MAGNETIC RESONANCE EVALUATION OF ANORECTAL MALFORMATION CASES TREATED BY POSTERIOR SAGITTAL ANORECTOPLASTY

Dipu Bhuyan¹, Sushant Agarwal², Pynskhemboklang Khongsi³, Aswin Padmanabhan⁴

HOW TO CITE THIS ARTICLE:

Dipu Bhuyan, Sushant Agarwal, Pynskhemboklang Khongsi, Aswin Padmanabhan. "Magnetic Resonance Evaluation of Male Anorectal Malformation Cases Treated By Posterior Sagittal Anorectoplasty". Journal of Evolution of Medical and Dental Sciences 2015; Vol. 4, Issue 23, March 19; Page: 3995-4007, DOI: 10.14260/jemds/2015/575

ABSTRACT: OBJECTIVES: Pelvic MRI is a useful tool for assessing patients with anorectal malformations before and after operation. The images obtained after PSARP can be reviewed for quality and shape of the sphincter muscle, position of the rectum, shape of the sacrum, and associated pelvic abnormalities related to the initial operation. These were then correlated with the clinical status of the patient during follow-up after colostomy closure. MATERIALS AND METHODS: Twenty six male patients with intermediate or high anorectal malformation were included in the study between January 2012 to December 2013. The findings thus obtained in the MRI were correlated clinically with the Kelley's scoring system. **RESULTS:** A total of 26 post-PSARP patients were included in the study. Eighteen of them were also selected for clinical correlation after colostomy closure. According to MRI findings, 2 patients had good degree of development of the pelvic floor muscles, 11 of them had fair amount of muscles and the rest 13 of them had significantly thinned out muscles. Four patients showed well-developed external sphincter muscle, 15 of them were having fair degree of development and in the rest 7 of them it was poorly-developed. Our study also showed that only 3 patients were showing symmetrical development of the sphincter complex whereas in the rest 23 of them it was asymmetrical. Eighteen patients had centrally-placed pulled-through rectum, whereas in the rest 8 of them the colon was located away from the center of the sphincter complex. Sixteen patients had mesenteric fat inadvertently pulled along with the bowel during PSARP; and in this same study, 58% of our patients were also having associated anomalies detected by MRI. On clinical correlation, it was found that external sphincter muscle and fat interposition play an important role in the overall clinical status of the patients after colostomy closure. **CONCLUSION:** Pelvic MRI is a useful tool for assessment of anorectal malformation after PSARP. Our study shows that development of the external sphincter muscle and the presence of mesenteric fat interposition has significant impact on the overall clinical status of the patient; however, other factors like operative technique and muscle innervations may also play important role in the overall continence of the patients. **KEYWORDS:** Anorectal malformation, MRI.

ABBREVIATIONS:

- ARM Anorectal malformation. MRI - Magnetic resonance imaging.
- CT Computerized Tomography.

INTRODUCTION: Anorectal malformations encompass a diverse group of congenital malformations of the anorectum and they are frequently associated with other anomalies. Incidence is one in 5000 livebirths.¹

The goals of surgical correction are to promote anatomic reconstruction, establish socially acceptable bowel function, and avoid undesirable sequelae such as fecal incontinence, urinary incontinence, and sexual dysfunction. However, with much advancement in the management of ARM, still many patients come back with many functional and technical complications.

Pelvic MRI is a useful tool for assessment of ARM before and after the initial repair. It has advantages over CT because of its multiplanar imaging facility and lack of ionizing radiation for infants and small children.^{2,3} It is superior to CT in identifying the sphincteric muscles due to its excellent soft tissue characterization.⁴

METHODS: A prospective study of twenty six male patients with high or intermediate type ARM and who had already undergone PSARP were included in the study between January 2012 to December 2013. Informed consent was taken from the parents of all the patients and permission for conducting the study was taken from the Institutional Ethical Committee. Exclusion criteria are: male patients with low ARM, female patients, and patients in whom PSARP have not been done. All these patients were operated upon by 5 different surgeons; 4 of them used the conventional technique of PSARP as advocated by Pena, whereas one surgeon did some modification by not cutting the vertical muscle complex and anoplasty was done with moderate tension, thereby allowing a skin-lined anal canal. All the patients underwent MRI study to evaluate their pelvic musculature. Location of the pulled-though rectum, and fat interposition were noted, and any associated anomalies which may directly or indirectly affect the outcome were also included. The MRI images were interpreted by a single radiologist without any knowledge of the clinical information. Eighteen patients had also completed colostomy closure and they were on follow-up; these were the patients selected for clinical correlation with the MRI findings, using the Kelley's Clinical Scoring system. The Chi-square test was used for statistical analysis of our findings.

