

Safety & Cost-Effectiveness of Primary Neonatal Posterior Sagittal Ano-recto Plasty (PNPSARP)

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Abstract

Background: Early repair of Anorectal malformation (ARM) within 6 months may be one of the factors that improve fecal continence. Delayed and multiple-stage repairs require dilatation, strict bowel preparation, fasting and total parenteral nutrition (TPN). PNPSARP requires neither bowel preparation nor parenteral nutrition. We believe it can be achieved within 72 hours of life.

Aim: To evaluate the safety, feasibility and cost effectiveness of PNPSARP within 72 hours of life versus delayed or multiple-stage repair of vestibular and perineal fistula.

Material & Methods: A retrospective study was carried out of all newborns with ARM at our institute between August 2016 and August 2019. PNPSARP within 72 hours of life was compared with delayed or multiple-stage repair. Neither bowel preparation nor parenteral nutrition was required in the PNPSARP group. Perioperative complications and costs were evaluated.

Results: Eight PNPSARP were compared with 7 delayed or multiple stage repair over the study period. Four babies (50%) were operated at day 1 post delivery in the PNPSARP group. Mean operative time (MOT) was 109 minutes (68-155). Mean follow up period was 22 months (12-36). One girl with a vestibular fistula had wound infection. This group had a good outcome with no morbidity, high satisfaction rate and low costs in comparison to delayed or multiple-stage repair.

Conclusions: PNPSARP for perineal and vestibular fistula with supportive ancillary services in the first few days of life appears to be safe and cost effective with minimal morbidity. PNPSARP is now the standard technique in our unit.

Background

Management of perineal and vestibular fistula by delayed primary repair depends on clinical circumstances with programmed anal dilatation and bowel preparation. Multiple stage repair with colostomy, PSARP and closure of colostomy is another modality. Colostomy morbidity includes bleeding, prolapse, retraction and obstruction. These two modalities carry high cost, multiple operations, multiple admissions, psychosocial impact on parents and infants plus cosmetic outcome.

The main aim of primary neonatal repair of vestibular and perineal fistula is to minimize multiple surgical trauma and to avoid the potential morbidities of colostomy¹. Early repair of ARM within 6 months of life is one of the factors that improve fecal continence^{2,3,4}. Some surgeons perform primary repair, many dilate with delayed repair, and many carry out an initial colostomy depending on their own clinical circumstances^{2,3}. Delayed repair of perineal and vestibular fistula requires pre-operative dilatation of the fistula, strict bowel preparation, fasting and TPN. We believe it could be accomplished within 72 hours of life under antibiotic cover with early post-operative oral feeding. We have been carrying out neonatal primary repair in the first few days of life for perineal and vestibular fistula since 2003 with good outcome. PNPSARP is a demanding procedure, which requires a multidisciplinary team approach. It

reduces stress on the parents, trauma to the baby, hospital admissions, number of procedures, cost and improves parent satisfaction. It also avoids a colostomy with its associated psychosocial impact.

Aim: To evaluate the safety, feasibility and cost effectiveness of PNPSARP within 72 hours of life versus delayed primary or multiple-stage repair of vestibular and perineal fistula.

Material & Methods

After ethical approval from the IRB hospital ethical committee Reference Ethical (REC 225), the parents' consented to be involved in the study. A retrospective comparative study was carried out of all newborns who underwent PNPSARP within 72 hours of life for vestibular and perineal fistula at our institute from August 2016 to August 2019.

Patients were divided into 2 groups: Group I who underwent PNPSARP in the first 72 hours of life and Group II who underwent delayed or multiple-stage repair. Urine analysis, sacral x-ray, abdominal ultrasound and echocardiography were performed shortly after birth to rule out other congenital anomalies¹. Preoperatively Gentamicin, Metronidazole and Ampicillin were given. Group I had neither bowel preparation nor TPN. Group II patients initially underwent colostomy and/or dilatation for vestibular and perineal fistula. Informed consent was taken. After endo-tracheal intubation and caudal anesthesia, Foley catheter was inserted and patient positioned prone with bottom up. All patients underwent PSARP as described by Pena. Surgical loupes and Pena stimulator were used to identify the superficial parasagittal muscle fibers and the "muscle complex" which is divided exactly at the midline in all cases². The rectum mobilized and fully separated from vagina in case of the vestibular fistula. Perineal body was reconstructed. Anoplasty was performed in the middle of the muscle complex. Glue or opsite spray was applied at suture line to avoid wound contamination.

