

Original Article

Frequency of Nail Abnormalities in Psoriasis and its Association with Disease Severity

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Abstract

Background: Psoriasis is a chronic inflammatory skin disease. Clinically, disease can present with cutaneous, joints and nails lesions. Nail abnormalities can be seen in up to two-thirds of patients with psoriasis.

Objective: to find out the frequency of nail abnormalities in psoriasis and its association with severity of psoriasis.

Methods: This cross-sectional analytical study was conducted in the Department of Dermatology and Venereology, Sylhet MAG Osmani Medical College Hospital from January 2017 to December 2019. One hundred and forty patients with psoriasis were included.

Results: Nail abnormalities were found in 63.6% (n=87) of psoriatic. Nail pitting was the most common, followed by subungual hyperkeratosis (40.4%) and nail discolouration (30.3%). Nail changes were more frequent in older at onset (38.12 ± 16.37 versus 32.61 ± 12.25 , $p=0.038$); in male [70 (78.7%) and (58.8%); $p=0.012$]; duration of psoriasis more than 5 years (47 (52.8%) versus 7 (13.7%); $p<0.001$) and severe psoriasis (69 (77.5%) versus 15 (29.4%); $p<0.001$) in univariate analysis. But in multivariate analysis, duration more than 5 years (OR=13.258, CI=1.968-89.310, $p=0.008$) and severe psoriasis (OR=8.186, CI=3.389-19.771, $p<0.001$) were independent risk factors of nail changes in psoriasis.

Conclusion: Nail abnormality is common in psoriasis with pitting and subungual hyperkeratosis are more frequent. Prolong disease duration and severe psoriasis are found to have in predict prevalence of nail abnormality in psoriasis.

Key words: Nail changes, Pitting, Subungual hyperkeratosis, Onycholysis

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Introduction

Psoriasis is a chronic inflammatory skin condition that affects approximately 2-3% of the worldwide population, making it one of the most prevalent dermatological diseases.¹ This multifactorial disease presents as red, scaly, and thickened plaques that can occur anywhere on the body.²

While skin manifestations are the most characteristic findings of psoriasis, nail involvement is an often-overlooked clinical symptom of the disease.³ Approximately 10-78% of patients with psoriasis have concurrent nail psoriasis,^{4,5} while isolated nail involvement is seen in 5-10% of patients.^{3,6}

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Although the pathology of nail abnormality in psoriasis is not completely understood, it implies the association of genetic, environmental and immunological factors (T-cell mediated inflammatory reaction). The clinical presentation of nail changes in psoriasis varies according to the severity and localization of the lesion. The nail matrix is the germinal center of the nail, with the proximal matrix forming the dorsal nail plate and the distal matrix forming the ventral nail plate. The nail plate is a specialized epidermal structure formed by a process known as onycholemmal keratinization, whereby matrix cells mature, lose their nuclei and organelles, and become commented in a thick mortar.⁷ Nail matrix involvement-result in features such as pitting, leukonychia, onycholysis, Beau's lines and crumbling of the nail plate. While nail bed involvement- leads to oil-drop discoloration, onycholysis, subungual hyperkeratosis and splinter haemorrhages.⁸ Nail pitting (psoriasis punctata unguinum, onychia punctata) is the most common changes in psoriasis of nail. Pits in the nails are superficial depressions in the nail plate, single or multiple, that indicate abnormalities in the proximal matrix. Oil drops are translucent, yellow discolorations of nail plate observed beneath the nail plate often extending distally toward the hyponychium, due to psoriasiform hyperplasia, parakeratosis, microvascular changes, and trapping of neutrophils in the nail bed subungual hyperkeratosis is due to hyperkeratosis of the nail bed and is often accompanied by onycholysis, which usually involves the distal aspect of nail.⁹

The nail abnormalities in psoriasis depend on the localization of the inflammation in the nail unit. Two patterns of nail disorders are commonly seen in psoriasis. Nail matrix involvement-result in features such as pitting, leukonychia, onycholysis, Beau's lines and crumbling of the nail plate. While nail bed involvement-leads to oil-drop discoloration, onycholysis, subungual hyperkeratosis and splinter haemorrhages.⁸ Williamson et al.¹⁰ have reported a direct proportion between the severities of skin lesions and nail psoriasis. Nail psoriasis affects patients of both sexes but the frequency increases with age. These nail changes can occur independently or in combination, and their frequency and severity can vary among patients.¹¹

Several authors have investigated the frequency of nail changes in patients with psoriasis. A study conducted by

Dogra and Arora,¹² found that nail changes were present in 90.9% of patients of psoriasis. Another study by Rich et al.¹³ reported nail changes in 71% of patients with psoriasis. The frequency of nail changes in psoriasis may vary depending on the duration and the severity of the disease. For instance, a study by Ahmed and Nasreen,¹⁴ found that nail changes were more common in patients with severe psoriasis.

Both nail bed and nail matrix involvement in the course of the disease result in heterogeneous specific clinical signs. The esthetic aspect, impaired functionality and increased psychological stress in these patients.¹⁵ In addition, nail involvement in psoriasis can have a significant impact on the quality of life of affected individuals, as it can cause pain, deformity, and difficulty performing daily activities.¹⁶ Moreover, nail psoriasis can be challenging to diagnose and treat, which further adds to the burden of the disease.¹⁷ Therefore, nail involvement in psoriasis requires a better recognition of clinical nail injury patterns and an accurate evaluation of the degree of nail psoriasis.¹⁵ Purpose of this study was to see the frequency and clinical presentations of nail changes in patients with psoriasis.

Material and Methods

This cross-sectional analytical study was conducted in the Department of Dermatology, Sylhet MAG Osmani Medical College Hospital, Sylhet from January 2017 to December 2019. Cases of psoriasis vulgaris including psoriatic arthritis of any age and both genders were enrolled after a written consent. Nail changes secondary to trauma, concomitant systemic disease, or other inflammatory conditions affecting joints, and co-existing dermatological disease other than psoriasis were excluded.

A detailed history was recorded including age of the patients, age of onset, site and duration of the disease, associated joint involvement, personal history, relevant drug history, past history and family history. A thorough clinical examination was done in each patient consisting of general survey, cutaneous and systemic examinations including assessment of joint involvement. Diagnosis of psoriasis was made on clinical history and examination. A skin biopsy and histopathology examination was performed in doubtful case. In this way 140 cases of psoriasis were included in this way. Finger and toenails were clinically observed and nail changes were noted.

The Psoriasis Area and Severity Index (PASI) was calculated that was based on the body surface area involved (in each of four anatomic areas – head, upper extremities, trunk and lower extremities) and clinical grading of lesional erythema, induration and scaling. The PASI ranges from 0 to 72.18 PASI score of <7, 7-12 and >12 was taken as mild, moderate and severe disease respectively.¹⁹

Data were analyzed using SPSS version 23, and descriptive statistics, frequencies, percentages, mean and standard deviation. Chi-square tests, unpaired t test and logistic regression model were used to analyze the data.

Results

A total of 140 patients of psoriasis were enrolled and nail change were found in 89 (63.6%) cases and nail change were absent in 51 (36.4%) cases.

Age of the patients with nail change ranged from 2 years to 70 years with the mean age of 44.03 ± 15.87 years.