RESULTS: Age of the patients ranged from 4 months to 16 years. Twenty four patients had high type whereas the other 2 of them had intermediate type of anomalies. According to MRI analysis of all these patients, 2 demonstrated good degree of development of the pelvic floor muscles; other 11 of them had fair amount of muscles, and the rest 13 had significantly thinned out or poorly developed muscles. Clinically, however, there was no significant change statistically. On the other hand, there was significant overlap of patients with fair or lax pelvic floor muscles with those in group 1 (Clinically good) and 2 (Clinically fair). Four patients were showing well-developed external sphincter; another 15 of them were having fair degree of development, whereas in the rest 7 of them the development of the external sphincter had a significant impact (p=0. 027) on the clinical outcome of the patients during follow-up. In this same study, only 3 patients were showing symmetry of the sphincter complex, whereas in the rest 23 of them MRI showed asymmetrical development of this muscle.

However, this had no significant relevance on the clinical outcome of the patients. Eighteen patients had centrally-placed pulled-through rectum, whereas in 8 of them the colon was located away from the center of the sphincter complex. This also was found to have no significant effect on the clinical picture of the patients overall. Sixteen patients had mesenteric fat pulled along with the bowel during PSARP, whereas in 10 of them there was no inadvertently pulled mesenteric fat

detected on MRI. On statistical analysis it appeared that the presence of mesenteric fat pulled with bowel through the sphincter during the initial repair interfered with the continence mechanism of the patient later on during follow-up after colostomy closure (p = 0.042). In addition, MRI could also detect that 58% of the patients with ARM were also having other associated anomalies, and genitourinary anomalies alone were seen in 53% of these cases. Four patients (26. 6%) were also found to have abnormalities of the spine and spinal cord.

STATISTICS:

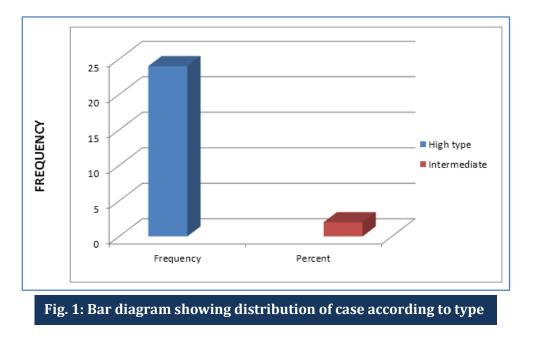
Type of ARM

Vaild	26
Missing	0

Type of AMR

	Frequency	Percent	Vaild Percent	Cumulative						
High	24	92.3	92.3	92.3						
Intermediate	2	707	7.7	100.0						
Total	Total 26 100.0 100.0									
Table 1: Showing distribution of case according to type										

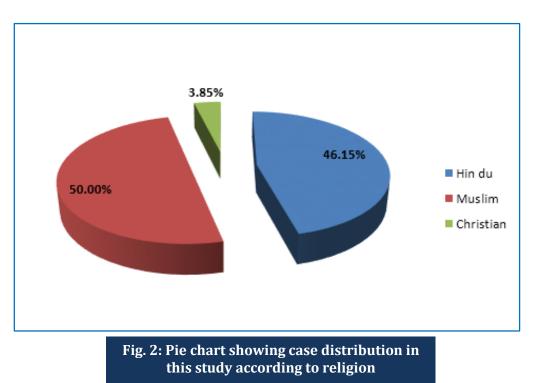
TYPE OF AARM



RELIGION: In the present Study, Out of the 26 case that ware including for our study 12 (46. 2%) ware from Hindu community, the other 13 case (50%) belonged to a Muslim community, and the rest i. e. 1 case (3. 8%) ware a Christian.

	Religion											
	Frequency Percent Vaild Percent Cumulative Percent											
	Hindu	12	46.2	46.2	46.2							
Vaild	Muslim	13	50.0	50.0	96.2							
Vallu	Christian	1	3.8	3.8	100.0							
	Total	26	100.0	100.0								
	Table 2: Showing distribution of according to religion											

Effect of development of pelvic floor muscle complex in relation to clinical findings of the patients.