Milk feeding commenced on the first postoperative day. The "neo-anus" was gently dilated in the third postoperative week. Laxative was not given routinely post repair. Parents were instructed for Hegar's dilatation guidelines.

Patients were followed regularly in the clinic to assess the outcome. Subjective data were collected from the outpatient clinic, chart review and patient satisfaction questionnaire. We compared both groups regarding cost effectiveness of surgical procedures, number of admissions and length of hospital stay (LOS). Quantitative measurement of outcome could be done only to measure the cost effectiveness.

Results

15 patients underwent repair of ARM during the period of the study. PNPSARP was performed for 8 patients while 7 patients underwent multiple stage or delayed primary PSARP. In group II; 3 out of 7 were girls with vestibular fistula and had multiple-stage repair. Four boys with perineal fistula had delayed repair. Seven patients (46%) were male and 8 patients (54%) were female. 7 cases (46%) had recto-vestibular fistula and 8 (54%) had perineal fistula. Six babies (40%) had associated anomalies with

cardiac and urinary systems being the most common. None of our patients has sacral deformities. In group I four babies (50%) were operated on day 1 post delivery, two (25%) on day 2, one (12.5%) on day 3 and one on day 4 (12.5%). Mean operative time (MOT) was 109 minutes (68-155). Mean follow up period was 26 months (12-36). One girl (12.5%) had superficial wound infection which was treated conservatively. All patients in Group I were breast-fed post-operatively and this was continued for a period of 3-6 months at home. Two patients had constipation, which responded to oral laxatives in one patient and enema in the second patient. Dilatation was continued in perineal fistula patients for 4-6 months and followed by a delayed PSARP. This was preceded by the administration of bowel preparation. Patients were given antibiotics, and kept NPO for two days pre-operatively, and five days post-repair with TPN. Girls with vestibular fistula in group II had a multiple stage repair (colostomy, PSARP and closure of colostomy). Group I had good outcomes with no scar (figure1). There was a high satisfaction rate amongst parents and lower cost compared to Group II ((Figure2&3). Seven out of eight parents in Group I were satisfied in comparison to one out of seven in Group II. Dissatisfaction was mainly due to a longer waiting time for the definitive surgical repair plus colostomy issues (Figure4). There was no significant statistical difference due to the small sample size.

Discussion

Management of ARM has gone through many modifications during the last three decades. PSARP is considered now the gold standard repair after well understanding of the anatomy of anal sphincter complex by Pena in 1982^{4,5,6}. ARM patients should have the best functional outcome with early efficient meticulous surgical repair plus management of associated anomalies^{7,8}.

Pena suggested primary repair of low-lying rectum after a period of regular dilatation⁵. Our series is representing only those babies who had PNPSARP for vestibular and perineal fistula in the first 72 hours of life without any preparation. PNPSARP is labeled as a demanding procedure which requires special technical skills. We agree with Nagdeve et al that it is easier to operate on a virgin neonatal plane as tapering of the rectal pouch is not required¹¹. The separation of the rectum from the hypertrophied vagina is easier due to the effect of the maternal hormones¹². PNPSARP is now our approach in all types of ARM except recto-vesical fistula as it saves the patient 1-2 operations and 2-3 admissions. We gained the technical skills required for PNPSARP by developing experience over time and regularly auditing our work. We use surgical loupes for easier dissection.

Some surgeons do primary repair, many dilate and delay the repair, and many do an initial colostomy depending on their own clinical circumstances^{2,3}. Delayed repair of ARM requires dilatation, strict bowel preparation, fasting and TPN. Financial advantage and psycho-social resistance of colostomy popularized primary repair over the staged repair especially in developing countries^{9,10}.

The driving forces for PNPSARP are parents and babies stress, multiple anesthesia, multiple admissions and high cost. Financial advantage and psycho-social resistance of colostomy popularized PNPSARP over the staged repair especially in developing countries and further multi centers studies are required to

support it, Although stoma diversion decreases the risk of infection and dehiscence in PSARP^{2,6,13}, it carries a risk of morbidities¹³. Only one girl with a vestibular fistula had wound infection in group I. The infection was superficial and treated conservatively without any consequence. She had straightforward PSARP, but unfortunately she was delivered elsewhere and started feeding before transfer to our institute. We have since modified our policy to exclude those who had commenced feeding.