Age of onset of psoriasis with nail change ranged from 1 year to 63 years with the mean age of onset was 38.12 ± 16.37 years.

There were 70 males (78.7%) and 19 females (21.3%) among the patients of psoriasis with nail change.

Of the patients with nail change, 47 (52.8%) had duration of psoriasis more than 5 years while remaining 42 (47.2%) had less than 5 years (Figure-1).

Mild psoriasis was in 20 (22.5%) cases, moderate psoriasis in 44 (49.4%) cases and severe psoriasis in 25 (28.1%) cases (Figure-2).

The most frequent nail change was pitting (61.8%) followed by subungual hyperkeratosis (40.4%), nail discolouration (30.3%) longitudinal ridging (19.1%) onycholysis (14.6%), nail thickening (13.5%), opaque nail (9.1%), Beau's line (9.1%) and oil spot (2.2%) (Table-I).

Patients having nail changes were significantly older both age (years) at presentation (44.03 ± 15.87 versus 36.31 ± 12.88 , $t=2.960$, $p=0.004$) and at onset (38.12 ± 16.37

versus 32.61 ± 12.25 , $t=2.093$, $p=0.038$) compared to those without nail change. Nail changes were more frequently male [70 (78.7%) male and 19 (21.3%) female versus [30 (58.8%) male and 21 (42.2%) female; $\chi^2=6.246$; $p=0.012$]; duration of psoriasis more than 5 years [47 (52.8%) versus 7 (13.7%); $\chi^2=20.902$; $p<0.001$] and severe psoriasis (69 (77.5%) versus 15 (29.4%); $\chi^2=31.276$; $p<0.001$] compared to those without nail change (Table-II).

We tried to fit a logistic regression model using nail involvement in psoriasis as the dependent variable and the other demographic characteristics and duration and severity of psoriasis as the independent ones. The logistic regression model showed that duration > 5 years (OR=13.258, CI=1.968-89.310, $p=0.008$) and severe psoriasis (OR=8.186, CI=3.389-19.771, $p<0.001$) were statistically significant in predicting nail involvement in psoriasis (Table-III).

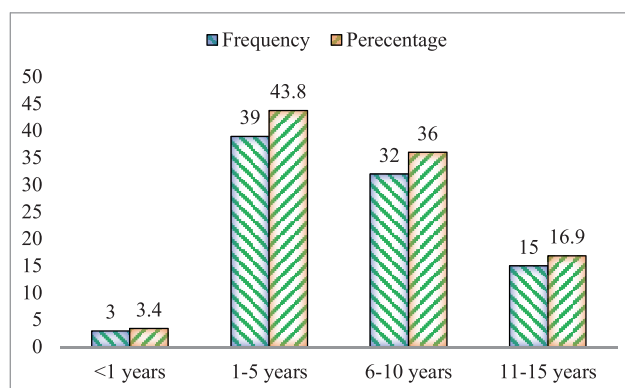


Figure-1: Distribution patients of psoriasis with nail change according to duration of psoriasis (n=89)

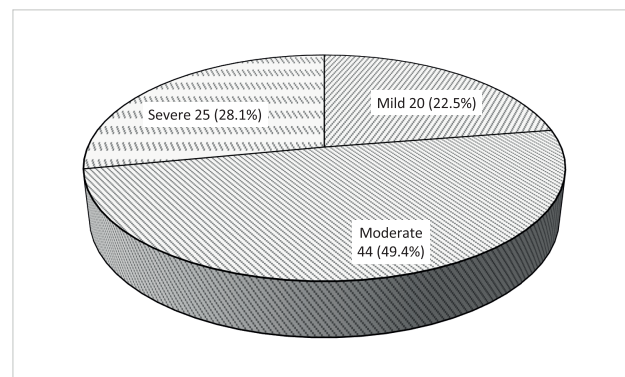


Figure-2: Distribution patients of psoriasis with nail change according to severity of psoriasis (n=89)

Table-I: Distribution patients of psoriasis with nail change according to pattern of nail changes

Nail changes	Frequency	Percentage
Pitting	55	61.8
Nail thickening	12	13.5
Onycholysis	13	14.6
Oil spot	2	2.2
Longitudinal ridging	17	19.1
Subungual hyperkeratosis	36	40.4
Opaque nail	9	10.1
Nail discolouration	27	30.3
Beau's line	9	10.1

Table-II: Comparison of various parameters between patients of psoriasis with or without nail change

Parameters	With nail changes (n=89)	Without nail changes (n=51)	p-value
Age			
At presentation	44.03 ± 15.87	36.31 ± 12.88	*p=0.004
At onset	38.12 ± 16.37	32.61 ± 12.25	*p=0.038
Sex			
Male	70 (78.7%)	30 (58.8%)	†p=0.012
Female	19 (21.3%)	21 (42.2%)	
Duration			
<5 years	42 (47.2%)	44 (86.3%)	†p<0.001
≥5 years	47 (52.8%)	7 (13.7%)	
Severity of psoriasis			
Mild to moderate	20 (22.5%)	36 (70.6%)	†p<0.001
Severe	69 (77.5%)	15 (29.4%)	

P-value was reached from †Chi-square (χ^2) tests and *unpaired t test

Table III: Results of logistic regression analysis for nail involvement in psoriasis

Variables	B	SE	Significance	Exp (B)	95.0% CI for exp (B)	
					Lower	Upper
Age	0.090	0.112	0.421	1.094	0.879	1.362
Age at onset	-0.098	0.109	0.369	0.907	0.732	1.123
Male gender	0.646	0.489	0.187	1.907	0.731	4.974
Duration > 5years	2.585	0.973	0.008	13.258	1.968	89.310
Severe psoriasis	2.102	0.450	0.000	8.186	3.389	19.771
Constant	-3.633	1.470	0.013	0.026		

Discussion

Nail changes in psoriasis are common but it is relatively less explored and often overlooked. In many cases it causes psychologic stress, pain, impairment of manual dexterity and a significant negative impact on a patient's quality of life. Such an impact of nail changes in psoriasis definitely warrants an insight into its clinical manifestations and treatment options by a present-day dermatologist.¹²

In this study nail change was recorded in 63.6% cases. This result correlated with the study of Ovcina-Kurtovic and Kasumagic-Halilovic,⁹ which reported nail abnormalities were present in 60.9% of patients with psoriasis. Ahmed and Nasreen,¹⁴ found that a frequency of nail changes was 58% in patients of psoriasis. Higher rate of nail changes in patients with psoriasis was reported in some,^{5,20,21} whereas lower rate of nail changes in patients with psoriasis was reported in others.^{15,22,23}

Age of the patients with nail change ranged from 2 years to 70 years with the mean age of 41.88 ± 15.63 years. Nearly similar age distribution the patients was reported other study that mean age of psoriatic with nail involvement was 43.62 ± 15.31 years.²⁴ Chowdary et al.²⁵ found the mean age of psoriatic with nail involvement was 42.56 years ± 14.45 years

Age of onset of psoriasis with nail change ranged from 1 year to 63 years with the mean age of onset was 36.37 ± 16.74 years. Marina et al.²⁶ found the mean age of onset of fingernail psoriasis was 46.29 ± 13.89 years.