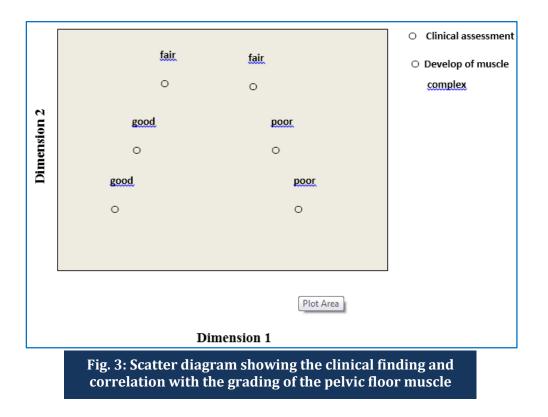
Development of	Clinical assessment								
pelvic floor muscle	Good	Fair	Poor	Active Margin					
Good	2	0	0	2					
Fair	5	4	0	9					
Poor	1	4	2	7					
Active Margin	8	8	2	18					
Table 3: Showing correlation of pelvic floor muscle development with clinical status of the patients									

SUMMARY:

	Circuralism		ch i		-	ortion ertia	Confidenc Va	e Singular lue
Dimension	Singular value	Inertia	Chi- square	p- value	Accounted for	Cumulative	Standard Deviation	Correlation 2
1	. 604	. 635	7.357	. 118 a	. 894	. 894	. 140	171
2	. 208	. 043			. 106	1.000	. 125	
Total		. 409			1.000	1.000		

a. 4 degrees of freedom.

Row and Column Points. Symmetrical Normalization.



Development of the external sphincter and its effect on the clinical finding of the patients: Correspondence Table:

Development of ext. sphincter	Good	Fair	Poor	Active Margin
Good	3	0	0	3
Fair	5	4	0	9

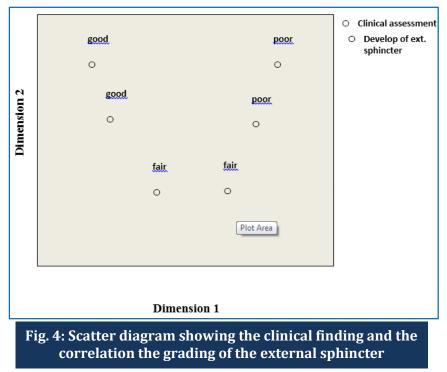
J of Evolution of Med and Dent Sci/eISSN-2278-4802, pISSN-2278-4748/Vol. 4/Issue 23/Mar 19, 2015 Page 3999

Poor	0	4	2	6					
Active	8	8	2	18					
Table 4: Showing relation of the development of external sphincter with the clinical statues of the patients									

SUMMARY:

Dimension	Singular value	Inertia	Chi- square	p- value	Proportion Inertia			e Singular lue
					Accounted Cumulative for		Standard Deviation	Correlation
								2
1 2 Total	. 749 . 222	. 562 . 049 . 611	11.000	027	. 919 . 081 1. 000	. 919 1. 000 1. 000	. 072 . 125	102

a. 4 degrees of freedom
Chi-square – 11. 00
Degree of freedom – 4.
p- value – 0. 027.
Row and Column Points.
Symmetrical Normalization.



The impact of the symmetry of the sphincter complex on the clinical picture of the patients: Correspondence Table.

J of Evolution of Med and Dent Sci/eISSN-2278-4802, pISSN-2278-4748/Vol. 4/Issue 23/Mar 19, 2015 Page 4000

Symmetry of sphincter complex	Clinical assessment					
Symmetry of sphincter complex	Good	Fair	Poor	Active margin		
Symmetric	1	2	0	3		
Asymmetric	7	6	2	15		
Active Margin	8	8	2	18		
Table 5: Symmetry of sphincter complex and its relation with clinical assessment						

SUMMARY:

Dimension	Singular Value	Intria	Chi- value	p- value	Proportion of Inertia		Confidence Singular Value
					Accounted for	Cumulative	Standard deviation
1 Total	. 224	. 050 . 050	. 900	. 638a	1.000 1.000	1.000 1.000	. 179

a. 2 degrees of freedom.
Chi – square of freedom = 2.
Degree of freedom = 2.
p- value = 0. 638.