Meticulous PNPSARP appears to provide the “best utilization of all existing resources” and might result in a better continence^{17,18}, although some authors disagreed¹⁹. Constipation was encountered in 60% of published series²⁰. In our study, content cannot be evaluated due to the short follow up period. The good outcome of PNPSARP is attributed to the accumulation of experience and the comprehensive ancillary services. We think that we must keep moving forward to operate our babies as early as possible, and hopefully without colostomy even for high lying rectum^{8,11,18}, provided we evaluate our cases in an objective way, apply a strict auto-criticism and long term follow up. We found that PNPSARP can be done safely by our trainees under supervision of experienced surgeons with supportive ancillary services. We suggest adding this procedure to the pediatric surgery training curriculum as it is not more challenging than tracheo-esophageal fistula repair. For patient safety the surgeon must take into consideration, his experience, the medical status of the patient and the surrounding circumstances on making a decision for PNPSARP. Our results suggest that PNPSARP is safe and cost effective.

Conclusions

PNPSARP for perineal and vestibular fistula is safe with minimal morbidity, good parent satisfaction and low cost provided that the supportive ancillary services are available. PNPSARP is now the standard technique in our unit except for bladder neck fistula, and we are recommending it as an excellent approach for ARM repair. Longer follow up for fecal and urinary incontinence is required to complement our findings.

Declarations

Ethics approval and consent to participate

Conflict of Interest Statement

Author 1, declares that she has no conflict of interest.

Author 2, declares that he has no conflict of interest

Author 3, declares that he has no conflict of interest

Author 4, declares that he has no conflict of interest

Author 5, declares that he has no conflict of interest

Author 6, declares that he has no conflict of interest

Author 7, declares that she has no conflict of interest

Statement of Ethics:

All procedures performed in studies 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 and approved by KFAFH IRB.

Author Contributions

Corresponding Author:

Drafting the work

Final approval of the version to be published

Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy; or the acquisition,; and revising it critically for important intellectual content;

co-author 1:

Substantial contributions to the conception or design of the work

co-author2

Substantial contributions to the conception or design of the work

co-author3:

Integrity of any part of the work are appropriately investigated and resolved

co-author4

Integrity of any part of the work are appropriately investigated and resolved

co-author 5:

Review, drafting, and final approval of the manuscript.

co-author 6:

Analysis, or interpretation of data for the work

co-author 7:

Analysis, or interpretation of data for the work

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Levels of Evidence: IV

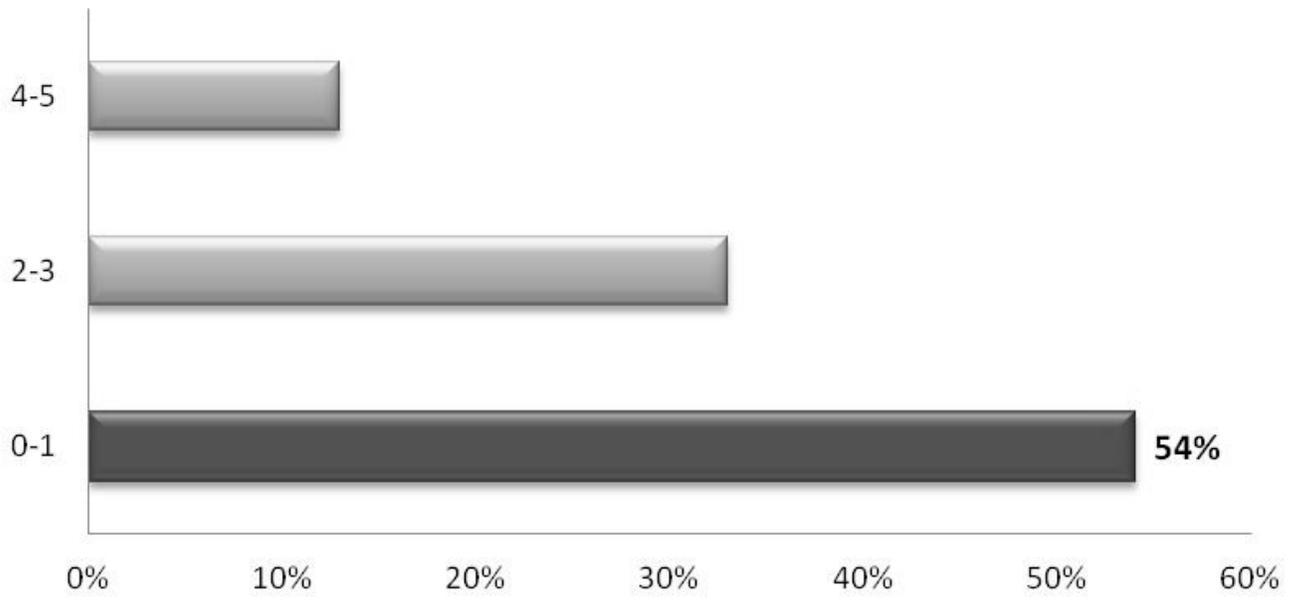
References

1. Alexander Holschneider, John Hutson, Albert Peña, Elhamy Beket, Subir Chatterjee, Arnold Coran, Michael Davies, Keith Georgeson, Jay Grosfeld, Devendra Gupta, Naomi Iwai, Dieter Kluth, Giuseppe Martucciello, Samuel Moore, Risto Rintala, E Durham Smith, D V Sripathi, Douglas Stephens, Sudipta Sen, Benno Ure, Sabine Grasshoff, Thomas Boemers, Feilin Murphy, Yunus Söylet, Martin Dübbers, Marc Kunst. (2005) Preliminary report on the International Conference for the Development of Standards for the Treatment of Anorectal Malformations. *J Pediatr Surg* 40, 1521–1526.
2. Pen~a A, De Vries P. (1982) Posterior sagittal anorectoplasty: important technical considerations and new applications. *J Pediatr Surg*; 17:796- 811.
3. Pena A. *Surgical Management of Anorectal Malformations*. New York: Springer Verlag; 1989. p. 15
4. Adeniran JO, Abdur-Rahman L(2005). One-stage correction of intermediate imperforate anus in males. *Pediatr Surg Int.*;21(2):88-90. Epub 2005 Jan 4.
5. Chen CJ. The treatment of imperforate anus: experience with 108 patients. *J Pediatr Surg*. 1999; 34(11):1728-32.
6. Bischoff A¹, Levitt MA, Peña A. (2013). Update on the management of anorectal malformations. *Pediatr Surg Int*. Sep;29(9):899-904.
7. A N Gangopadhyay, S Chooramani Gopal, Shilpa Sharma, D K Gupta, S P Sharma, T Vittal Mohan. (2006). Management of anorectal malformations in Varanasi, India: a long-term review of single and three stage procedures. *Pediatr Surg Int.*; 22(2):169-72. Epub 2005 Nov 29.
8. C T Albanese, R W Jennings, J B Lopoo, B J Bratton, M R Harrison. (1999). One-stage correction of high imperforate anus in the male neonate. *J Pediatr Surg*; 34:834-836.