There were 89 males (78.7%) and 19 females (21.3%) among the patients of psoriasis with nail change. Male preponderance of nail psoriasis was reported in several studies.^{14,20,23,24}

In this study 52.8% patients had duration of psoriasis more than 5 years and 47.2% had less than 5 years. Mirza et al.²⁰ found that 56% patients were suffering from psoriasis above 5 years and 44% patients suffering from psoriasis for up to 5 years. Ahmed and Nasreen,¹⁴ found that 83% of the patients with nail psoriasis had been suffering from psoriasis for more than 5 years while remaining (17%) for less than 5 years. Whereas, Idrees et al.²¹ found that 59% patients suffering from psoriasis for up to 5 years and remaining 41% patients were suffering from psoriasis above 5 years.

The most frequent nail change in this study was pitting (6.18%) followed by subungual hyperkeratosis (40.4%), nail discolouration (30.3%) longitudinal ridging (19.1%) onycholysis (14.6%) and nail thickening (13.5%), opaque nail (9.1), Beau's line (9.1%) and oil spot (2.2%). Ovcina-Kurtovic and Kasumagic-Halilovic,⁹ reported nail changes in patients with psoriasis were pitting (47.8%), discoloration of nail plate (17.9%), subungual hyperkeratosis (14.9%), oil spot (8.9%), nail thickening (5.9%) and onycholysis (4.5%). Hayat et al.²⁷ found the most common nail finding was pitting (24.6%), followed by onycholysis (19.8%) and subungual hyperkeratosis (19.8%).

In this study patients with nail change were significantly older than psoriatic patients without nail abnormalities ($p=0.004$) in univariate analysis but no significant difference in multivariate analysis ($OR=1.094$, $CI=0.879-1.362$, $p=0.421$). This result was consistent with several studies.^{23,28} But other studies found that age of the patients with psoriatic nail change did not differ significantly compared to psoriatic patients without nail abnormalities.^{5,21}

In the present study patients with psoriatic nail change were more frequently male, but no significant difference in multivariate analysis ($OR=1.907$, $CI=0.731-4.974$, $p=0.187$). This finding was supported by several studies.^{5,29} But other studies found that no gender difference between psoriatic patients with and without nail abnormalities.^{14,22,23}

In this study patients with psoriatic nail change had prolong duration of psoriasis more than 5 years compared to those without nail change in univariate and also in multivariate analysis ($OR=13.258$, $CI=1.968-89.310$, $p=0.008$). Idrees et al.²¹ supported this finding that duration of psoriasis was significantly higher in patients with psoriatic nail change than psoriatic patients without nail change. But Yap and Pubalan,³⁰ found no relation of psoriatic nail change and duration of psoriasis.

In current study patients with psoriatic nail change had more frequent in severe psoriasis in univariate and also in multivariate analysis ($OR=8.186$, $CI=3.389-19.771$, $p<0.001$). This finding correlated with several studies.^{21-24,29,30} This observation, however, is not supported by findings of a study of Chinese patients.³¹

Limitations of the study were: (1) single centre study and (2) small sample size.

Conclusion

Nail involvement is common in patients with psoriasis. Pitting and subungual hyperkeratosis are the most frequent nail abnormality. Longer duration of disease and severe psoriasis are found to have predicted the prevalence of nail abnormality in psoriasis. However further prospective studies including larger sample size are needed to establish a definite conclusion.

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Original Article

Dimensions of Knee Joint Space Width and Their Gender Variations Among Adult Bangladeshi Population

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Abstract

Background: Human knee joint shows great variation in different radiographic dimensions. Dimensions of different knee joints can be studied by following techniques: visual observation, anthropometric measures and radiographic measures. Among these methods, radiographic measurements were regarded as most important for assessing skeletal alignment of knee joint in a static weight bearing position.

Objective: The present study aimed to find out the differences in dimensions of knee joint space between adult Bangladeshi male and female.

Methods: A cross-sectional analytical type of study carried out in the Department of Anatomy, Dhaka Medical College, Dhaka from July, 2018 to June, 2019. The study population was adult male and female attending the Radiology and Imaging department of Dhaka Medical College and Hospital (DMCH) for the X- ray of any region of the body advised by their physicians. Digital radiographs of knee joint antero-posterior and lateral view were taken. The study was performed on 80 Bangladeshi population where 40 was male and 40 was female.

Results: In the present study, the mean medial and lateral compartment of knee joint space width was significantly higher ($P<0.01$) in male than in female when compared no significant differences ($p=0.588$) and $p=0.297$) was observed

Conclusion: The present study was an attempt to produce a radiographic standard data on different dimensions of knee joint space and to find out the differences in dimensions of knee joint space between adult Bangladeshi male and female.

Key words: Dimensions of knee joint, Gender variation, Joint space width

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Introduction

The knee is a weight-bearing synovial joint comprising of two condylar joints and a saddle joint. Condylar joints are formed between femoral and corresponding tibial condyles. The tibio-femoral articulations are separated by articular cartilages and menisci with little contributions from cruciate ligaments.

Normal knee joints are analyzed to determine a quantitative relationship between the joint line of knee and

the bony landmarks. Wide variability is found in the absolute distance from each landmark to the joint line of knee, including significant differences between sexes.

The bony landmarks are identified on a digital radiograph of knee joint in both antero- posterior and lateral view. Anatomical landmarks of knee joint are adductor tubercle, tibial tuberosity, medial epicondyle, lateral epicondyle, intercondylar eminence etc.

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Joint space are seen on frontal radiograph as a radiolucent area between the tibial and the femoral condyles. As a result of wear and tear over time, there is a gradual reduction in joint space with increasing age. Knee joint space narrowing is the most common joint disorder and responsible for substantial economic, social and psychological costs. In case of joint space narrowing, medial compartment of knee appears earlier

The values of different radiographic dimensions of knee of adult Bangladeshi population may be helpful to the anatomists for a normative reference. For the radiologists, the normative values may be helpful in their diagnosis. For proper designing of a prosthetic knee by the ergonomist and for surgical reconstruction by the orthopedic surgeons, knee radiographic data is essential.

Materials and Methods

This was a cross-sectional analytical type of study. The study was performed on 80 Bangladeshi population of 24-49 years of age were selected from attending the Radiology and Imaging department of Dhaka Medical College and Hospital (DMCH) for the X-ray of any region of the body advised by their physicians. The subjects were requested to give permission for another exposure of anterior posterior and lateral view radiographs of knee joint. Subjects were purposively selected. Informed written consent was taken from each subject. Permission was also taken from the Head of the Department of Radiology and Imaging of DMCH. The study was performed on 80 Bangladeshi population where 40 was male and 40 was female.

Information regarding inclusion of subjects: Bangladeshi by nationality. Twenty-four to forty-nine years of age (confirmed by her/his national identity card). For exclusion of subjects: having any congenital lower limb deformities, individual having abnormal gait, trauma or surgery of knee joint, individual with clinical or radiographic evidence of inflammatory/degenerative arthritis, over weight bearing limb and any active or chronic joint disease. The ossification of all bones of lower limb is completed by the age of twenty to twenty-four years. So fully ossified foot achieves its adult form and fixed measurements after this age.

An International Agreement for the unification of anthropometric measures on the living was adopted in the Fourteenth International Congress of Prehistoric Anthro-

pology and Archeology held in Geneva, September 9-14, 1912. According to this agreement for paired measurements, it is recommended to operate on the left side. Measures were taken to minimize the radiation hazards of the subjects: A good quality radiograph was taken at the first time to reduce the number of repeated exposures. Collimated primary X-ray beam was used to reduce scattered radiation. Gonadal shields and protective eye wear were used for protection of testes and eye.