Position of the pulled -through rectum and its clinical relationship. Correspondence Table.

Position of pulled- through rectum	Clinical assessment					
Fosition of punea- through rectain	Good	Fair	Poor	Active margin		
Central	7	4	1	12		
Eccentric	1	4	1	6		
Active Margin	8	8	2	18		
Table. 6: position of the pull-though rectum and correlation with the clinical assessment						

SUMMARY:

Dimension	Singular	Intria	Chi-	p- value	Proportion of Inertia		Confidence Singular Value
Dimension	Value	muna	value		Accounted for	Cumulative	Standard deviation
1	. 395	. 156			1.000	1.000	. 199
Total		. 156	2.813	. 245ª	1.000	1.000	

a. 2 degrees of freedom.

Chi –square test = 2.813 Degree of freedom = 2

p- value = 0245

Presence or absence of the mesenteric fat its effect on the clinical status of the patients after definitive surgery:

Correspondence Table:

Mesenteric fat	Clinical assessment								
Mesentericiat	Good	Fair	Poor	Active margin					
Present	2	7	1	10					
Absent	6	1	1	8					
Active Margin	8	8	2	18					
Table 7: presence of mesenteric fat its relation with the clinical outcome Summary									

Dimension	Singular Value	Intria	Chi- value	p- value	Proportion of Inertia		Confidence Singular Value
					Accounted for	Cumulative	Standard deviation
1 Total	. 594	. 353 . 353	6.356	. 042ª	1.000 1.000	1.000 1.000	181

a. 2 degree of freedom.
Chi –square test -6. 356.
Degree of freedom = 2.
p- value = 0. 042.

Other incidental finding detected by MRI in our anorectal malformation case:

Case 1: Testes in inguinal region on both sides.

Case 2: Diverticulum on the posterior inferior aspect of the bladder wall.

Small nodules on the left pararectal, ischiorectal, and ischoanal fossa.

Case 5: Posterior urethral diverticulum. Filar lipoma.

Case 7: Lower segment of coccyx and sacrum are not formed.

Case 9: Nodular lesions in left pararectal, ischioanal, and ischiorectal fossa.

Case 10: Nodular lesion in the left pararectal, ischionanal, and ischiorectal fossa.

Case11: Testes in inguinal region bilaterally.

Case14: Utricle cyst.

Case15: Nodular lesions in left pararectal ischiorectal, and ischioanal fossa.

Case 18: Nodular lesion in left pararectal, ischiorectal, ischioanal fossa.

Case 20: Right testis in inguinal region.

Nodular lesion in left pararectal, ischiorectal and ishioanal fossa.

Case 21: L3 hemivertebra.

Case 22: Left testis in inguinal region.Case 23: Left hydrouretronephosis.Case 24: S4, S5, and coccyx are not visualized.

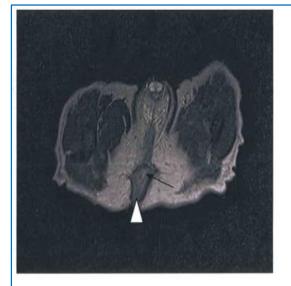




Fig.1 (a) – 2month old baby with normal internal (arrow) and external sphincter (arrow head)

(b) – same patient with well-developed levator ani

Fig. 1(a) (b)

Prominent pararectal, internal iliac, eternal, iliac, and bilateral inguinal nodes. Prominent pararectal, internal iliac, external iliac, and bilateral inguinal nodes.



(c) – well-developed pelvic floor muscles like the shape of an inverted umbrella





external sphincter(thicker on left side)



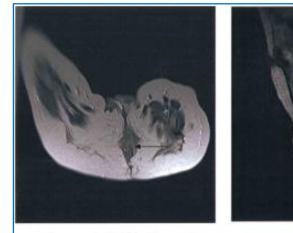
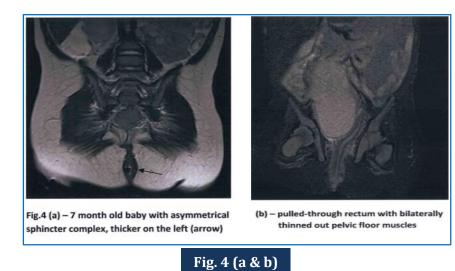


Fig.3 (a) – 9 month old child with centrallyplaced rectum and visible mesenteric fat (arrow)

(b) – pelvic floor muscles are thinned out (arrow)





DISCUSSION: Bowel control implies the ability to detect and retain flatus and stool until the appropriate time for evacuation. It is the result of complex interplay among sphincter function, anorectal sensation, and colonic motility. All these factors are affected in children with anorectal malformations.