9. Basant Kumar, Deepak K Kandpal, Shyam B Sharma, Leela Dhar Agrawal, Virendra Narayan Jhamariya. (2008). Single-stage repair of vestibular and perineal fistulae without colostomy. *J Pediatr Surg.* Oct; 43(10):1848-52.
10. Mishra BN, Narasimhan KL, Chowdhary SK, Samujh R, Rao KL. (2000). Neonatal PSARP versus staged PSARP: A comparative analysis. *J Indian Assoc Pediatr Surg.*;5:10–3.
11. Nilesh G. Nagdeve, Pravin D. Bhingare, and Harish R. Naik. (2011). Neonatal posterior sagittal anorectoplasty for a subset of males with high anorectal malformations. *J Indian Assoc Pediatr Surg.* Oct-Dec; 16(4): 126–128.
12. Max Elstein M.B., Ch.B. (1963). *Vaginal Cytology Of The Newborn.* *Bjog.* December, volume 70, Issue 6, Pages 1050-1055
13. N Patwardhan, E M Kiely, D P Drake, L Spitz, A Pierro. (2001). Colostomy for anorectal anomalies: high incidence of complications. *J Pediatr Surg.*; 36(5):795-8.
14. Goon H. (1990). Repair of anorectal anomalies in the neonatal period. *Pediatr Surg Int.*; 5: 246–249.
15. M Endo, A Hayashi, M Ishihara, M Maie, A Nagasaki, T Nishi, M Saeki. (1999). Analysis of 1,992 patients with anorectal malformations over the past two decades in Japan. Steering Committee of Japanese Study Group of Anorectal Anomalies. *J Pediatr Surg.*; 34(3):435-41.
16. Guochang Liu, Jiyan Yuan, Jinmei Geng, Chunhua Wang, Tuanguang Li. (2004). The Treatment of high and intermediate anorectal malformations: one stage or three procedures. *The J Pediatr Surg.*;39(10):1466-71.
17. Nah SA¹, Ong CC, Lakshmi NK, Yap TL, Jacobsen AS, Low Y. (2012). Anomalies associated with anorectal malformations according to the Krickenbeck anatomic classification. *J Pediatr Surg.* Dec;47(12):2273-8.
18. Mirshemirani, J. Kouranloo, M. Rouzrokh, M. N. Sadeghiyan and A. Khaleghnejad. (2007). Primary Posterior Sagittal Anorectoplasty Without Colostomy In Neonates With High Imperforate Anus. *Acta Medica Iranica*, Vol. 45, No.2 .
19. Mishra BN, Narasimhan KL, Chowdhary SK, Samujh R, Rao KL. (2000). Neonatal PSARP versus staged PSARP: A comparative analysis. *J Indian Assoc Pediatr Surg.*;5:10–3.
20. Rintala RJ, Lindahl HG. (2001). [Fecal continence in patients having undergone posterior sagittal anorectoplasty procedure for a high anorectal malformation improves at adolescence, as constipation disappears.](#) *J Pediatr Surg.* Aug;36(8):1218-2