Medial compartment joint space width (MJSW): the distance between the distal femur and the proximal tibia. The maximum height of medial joint space width is the mid-portion of the medial compartment of the knee

- Lateral compartment joint space width (LJSW) the distance between the distal femur and the proximal tibia. The maximum height of the lateral joint space width is the mid-portion of the lateral compartment of the knee (Fig-1)



Fig-1: Digital radiograph of the left knee joint anterior-posterior view showing the medial compartment joint space width (MJSW) lateral compartment joint space width (LJSW)

This protocol was first presented and approved by the Research Review Committee (RRC) of Dhaka Medical College. A written clearance from the Ethical Review Committee (ERC) of Dhaka Medical College, Dhaka was taken. Data were collected by quantitative method and were checked and edited after collection. All the data obtained were recorded using an Excel spread sheet (Microsoft Office 2010; Microsoft, Redmond, WA). All statistical analyses were performed by using the statistical software SPSS software package (IBM SPSS Statistics Base, version 22), keeping the objective of the study in view. Statistical test such as Unpaired Student's 't' test was done. Then the calculation of mean and standard deviation (SD) with minimum and maximum values were done for each variable in both groups. Statistical significance was accepted at P- value equal to or less than 0.05 ($p < 0.05$).

Results

The study was performed on 80 Bangladeshi population of 24-49 years of age were selected from attending the Radiology and Imaging department of Dhaka Medical College and Hospital (DMCH). All the study subjects were divided into two groups according to sex. After collection of data, the findings of this study were analyzed by SPSS (Statistical Package for Social Sciences) version 22.0.

Medial compartment joint space width (MJSW):

In male, medial compartment joint space width (MJSW) ranged from 1.12 mm to 5.29 mm and the mean length was 2.25 ± 0.97 mm.

In female, medial compartment joint space width (MJSW) ranged from 1.02 mm to 4.01 mm and the mean length was 2.14 ± 0.79 mm.

No significant difference ($p=0.588$) of medial compartment joint space width was observed when compared between male and female (Fig-2).

Lateral compartment joint space width (LJSW):

In male, lateral compartment joint space width (LJSW) ranged from 2.38 mm to 6.35 mm and the mean length was 4.05 ± 0.91 mm.

In female, lateral compartment joint space width (LJSW) ranged from 2.13 mm to 5.02 mm and the mean length was 3.82 ± 0.91 mm.

No significant difference ($p=0.297$) of lateral compartment joint space width was observed when compared between male and female.

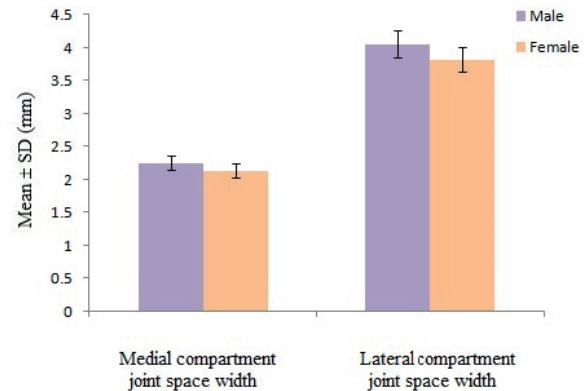


Fig 2: Bar diagram showing medial compartment joint space width (mm) and lateral compartment joint space width (mm) in male and female

Discussion

This study was carried out to get different radiographic knee joint space dimensions of adult Bangladeshi male and female. The morphometry of knee joint is influenced by gender, age, geographical, socioeconomic, food habits, ethnicity and racial factors. So, each population has specific baseline knee radiographic data to optimize the accuracy of identification.

In the present study, the age ranges of study subjects were within 24-49 years. The age limit was considered within such an age when the developments of the respective body parts are complete and the age-related changes in the knee joint features are minimum.

In the present study, the mean value of medial compartment joint space width and lateral compartment joint space width was significantly higher in male than in female and when compared statistically significant ($p=0.0000$) difference was observed.

According to Anas, et al. conducted a study in the Department of Radiology, Aminu Kano Teaching Hospital and Federal Medical Center, Nigeria. According to their study medial compartment joint space width and lateral compartment joint space width (mm) was significantly higher in male than in female and when compared no significant differences was observed

According to Jain, Abhishek and Mahajan studied on 49 medical staffs (26 male and 23 female) in the Department of Orthopedics, SAMC&PGI, Indore, Madhya Pradesh, India medial compartment joint space width and lateral compartment joint space width (mm) was significantly higher in male than in female and when compared no significant differences was observed

Results of different radiographic variables of this study were compared with the radiographic variables of other studies from different countries. But so far it is known, there is no published data on radiographic knee dimensions in our country. So the present study could not be compared with any previous similar study on Bangladeshi adult population.

Limitations could be identified as the present study was conducted only in a single institute, at the Radiology Department of Dhaka Medical College Hospital. So, the results of the study may not be the full representation of the whole community of Bangladesh. During selection of study subjects the assessment of the exclusion criteria was based totally on the information gathered through verbal enquiry and on visual impression. This might have failed to identify some study subjects who could have revealed some exclusion criteria on more sophisticated screening. The study was based on measurement of different variables on soft copy of X-ray film by MB ruler which might have produced minor errors.

Conflict of Interest : None

Conclusion

In this study, medial and lateral compartment joint space width (mm) was significantly higher in male than in female. The study findings suggest that morphological measurements of the left knee joint space dimensions showed significant variations among adult Bangladeshi male and female people.

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Original Article

Assessment of Antibiotic Prescription Pattern in Two Departments of a Selected Medical College Hospital of Bangladesh

Afroz R¹, Akhter S², Rahman TR³, Akter N⁴, Ghosh S⁵, Das AK⁶

Abstract

Background: Antibiotic therapy is considered to be a principal component of infectious disease management in health care settings. Inappropriate antibiotic use leads to adverse drug effects, increase cost, increase the mortality morbidity and emergence of resistance. Poor education, absence of guideline, lack of resources, poor training, patient load and inaccessibility of antibiotics can cause irrational use of antibiotics.

Objective: To assess the pattern of antibiotic use among hospitalized patients according to BSMMU guideline and WHO Access, Watch, Reserve (AWaRe) classification.

Methods: An observational descriptive cross-sectional study was carried out in the department of Surgery and department of Gynaecology & Obstetrics of Khulna Medical College Hospital from January 2022 to December 2022. A total 300 prescriptions containing antibiotics was purposively selected. The BSMMU guideline and World Health Organization AWaRe categorization of antibiotics were used to assess the prescribing practices of physicians.

Results: In this study the demographic profile showed the predominant age group was 21-40 years. Combination of two antibiotics prescribed mostly 59.3% was from Gynaecology & Obstetrics department. Antibiotics were prescribed mostly in trade name 89.3% from Surgery department. Most of the antibiotics prescribed by parenteral route 76% from Gynaecology & Obstetrics department. Most of the frequency of administration was 12 hourly, which was from Gynaecology & Obstetrics department 90.7%. Most of the type of therapy was empirical from Surgery Department 83.3%. Most commonly prescribed antibiotic group was Nitroimidazole group 78.67% from Gynaecology & Obstetrics department. Most of the prescriptions 64.0% were adhered to BSMMU antibiotic guideline from Surgery department. Most of the prescribed antibiotics 58.0% were from watch group of WHO AWaRe classification belongs to Gynaecology & Obstetrics department.

