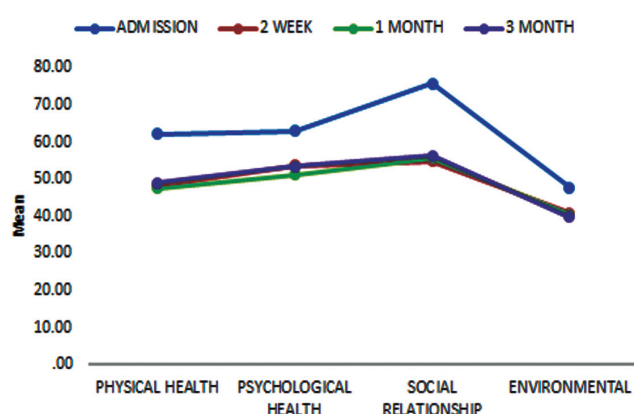


Figure 2: Four domains of quality of life

The mean transformed score of psychological health at admission was 62.8 which changes over period of 3 month (mean transformed scores at 2 weeks, 1 month and 3 months was 53.6,53.2 and 53.43 respectively). The main concern of parents was marriage of female child and future of child that created negative feeling among mothers. The effect size between base line and 2 week was -0.68, between 2 week and 1 month -0.19 and between 1 month and 3 month 0.16.

The mean transformed score of social relationship at admission was 75.63 which changes over 3 months (at 2 weeks, 1 month and 3 months was 54.8, 55.6 and 56.2 respectively). The main changes seen in support from other during this period. The effect size between base line and 2 week was -1.79, between 2 week and 1 month 0.06 and between 1month and 3 month 0.04.

The mean transformed score of environment at admission was 47.67 which changes over 3 months (at 2 weeks, 1 month and 3 months was 40.87, 40.47 and 39.8 respectively). The main changes seen in opportunity for

leisure activity and transport facility. The effect size between base line and 2 week was -0.61, between 2 week and 1 month -0.04 and between 1 month and 3 month -0.06.

### Psychosocial Burden

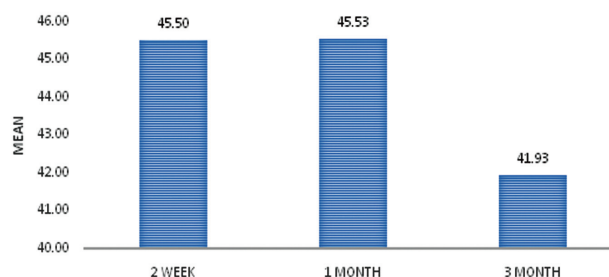


Fig. 3: Psychosocial Burden

Most parents in our study suffer moderate to severe grade of psychosocial burden. The mean burden at 2 weeks, 1 month and 3 months was found to be 45.50,45.53 and 41.93. The effect size between 2 weeks and 1 month was 0.002 and between 1month and 3 month was -0.24. Although burden was in moderate to severe grade but there was slight improvement in burden noticed at 3 month.

### Comparison Between Psychosocial Burden And Quality Of Life Between Staged And Definitive Group

Psychosocial burden was found to be moderate to severe in staged group while it is mild to moderate in the definitive group. The cause of this moderate to severe burden in staged group was stoma related complications, postoperative scar and multiple surgeries. The effect size showed improvement in the burden in definitive group vs staged group

#### Physical Health

Transformed score of physical health of parents between staged and definitive group Mean transformed score of physical health in staged surgery group was found to be 57.53, 42.47, 41.41 and 43.88 at admission, 2 weeks, 1 month and 3 month while in definitive surgery group it was 68.08, 55.69, 55.38 and 55.54 at admission, 2 weeks, 1 month and 3 month. The physical health was suffered mainly in staged group because of stoma care, frequent change of stoma bag and fecal smell that led to sleep disturbances and decreased energy level to do daily work.

#### Psychological Health

Transformed score of psychological health of parents between staged and definitive group. The mean transformed score of psychological health in definitive

surgery group at admission was 68.38, at 2 weeks 57.77, at 1 month 56.85 and 3 months 61.15. The trend showed that there was improvement in psychological health of parent after surgery while in staged group it showed continuous decrease. The cause of this continuous decrease was decreased enjoyment of life because child need more care and in case of female child the mother had negative feeling regarding marriage of child and in the male child multiple surgeries was the problem.

### **Social Relationship**

Transformed score of social relationship of parents between staged and definitive group. The social relationship of parent was seen to be in an increasing trend postoperatively in definitive surgery group while in staged group it was in a decreasing trend.

### **Environment**

Transformed score of environment of parents between staged and definitive group. The environments of parents were not affected much in staged vs definitive surgery group.

### **Discussion**

The birth of a baby brings lots of joy to the family and parents but baby with congenital malformation leads to severe impact on parents. Birth of a baby with congenital malformation that requires hospitalization and surgery affect parents in many ways not only economical but it affected many domain of life. The physical, psychological, social and environmental health of parent is affected depending upon the severity of congenital anomalies. Parents also experience burden while caring and rearing of child with congenital malformation. Parents have to deal not only with their child's disease but also need to follow the prolonged therapy schedule, which could be as distressing as the disease itself [9].