Figures

Stony Brook Scar Scale

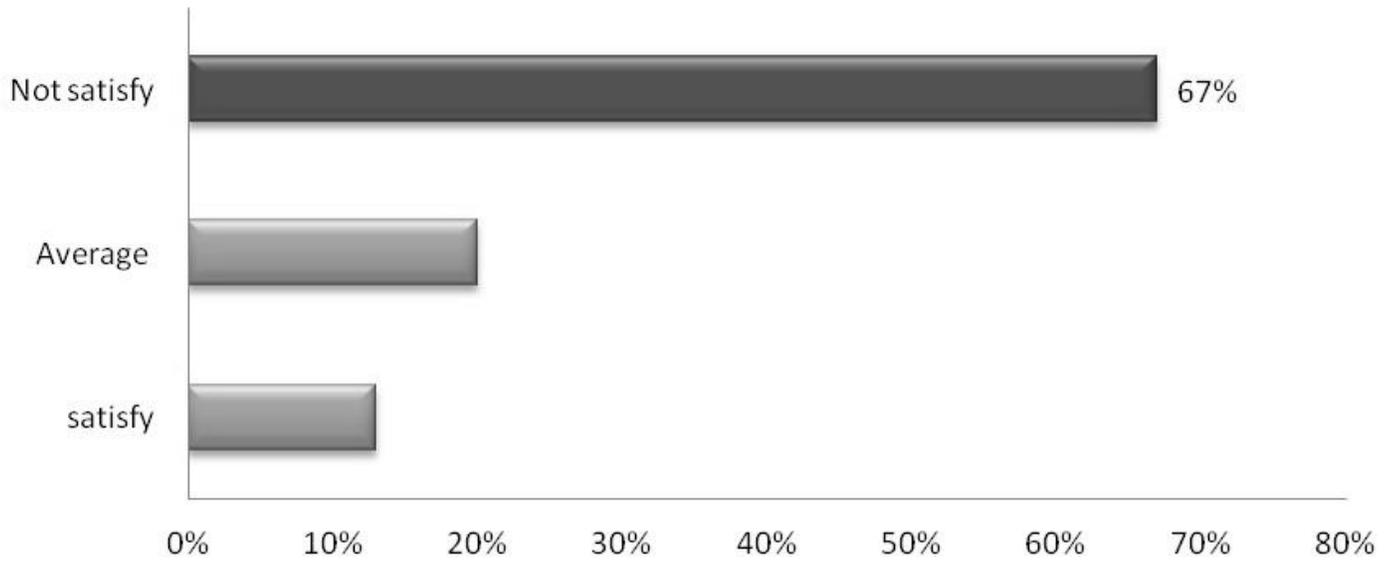


	0-1	2-3	4-5
Percentage	54%	33%	13%

Figure 1

Stony Brooke Scar Scale

Overall Parents Satisfaction



	satisfy	Average	Not satisfy
■ Percentage	13%	20%	67%

Figure 2

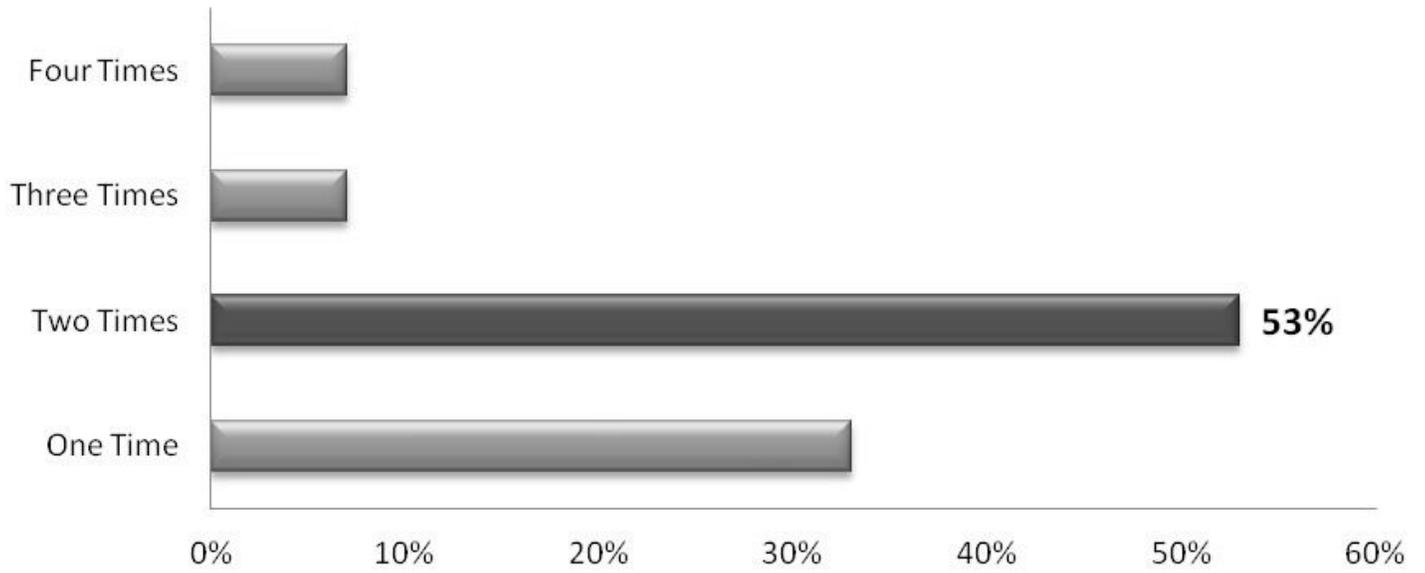
Over All Parents' satisfaction

Operation		Site	Procedure cost	Admission cost	Appliance	Total
3 Stages	Colostomy diversion	NICU	12000 SR	$5_{\text{days}} \times 3000_{\text{SR}} = 15000 \text{ SR}$	$33_{\text{SR}} \times 60_{\text{peice/month}} \times 6_{\text{month}} = 11880 \text{ SR}$	38000 SR
	PSARP	Pediatric	20000 SR	$5_{\text{days}} \times 1000_{\text{SR}} = 5000 \text{ SR}$		25000 SR
	Colostomy closure	Pediatric	9500 SR	$5_{\text{days}} \times 1000_{\text{SR}} = 5000 \text{ SR}$		14500 SR
	Total					77500 SR
Single stage		NICU	20000 SR	$5_{\text{days}} \times 3000_{\text{SR}} = 15000 \text{ SR}$		35000 SR

Figure 3

Total Costs for Both Groups

Times of Surgeries Postpone



	One Time	Two Times	Three Times	Four Times
■ Percentage	33%	53%	7%	7%

Figure 4

Postponing Definite Surgery