Conclusion: Use of antibiotic therapy should be evidence based. Formulation of institutional antibiotic guideline and formulation of individual Hospital Antibiotic Policy may be helpful.

Key words: Antibiotic, BSMMU antibiotic guideline, WHO AWaRe classification.

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Introduction

Antibiotics have two sided effects-one is control of infection, the other is the resistance of an organism. Continuous, indiscriminate and excessive use of antimicrobial agents promotes the emergence of resistant organisms which is a global threat today which also increases the health costs. Prescription with antibiotic is

mostly made on empirical basis and broad-spectrum. Worldwide more than 50% of all medicines are prescribed, dispensed, or sold inappropriately, while 50% of patients fail to take them correctly¹. In India, the antimicrobial agents use varies from 24 to 67% and 64% of total prescribed antibiotics are either inappropriate or not

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indicated in terms of drug or dosage². Pattern of antibiotic use in different departments of Dhaka Medical College Hospital showing: ceftriaxone 49.75%, metronidazole 17.5% and ciprofloxacin 12.75%³. Different studies conducted in Bangladesh during last decade showing that the use of antibiotics is not satisfactory and the condition is contributing to emergence of resistance day by day. Antimicrobial stewardship program (ASP) is considered as the most effective approach to improve antimicrobial prescribing. ASP is designed by BSMMU to support the implementation of updated antimicrobial guideline. The reported prevalence of antibiotic utilization in admitted patients is 59.4% in four selected departments of Bangabandhu Sheikh Mujib Medical University (BSMMU)⁴. By 2023, the WHO aims for at least 60% of all antibiotic consumption to come from the Access group⁵.

Methodology

An observational descriptive cross-sectional study was carried out in the department of Surgery and department of Gynaecology & Obstetrics of Khulna Medical College Hospital. The duration of the study was one year from January 2022 to December 2022. A total 300 prescriptions containing antibiotics was purposively selected. Patients above 18 years of age and both gender, admitted to the Department of Surgery and Gynaecology & Obstetrics of Khulna Medical College Hospital, who were prescribed with antibiotics, were included. Admitted patients who were unwilling to take part in the study and patients who were not prescribed with antibiotics, were excluded. The protocol of the study was approved by Ethical Review Committee of Dhaka Medical College. Written permission was then obtained from the authority of KMCH after giving verbal explanation on the nature of the study, its purpose, and the potential benefit of the study. Study subjects were selected by purposive sampling technique. Before recruitment, aim, benefit and procedure of the study was explained and informed written consent was taken from each study subject and the picture of those prescriptions were captured by a photo capturing device. Later on, the photo of antibiotic containing prescription was screened and reviewed to record in the data collection form. Data was analyzed using descriptive statistics. Continuous data was expressed as mean $\pm$ SD (standard

deviation) and the nominal data was expressed as percentages. Analysis of data was carried out by using Statistical Package for Social Sciences (SPSS) 26 version.

Results

Table-I: Distribution of the study patients according to age(n=300)

Age group (years)	Obs & Gynae No.(%)(n=150)	Surgery No.(%)(n=150)	Total No.(%)(n=150)
<20	16(10.7%)	8(5.3%)	24(8.0%)
21-40	99(66.0%)	51(34.0%)	150(50.0%)
41-60	28(18.7%)	57(38.0%)	85(28.3%)
61-80	7(4.7%)	33(22.0%)	40(13.3%)
>80	0(0.0%)	1(0.7%)	1(0.3%)
Total	150(100.0%)	150(100.0%)	300(100.0%)

Table-I shows the distribution of study patients based on their age in years where majority of patients 99(66.0%) were from Obs & Gynae department between the ages of 21 to 40 years.

Table-II: Proportion of number of antibiotic per prescription

Number of antibiotics	Obs&Gynae (n=150)No.(%)	Surgery (n=150)No.(%)	Total
One	28(18.7)	70(46.7)	98 $\times$ 1=98
Two	89(59.3)	75(50.0)	164 $\times$ 2=328
Three	33(22.0)	5(3.3)	38 $\times$ 3=114
Total	150(100.0)	150(100.0)	540

Mean 1.8

Table-II shows proportion of number of antibiotic per prescription. Where combination of two antibiotics were prescribed mostly from Gynaecology & Obstetrics Department 89(59.3%).

Nomenclature	Obs & Gynae No.(%)(n=150)	Surgery No.(%) (n=150)
Generic	64(42.7)	10(6.7)
Trade	36(24.0)	134(89.3)
Both (Generic +Trade)	50(33.3)	6(4.0)
Total	150(100.0)	150(100.0)

Table III shows the distribution of prescribed antibiotic by nomenclature. Where most them were in trade name 134(89.3), from Surgery department.

Table IV: Distribution of study patients by route of antibiotic administration (N=300)

Route	Obs & Gynae No.(%) (n=150)	Surgery No. (%) (n=150)
Parenteral	114(76.0)	90(60.0)
Oral	33(22.0)	56(37.3)
Both (Parenteral +oral)	3(2.0)	4(2.7)
Total	150(100.0)	150(100.0)

Table IV shows the distribution of study patients by route of antibiotic administration. Where most of the antibiotics prescribed by parenteral route 114(76.0%), were from Gynaecology & Obstetrics department.

Table -V: Distribution of study patients by frequency of antibiotics administration (n=300)

Frequency	Obs & Gynae No.(%) (n=150)	Surgery No. (%) (n=150)
6 hourly	3(2.0)	5(3.3)
8 hourly	7(4.7)	13(8.7)
12 hourly	136(90.7)	125(83.3)
24 hourly	4(2.7)	7(4.7)
Total	150(100.0)	150(100.0)

Table V shows distribution of study patients by frequency of antibiotics administration. Where most of the frequency of antibiotics administration was 12 hourly 136(90.7%), from Gynaecology & Obstetrics Department.

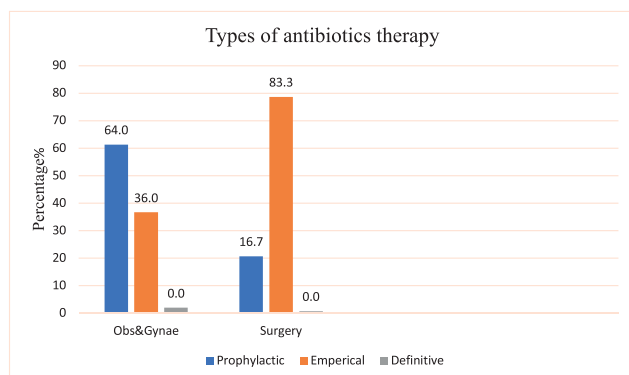


Figure1: Distribution of study patients by types of antibiotics therapy

Table-VI: Distribution of study patients according to group of antibiotic

Antibiotic	Obs& Gynae No.(%) (n=150)	Surgery No. (%) (n=150)
Cephalosporin	99(66.0)	111(74.0)
Penicillin	3(2.0)	34(22.67)
Fluroquinolone	50(33.33)	5(3.33)
Aminoglycosides	27(18.0)	2(1.33)
Macrolides	4(2.67)	4(2.67)
Carbepenem	3(2.0)	16(10.67)
Nitroimidazole	118(78.67)	47(31.33)
Clindamycin	1(0.67)	2(1.33)
Linezolid	0(0.0)	14(9.33)

Table VI shows that most of the prescribed antibiotics were from Nitroimidazole group 118(78.67%) in Gynaecology & Obstetrics department.