The pelvic floor is a complex, integrated, multilayer system that provides active and passive support. Fascia and ligaments provide passive support, while the muscles of the pelvic floor, mainly the levator ani, provide active support. The pubo rectalis muscle has been considered the essential muscle for continence, and adequate placement of the neorectum in the puborectalis sling has been emphasized in the surgical literature.⁵ Recently, the important contribution of the external anal sphincter for continence, has been stressed.^{6,7} Adequate placement of the neorectum not only through the puborectalis sling but also within the external sphincter is necessary for an adequate functional outcome after surgical correction of anorectal anomalies.⁴

Several reports have assessed the efficacy of CT and MRI in evaluating anorectal malformation.^{8,9,10,11} MRI with axial and coronal imaging has allowed direct visualization of the sphincter muscle complex with excellent anatomic detail due to its excellent soft tissue characterization. MRI has other advantages over CT because of its multiplanar imaging facility and lack of ionizing radiation for infants and small children.

Some previous studies have used the measurement of the thickness of the sphincteric muscles. However, muscle thickness changes according to the patient's age.^{10,11} Fukuya T et al⁴ evaluated the role of muscle development for continence. Their results showed significant overlap between clinically continent and incontinent patients; and they concluded that grading based solely on the degree of the development of muscles can thus be misleading unless there are markedly hypoplastic muscles.

The present study indicates that there was no significant difference in development of the pelvic floor muscles on clinical correlation with patients in group 1 and 2, with most of them were either fairly or poorly-developed, and only 2 of the patients in group 1 had well-developed muscles. Group3 patients however, were both showing poorly-developed muscles on MRI.

In the study by Shah AA et al,¹² in patients with high anomaly, the external sphincter development was better than development of the levator and sphincter muscle complex. Therefore, with proper placement of rectum, many of them were able to achieve acceptable continence, thus emphasizing that even patients with high malformation can have good external sphincter with good prospects of continence. Similarly, our study also supports this finding and showed that external sphincter has got significant contribution on the clinical outcome of the patients in terms of continence.

Malpositioning of the rectal pull-through can be identified on axial, coronal, and sagittal images of the MRI scan. The importance of siting the pulled-through rectum accurately and symmetrically in the sphincter ani at primary operation is stressed, as fecal incontinence can occur with asymmetrical placement. In contrast to many studies, ¹³ our finding showed 33. 3% of the patients had their pulled-through rectum sited away from the center, but analysis showed that this does not have significant impact on the outcome of the patients clinically. Presence of peritoneal fat inadvertently pulled along with the rectum was shown to play significant contribution on the continence mechanism of the patients after PSARP. This finding is also supported by other studies done by Yong C et al and others.^{12,14}

However, there are several factors that may be related to continence: passive forces; motor action of the sphincter; sensitivity of the skin, mucosa, and sphincter; function of the intestine; and nervous system pathways such as innervation of the muscles. These factors and in fact, the technique of operation have got significant contribution on the fecal continence of the patients. This statement supports our findings on some of our patients in whom modified technique of PSARP was done. In these cases, the vertical muscle complex was not cut and pull-through of the bowel was done with moderate tension; hence there was less disturbance to the muscle innervations and the sphincteric complex was not divided, and anoplasty was done under some tension, therefore allowing a skinlined anal canal. Most of these patients are showing better clinical outcome. This finding however, will need better randomization with more number of cases and a longer follow-up to support our observations.

CONCLUSION: Pelvic MRI is a useful tool for assessment of anorectal malformation before and after PSARP. It has several advantages because of its multiplanar imaging facility and lack of ionizing radiation especially for small infants and small children. Because of its excellent soft tissue characterization, it allows identification of the sphincteric muscles accurately. This study indicates that there was significant overlap in the clinical outcome of the patients when correlated with the degree of development of the pelvic floor muscles. Similarly, the location of the pulled-through rectum within the levator ani sling and the symmetry of the sphincteric complex does not appear to play significant role on the outcome. However, the development of the external sphincter and the presence of inadvertently pulled mesenteric fat appears to have significant contribution on the overall clinical outcome of the patients. Our observation that some of our patients who underwent modified PSARP showed better outcome may be attributed to other factors affecting continence such as the technique of operation and muscle innervations in the already compromised and maldeveloped pelvic musculature.