Demographic data of caregiver in our study revealed that out of 30 care giver 29 were mother and in 1 case father was caregiver. We choose mother as primary caregiver because in our society rearing and caring of child is responsibility of mother. Most of our caregiver were illiterate (43.3%) and belonged to nuclear family. The age of the caregiver ranged from 19-44 years with mean age of 26.6 year. The minimum monthly income of caregiver is 6000, maximum 40000 and mean 17400 INR, the cause of high mean monthly income in our study was 3-4 caregiver had monthly income greater than 25000 INR. Surgical advances as well as perioperative care reduced mortality approximately 10% even though (apart from CDH) this has caused much more morbidity with effects possibly extending into adulthood and placing a heavy

burden on patients & parents as well as on healthcare [10]. Among the parents, mothers may suffer more than the fathers as it has been shown that responsibility of managing the child's bowel habits and incontinence is often taken by the mother who most likely performs the regular enemas causing high level of anxiety associated with the child's ARM and the extensive follow-up treatments [11]. Parents have a crucial role to play in the life of a child suffering from anorectal malformation. Parents teaches child to learn to acclimatize to his/her disability [12]. The quality of life not only depend of physical wellness but it principally depends upon income, education and marital status.

In our study 17 cases (56.7%) underwent staged surgery (colostomy followed by posterior sagittal anorectoplasty followed by stoma closure) and 13 cases (43.3%) underwent definitive surgery (posterior sagittal anorectoplasty). Out of total 17 patients who underwent staged surgery, 14 (82%) patients developed surgical complications in which 3 patients required surgical intervention while rest were managed conservatively. Most common surgical problem were skin excoriation (64%). 2 patients (11%) out of 17 developed stoma retraction which required stoma revision. 1(5%) out of 17 developed retraction of pulled through segment. Colostomy is associated with high complication rate in our patients as reported by Chirdan LB et al [13].

8 patients (61%) out of 13 developed surgical complication in which only one patient required surgical intervention for refistulisation, rest were managed conservatively. The complication following definitive surgery in our study is similar to the study conducted by Rintala R et al [14].

In our study, only 16 patients were assessed for PENA'S SCORE for continence. 10 patients (62%) out of 16 had constipation and 5 out of 16 had normal bowel function. Our result was similar to the result of the study conducted by Huang C-F et al (2012) [15].

Psychosocial burden on parents was evaluated by the Zarit Burden Interview. Most parents in our study suffer moderate to severe grade of psychosocial burden. The mean burden at 2 weeks, 1 month and 3 months was found to be 45.50, 45.53 and 41.93. The burden in postoperative period upto 1 month found to be constant but at 3 months there was slight improvement in burden. This was due to acclimatization of caregiver to disease of child. The study conducted by GK Pruthi and A Mohta (2010) and Nissell *et al* (2009) showed mild level of burden [16-17].

In our study, we have studied the effect of immediate (short term) post-operative outcome on psychosocial

burden on parents so burden was found to be moderate to severe grade. The mean physical health domain of WHOQOL was found to decreasing trend upto 1 month from baseline but at 3 months there was slight improvement in physical health. The main reason for this decrease in postoperative period was sleepless night and frequent stoma care of child. Slight improvement was seen at 3 month which was due to better understanding of stoma care by mother and acclimatization to the condition. The mean psychological health domain of WHOQOL was found to decreasing trend upto 1 month from baseline but at 3 months there was slight improvement. The main cause of deterioration in psychological health was thinking about future of the child especially of female children and their marriage.

Similarly, social relationship and environment was affected. The main reason behind decrease in social relationship was lack of support from others especially economic support and in environment, was the lack of transport facility. Among all domains of quality of life, social relationship was affected most followed by physical and psychological. Environmental domain was least affected.

The comparison of psychosocial burden and quality of life of parents between staged and definitive group were made .Our study concluded that parents experience less burden and have better quality of life in definitive group than in staged group. The reason behind this difference is the low type of ARM, less hospital stay and good cosmetic result. In the study conducted by Pruthi (2010), it was seen that the most affected domain was psychological health and the least environmental. The cause behind this disparity of our result was due to lack of support from other especially economic in the immediate post operative period [16]. Nisell M. et al conducted a study on the psychosocial experiences of parents of children with imperforate anus (IA) and to describe their potential positive experiences. Parents of IA children and a comparison group answered a questionnaire, which was analyzed quantitatively and with manifest content analysis. Result of this was social relationships and respects for the child's will were more affected among IA mothers [17]. The results of our study (as our results corroborates with previous studies) should be taken into account in policy making to provide better and more specific supports and interventions for this group of diseases. More attention should be given to parents (and in particular mothers) needs. Social support and different coping strategies should be developed to respond positively to individual changing needs and in buffering parents from the stress of having a child with anorectal malformation.

## Conclusion

The study concluded that most of our parents experienced greater psychosocial burden and their quality of life worsened following surgery. Among all domains of quality of life, social relationship was affected most followed by physical and psychological. Environmental domain was least affected. Our study was first study which compares the psychosocial burden and quality of life of caregiver between staged and definitive group, and study revealed that there was greater psychosocial burden and poor quality of life in staged surgery group.

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<b>Ethics:</b>	There is no ethical violation as it is based on voluntary anonymous interviews
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