Table-VII: Distribution of study patients by name of antibiotic

Name of antibiotic	Obs &Gynae No.(%) (n=150)	Surgery No.(%) (n=150)
Cefixime	11(7.33)	29(19.33)
Ceftriaxone	84(56.0)	62(41.33)
Cefuroxime	3(2.0)	6(4.0)
Ciprofloxacin	50(33.33)	1(0.67)
Co-amoxiclav	1(0.67)	3(2.0)
Flucloxacillin	2(1.3)	31(20.67)
Meropenem	3(2.0)	13(8.67)
Metronidazole	114(76.0)	47(31.33)
Moxifloxacin	-	1(0.67)
Amikacin	26(17.33)	2(1.33)
Clindamycin	1(0.67)	2(1.33)
Azytrhomycin	4(2.67)	3(2.0)
Clarithromycin	-	1(0.67)
Ornidazole	4(2.67)	-
Levofloxacin	-	3(2.0)
Cefepime	-	4(2.67)
Cefuroxime+Clavulanicacid	2(1.33)	10(6.67)

Table VII shows that most of the prescribed antibiotics were Metronidazole 114(76%), from Gynaecology & Obstetrics Department.

Table-VIII: Distribution of antibiotics on the basis of Adherence to BSMMU guideline (N=300)

Adherence type	Obs & Gynae No.(%)(n=150)	Surgery No.(%)(n=150)
Adherence	76(50.7)	96(64.0)
Non-adherence	74(49.3)	54(36.0)
Total	150(100.0)	150(100.0)

Table VIII shows the distribution of antibiotics on the basis of Adherence to BSMMU guideline. Where most of the prescriptions were adhered to BSMMU antibiotic guideline, were from Surgery Department 96(64.0%).

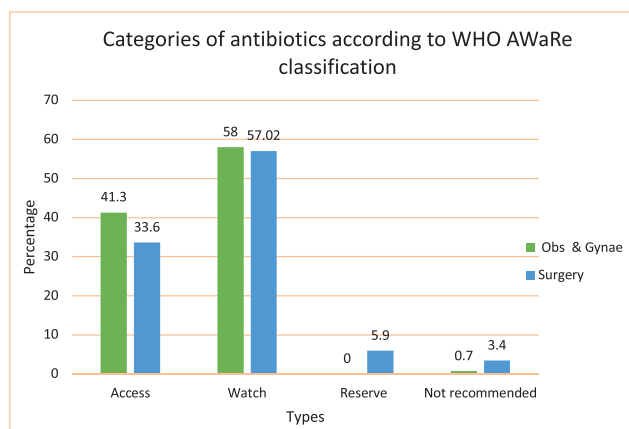


Figure 2: Distribution of antibiotics according to WHO AWaRe classification

Discussion

Antibiotics are most commonly prescribed drugs in health care facilities. The effectiveness and easy access to antibiotics have also led to their overuse. Education to Health care providers, dispensers and patients about the causes and control mechanism of antimicrobial resistance will be effective approach to prevent dissemination of infection⁶. In this study the demographic profile showed the predominant age group was 21-40 years 99(66.0%) from Gynaecology & Obstetrics department, which was similar to the study conducted in Assam, India where 21-40 years age group was predominant 53%⁷.

The average number of drugs per prescription is an important parameter of a prescription audit. To minimize the risk of drug interactions, development of bacterial resistance and hospital costs, it is preferable to keep the

number of drugs per prescription as low as possible. In our study we concentrated only on antibiotics. The average number of antibiotics per prescription was 1.8 which was similar to the study in western Nepal (1.7)⁸ and northern India (1.61)⁹. Two antibiotic was prescribed mostly in Gynaecology & Obstetrics department 89 (59.3%), which was similar to the study in a tertiary care hospital in India (41.6%)¹⁰. In a study of Yemen showed two antibiotics was prescribed (55.73%)¹¹ and in Uttarakhand India (39%)¹².

Prescribing drug by generic name can promote the rational use of drugs with regards to safety, efficacy and cost by identifying the product with its scientific name. But in this study most of the prescribed antibiotics was in trade name 134(89.3%), from Surgery department. A study conducted in Bangladesh in which all the antimicrobial agents were prescribed by their brand names (100%)¹³. Which was not in accordance with the WHO guidelines of rational prescribing that requires generic prescriptions. Prescription by generic name may reduce overall expenditure on drugs, particularly on newer antimicrobials and avoiding medication errors, generic name however are not widely manufactured in Bangladesh.

In this study most of the antibiotics prescribed by parenteral route, was from Gynaecology & Obstetrics 114(76.0%) and frequency of administration for most of them were 12 hourly 136(90.7%) from Gynaecology & Obstetrics. In some studies-conducted in Kerala, India (60%), in Eastern India (82.91%), in Yemen (99.7%), in Mongolia (81.9%) encounters were prescribed by parenteral route^{14,15,11,16}.

In this study, among antibiotic usage in patient of both departments supported by empirical therapy was in Surgery Department 125(83.3%), probable reasons of this high percentage of empirical therapy may be due to in inpatient department most of the patient were severely ill, there were too much patient load than hospital capacity, lack of 24 hours availability of culture and sensitivity opportunity in the hospital and lack of ability of patients to perform culture and sensitivity test. In this aspect, the empirical therapy might be thought to be more beneficial than risk. Similar findings were found to a study of Bangladesh where empirical therapy was 93.6%⁶, and in India empirical therapy was 61.6%¹⁷.

In this current study, the most commonly prescribed antibiotic group was Nitroimidazole 118 (78.67%) from Gynaecology & Obstetrics department, where they were co-prescribed commonly with Cephalosporin 99 (66.0%), and in Surgery department Cephalosporin was 111 (74.0%), Which was similar to the study conducted in India where Cephalosporins were mainly indicated for intra-abdominal infections (34.4%) followed by surgical prophylaxis (24%). Among them 66.4% were co-prescribed with other antibiotics, of which metronidazole (69.9%) was found to be common¹⁷.

In this study most of the prescriptions were adhered to BSMMU antibiotic guideline, in Surgery department 96 (64.0%) and in Gynaecology & Obstetrics department 76 (50.7%). Similar findings were found to a study done in Bangladesh where the adherence to guideline was highest in the department of Obstetrics and Gynecology 91.3%¹⁸.

In this current study mostly prescribed antibiotics 58.0% from Gynaecology & Obstetrics department were from watch group and in Surgery Department that was 57.02%. A study done in Bangladesh in 2022 showed that 62.9%, 36.4%, and 0.2% of the patients were treated with Watch, Access, and Reserve group of antibiotics respectively in Surgery ward, and in Gynaecology & Obstetrics wards, 55.4%, 44.4%, and 0.2% of the patients were treated with Watch, Access, and Reserve group respectively¹⁹. This study was done in a tertiary care hospital and sample size was small, however the condition might be worse in remote corners of Bangladesh.