REFERENCES:

- 1. Pena A, Levitt MA. Anorectal malformations. In: Grosfeld JL, O'Neill JA Jr, Fonkalsrud EW, Coran AG, eds. Pediatric Surgery. Philadelphia, PA: Mosby, Elsevier, 2006: 1566-1589.
- 2. Vade A, Reyes H, Wilbur A, Gyi B, Spigos D. The anorectal sphincter after pull-through for anorectal anomalies. Pediatr Radiol, 1989; 19: 179-183.
- 3. Sato Y, Pringle KC, Bergman RA, et al. congenital anorectal anomalies: MR imaging. Radiology, 1988; 168: 157-162.
- 4. Fukuya T, Honda H, Kubota M, et al. Post operarive MRI evaluation of anorectal malformations with clinical correlation. Pediatr Radiol, 1998; 23: 583-586.
- Stephens FD, Smith ED. Anatomy and function of the normal rectum and anus. In: Stephens FD, Smith ED, eds. Ano-rectal malformations in children. Chicago: Year Book Medical Publishers, 1971: 14.
- 6. DeVries PA, Cox KL. Surgeries of anorectal anomalies. Surg Clin North Am, 1985; 65: 1139.
- 7. Pena A. posterior sagittal approach for the correction of anorectal malformations. In: Mannick JA, Cameron JL, Jordan GL Jr, et al (eds). Advances in surgery. Year Book, Chicago, p 69.
- 8. Kohda E, Fujioka M, Ikawa H, Yokoyama J. Congenital anorectal anomaly: CT evaluation, 1985; 157: 349.

J of Evolution of Med and Dent Sci/eISSN-2278-4802, pISSN-2278-4748/Vol. 4/Issue 23/Mar 19, 2015 Page 4006

- 9. Taccone A, Martucciello G, Dodero P et al. New concepts in preoperative imaging of anorectal malformations. Pediatr Radiol, 1992; 22: 196-199.
- 10. Arnbjornsson E, Laurin S, Mikaelsson C. Computed tomography of anorectal anomalies: correlation between radiologic findings and clinical evaluation of fecal incontinence. Acta Radiol, 1989; 30: 25.
- 11. Sato Y, Pringle KC, Bergman RA, et al. congenital anorectal anomalies: MR imaging. Radiology, 1988; 168: 157-162.
- 12. Shah AA, Kothari MR, Bhattacharjee N, Shah AJ, Shah AV. Magnetic resonance imaging in anorectal malformations. J Indian Assoc Pediatr Surg, 2001; 6: 4-13.
- 13. Eltomey MA, Donnelly LF, Emery KH, Levitt MA, Pena A. Postoperative pelvic MRI of anorectal malformations. AJR, 2008; 191: 1469-1476.
- 14. Yong C, Ruo-Yi W, Yuan Z, Shu-Hui Z, Guang-Rui S. MRI findings in patients with defecatory dysfunction after surgical correction of anorectal malformation. J Pediatr Radiol, 2013; 43 (8): 964-970.

AUTHORS:

- 1. Dipu Bhuyan
- 2. Sushant Agarwal
- 3. Pynskhemboklang Khongsi
- 4. Aswin Padmanabhan

PARTICULARS OF CONTRIBUTORS:

- 1. Associate Professor, Department of Radiology, Gauhati Medical College & Hospital, Guwahati.
- 2. Post Graduate Student, Department of Radiology, Gauhati Medical College & Hospital, Guwahati.
- 3. Senior Resident, Department of Paediatrics Surgery, Gauhati Medical College & Hospital, Guwahati.

FINANCIAL OR OTHER COMPETING INTERESTS: None

 Post Graduate Student, Department of Radiology, Gauhati Medical College & Hospital, Guwahati.

NAME ADDRESS EMAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Dipu Bhuyan, Associate Professor, Department of Radiology, Gauhati Medical College & Hospital, Guwahati-32. E-mail: dipubhuyan@gmail.com

> Date of Submission: 05/02/2015. Date of Peer Review: 06/02/2015. Date of Acceptance: 09/03/2015. Date of Publishing: 18/03/2015.