Conclusion

On the basis of the study findings, it can be concluded that antibiotic therapy was mostly empirical. Use of antibiotic therapy should be evidence based. Formulation of institutional antibiotic guideline and formulation of individual Hospital Antibiotic Policy may be helpful.

Conflict of Interest : None

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Original Article

Incidence of Fistula After Two Flap Palatoplasty: 11 years Experience of a Single Surgeon

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Abstract

Background: Cleft lip and cleft palate is a common congenital anomaly. Palatoplasty is a vital surgical procedure aimed at correcting cleft palate, which can severely impact a patient's ability to speak, eat, and maintain oral hygiene. There are many factors which contribute to achieve good outcome after palatoplasty. Postoperative palatal fistula is one of the most significant long-term complications.

Aims: The purpose of this study was to examine the palatal fistula rate after repair with the two-flap palatoplasty technique and to find out the factors contributing to fistula formation post-surgery.

Methods: It was a retrospective study conducted between July 2013 to June 2024 at a single centre in Bangladesh. During this period 382 cases of primary cleft palate were operated by the same surgeon. Two flap palatoplasty technique was applied for all cases by a single surgeon. Same post-operative protocol was followed in all cases.

Results: Two flap palatoplasty technique was used in 382 palate repair and fistula occurred in 15(3.92%) patients. 11(3.65%) fistula developed in patients who had associated cleft lip and cleft palate and 4(4.93%) fistula developed in patients who had isolated cleft palate. 6 fistula occurred in patients who had Bilateral cleft & palate and 5 fistula occurred in patients having unilateral complete cleft lip & palate. The highest number of fistula 9(11.25%) occurred in patients when cleft palate repaired after 2 years age. 6(1.98) fistula occurred when cleft palate repaired before 2 years age.

Conclusion: The incidence of fistula after primary cleft palate repair is a significant concern for both patients and surgeons. Understanding the factors that influence fistula formation is essential for providing successful surgical outcomes. Cleft palate repair technique is an important contributing factor to fistula formation.

Keywords: Cleft palate, Palatoplasty, Fistula, Risk factor

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Introduction

Cleft lip and cleft palate are the most frequently occurring congenital craniofacial deformities. The average prevalence of cleft lip with or without cleft palate was 7.75 per 10,000 live births in the United States and 7.94 per 10,000 live births internationally.¹ Cleft palate repair is a common surgical procedure performed to correct a congenital birth defect where there is a gap in the roof of

the mouth. The goal of cleft palate surgery is to divide the nasal and oral cavities, maintain normal velopharyngeal function, and minimize restricted postoperative facial growth. While advancements in surgical techniques have improved outcomes, fistula formation remains a challenging complication that can occur after cleft palate repair.

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A palatal fistula is an abnormal communication between nasal and oral cavity. When it occurs after cleft palate repair, it can significantly impact the patient's quality of life and may require additional surgical interventions to correct. Velopharyngeal insufficiency is higher in those who experienced a post-operative fistula.² Palatal fistula occurs when the primary surgical repair of the palate breaks down or healing fails and like cleft palate, it induces hyper nasal speech, articulation disorders, and nasal regurgitation of food and drink.³

The incidence of palatal fistula following cleft palate repair was reported to be 0%–60% historically and 3%–35% in recent studies.⁴⁻⁸ Furthermore, many factors, including the surgical technique, timing of surgery, extent of cleft, presence of cleft lip, surgeon's experience, and associated syndromes, are known to increase the risk of palatal fistula.^{8,9}

To prevent the fistula formation, it is important to find out the factors contributing the fistula formation after palatoplasty. Although there are many studies to find out the incidence of fistula after palatoplasty in the world, in Bangladesh no study was carried out regarding this so far we know. This will be helpful to reduce fistula formation after cleft palate repair.

Methods

This retrospective study was done from July 2013 to June 2024 at a private hospital in Bangladesh which is a partner hospital of smiletrain. During this period 382 cases primary cleft palate were operated by the same surgeon. All cases were operated under general anaesthesia with orotracheal intubation. In our study, two-flap palatoplasty technique was used to provide tension-free, multilayer repairs. Patients having cleft palate with cleft lip and isolated cleft palate operated at this center by a single surgeon were included in the study. Patients operated by the same surgeon outside this center were excluded from this study. For wide cleft, nasal side closure was performed with vomerine flaps. Same post-operative protocol was followed in all cases. Liquid diet was allowed for 4 weeks and semisolid diet for next 2 weeks was given. Proper oral hygiene was maintained. Patients were discharged home 48 hours post op after thorough inspection of surgical sites. Oral toileting instructions were reiterated with emphasis at

discharge. Patients were advised calculated doses of analgesics (NSAIDs), and antibiotics for 7 days. Post operative follow up was given on 2nd, 4th, 8th and 24th week. The location of the fistula was classified using the Pittsburgh classification.

Data were collected on the sex of patients, date of birth, type of cleft deformity, the technique of repair, associated anomalies and surgical outcomes including complications. Written informed consent was taken from guardians. Data collected were subjected to simple statistical analysis using the Statistical Package for Social Sciences (SPSS). Both manual and computer-based statistical analyses of data were done. P value <.05 was considered as significant. Funding of treatments and care was covered under Smile Train grant at the Hospital. Approval of the study protocol was obtained from the Institutional Research and Ethics Committee.

Results

Among 382 palatoplasty, fistula occurred in 15(3.92%) patients. 8 patients were male and 7 patients were female. 11(3.65%) fistula developed in patients who had associated cleft lip and cleft palate and 4(4.93%) fistula developed in patients who had isolated cleft palate. 6 fistula occurred in patients who had bilateral cleft lip & palate and 5 fistula occurred in patients having unilateral complete cleft lip & palate. Two flap palatoplasty technique was used in all palate repair and fistula occurred in 15(3.92%) patients. The highest number of fistula 9(11.25) occurred in patients when cleft palate repaired after 2 years age. 6(1.98) fistula occurred when cleft palate repaired before 2 years age (Table-I).

The most common location of fistulas was the hard palate (53.33%). This is followed by at the junction of the hard and soft palate (40%) and in the soft palate (6.67%) (Table-II). The incidence of fistula was not significantly associated with sex, age at cleft palate repair and with cleft lip, and the extent of the cleft (Table 1). The commonest (9, 60%) cause of fistula formation was wound infection & dehiscence. The next common causes were insertion of hard object in mouth & early intake of solid food which were 2(13.33) for each. 1(6.67%) fistula occurred after palatoplasty of a wide cleft & malalignment of dental arch (Table-III).



Fig-1: Two flap palatoplasty



Fig-2: Site of Palatal fistula

Table-I: Comparison of Fistula Formation on different variables

Variable	No. of patients	No. of fistula (%)	P-value
Sex			
Male	194	8(4.12)	.973
Female	188	7(3.72)	
Age of Cleft palate repair			
0-2 years	302	6(1.98)	.429
>2 years	80	9(11.25)	
Associated Cleft Lip			
Yes	301	11(3.65)	.3966
No	81	4(4.93)	

Table-II: Site of Fistula Formation

Site	Number	Frequency(%)
Hard palate	8	53.33
At Junction of hard & soft palate	6	40
Soft palate	1	6.67

Table-III: Factors contributing to palatal fistula formation

Events	Number	Frequency(%)
Wound infection, dehiscence	9	60
Insertion of hard object in mouth	2	13.33
Early Solid Food intake	2	13.33
Malalignment of dental arch	1	6.67
Wide cleft	1	6.67

Discussion

Cleft palate repair surgery is a complex procedure that aims to close the gap in the roof of the mouth that is characteristic of a cleft palate. This surgical intervention is typically performed early in life to improve feeding, speech, and overall facial development in children born with a cleft palate. Despite advancements in surgical techniques and technology, fistula formation remains a common complication that surgeons commonly face.

Several factors can influence the likelihood of developing a fistula after cleft palate repair surgery. These may include age of the patient, type and severity of the cleft palate, surgeon's experience, techniques of palatoplasty, wound infection, postoperative food regimens, presence of other medical conditions, oral hygiene and caregiver education.

Previous studies have reported a wide range of fistula rates after cleft palate repair, ranging from 0% to 60%.^{4-8,19} Those studies included different types of palatoplasty techniques. We report a 3.92% incidence of palate fistulas in our study. Fistula rate is lower due to application of single technique of two flap palatoplasty in all cases. The incidence of palatal fistula was 3.1% in a study by Park et al.³

Previous studies have reported that high-volume operators produce better outcomes than low-volume operators.⁸⁻¹² Moreover, high-volume operators had better results in terms of dental arch relationships and facial appearance. In this study the surgeon was a high volume operator who operated 382 patients over 11 years, which may have contributed to the lower incidence of fistula.

The optimal age for cleft palate repair is important factor for good outcome specially for favourable speech outcomes. At our hospital, we recommend performing cleft palate repair at the age of 9-12 months. However, there were cases in which the repair was performed after the age of 2 years due to a delayed diagnosis, presentation, poverty and other medical issues. Age range in this study was 9 month to 19 years. While some studies have argued that the incidence of fistula is directly proportional to the patient's age at operation^{6,7}, other studies have reported that there was no significant difference in fistula formation between patients younger than 18 months and older patients.²⁴ Eliason et al. showed that post-operative fistula formation occurs at a higher rate when performed prior to 6 months old.² Our study found no significant association between the incidence of palatal fistula and age at the time of cleft palate repair.

Patient and family education is crucial to improving postoperative outcomes. Many studies have reported that therapeutic education for patients was helpful in optimizing clinical outcomes and it was also reported that patient and family education lowered the incidence of palatal fistula more than 2-fold.¹⁶ In this study post-opera-

tive monitoring and care, caregivers education on wound care and oral hygiene, instruction to all the guardians about postoperative management and good communication were maintained. The use of arm restraints is a commonly accepted practice and may be beneficial in preventing trauma to the repair.¹⁷ We instructed the guardian not to put finger or any hard object into the mouth. We advised guardian to put foot socks in hands of child so that fingers couldn't be separated and the child was unable to put his/her finger into mouth. Even then 2(13.33%) fistula occurred due to insertion of hard object into mouth. According to our experience, good communication between physicians and patients' parents played a critical role in reducing the incidence of fistula.

Postoperative wound infection is a common cause of wound dehiscence leading to fistula formation. Proper antibiotic, maintaining oral hygiene is mandatory to reduce wound infection. Although wound infection was managed properly even then 9(60%) fistula occurred due to delayed presentation with wound infection in this study.

Palatoplasty techniques have greater influence on fistula formation. Cohen et al demonstrated better outcomes with a Langenbeck repair than with the Wardill closure.¹⁸ The Sommerlad repair technique has a published fistula rate of 5% in native group and 16% in non-native group.²⁰ Bekerecioglu et al found no difference in fistula rate between 2- and 4-flap palatoplasty, whereas Dong et al found no difference in outcomes between Furlow and 2-flap palatoplasty.^{21,22} There was a significantly higher incidence of fistula in patients who underwent surgery using the Furlow double-opposing Z-plasty technique (12.1%) than in cases where the Busan modification (3.0%) or two-flap technique (2.0%) was used.³ In our study fistula rate was 3.92% which has similarity to study by Park et al (3.1%).³

Postoperative feeding is an important factor contributing to fistula formation and this feeding regimens following cleft palate surgery remain controversial. Studies have suggested that unrestricted feeding with liquid diet is appropriate immediately after surgery. Some authors advocate feeding with liquids postoperatively for 10 – 14 days followed by semisolid diet for the next three to four weeks, while others will continue with liquid diets for three weeks, transitioning to a semisolid diet for an additional

three weeks. In a study by I I Onah et al. showed a significant increase in wound breakdown in patients commenced early (one-three weeks) on semisolid/solid diets as compared to those commenced on semisolid/ solid diet later at one month. Particulate matter from semisolid/solid food adhere to the repair site could evoke inflammatory changes that impair wound healing and predispose to wound dehiscence, wound breakdown and subsequent fistula formation.¹⁰ In our study we allowed liquid foods for 4 weeks followed by semisolid for next 2 week in all cases. Plenty of water was advised after each feed. This feeding regimen reduced the fistula formation.

The present study analyzed the location of the fistula which revealed a greater incidence (8, 53.33%) in the hard palate (Pitsburg IV, V, VI) followed by the junction of the hard and soft palate (6, 40%) and the soft palate (1, 6.67%). This has similarity with many studies. Anterior palatal fistulas (65.57%) were most commonly reported by Mahajan et al, followed by middle (24.86%) and posterior (9.18%).¹¹ Study by Garg et al. showed fistula rate 9.6% and a significantly higher number of which were found in Veau Types III and IV. They performed 2 flap palatoplasty technique in 68.2% of these fistula patients.¹² Many studies have shown that greater cleft width is one of the main predictor in fistula formation.^{6,13} Fistula occurred most frequently at the junction of hard and soft palate in case of wide cleft because cleft palate is widest in this region, as well as mucosa atrophy, inadequacy of muscle bulk and insufficient muscle mobilization for closure at that central area, resulting in greatest tension at closure¹⁴ coupled with constant motion of the soft palate as it hinges on the hard palate during velopharyngeal function. Yarhere et al. showed most common site of fistula formation at the junction of hard and soft palate.¹⁵ In our study, for a wide cleft, we repair the nasal layer by using vomerine flap and adequate mobilization of mucoperiosteal flap was done. A prospective study performed by Losken et al reinforced the importance of skeletonization of the vascular pedicle during medialization of the mucoperiosteal flaps, aggressive undermining and mobilization of musculature, and meticulous 2-layer mattress suture closure.²³ In this study 1(6.67%) fistula occurred in a patient having wide cleft and it was at the junction of hard and soft palate.

This study had some limitations. It was a retrospective study. The uneven distribution of variables (sex, age at the

time of cleft repair, cleft lip, the extent of the cleft) among patients makes the interpretation of results of this study more complex. We only evaluated a few risk factors. More risk factors should be evaluated in future prospective studies to address these issues. In our study it was not possible to examine the size or shape of the fistula due to a lack of information in the records.

Conclusion

In conclusion, the incidence of fistula after primary cleft palate repair is a significant concern for both patients and surgeons. Understanding the factors that cause fistula formation is essential for achieving successful surgical outcomes. Two flap palatoplasty technique along with addressing other factors can reduce fistula formation. Further research is needed to develop strategies to reduce the risk of fistula and improve overall patient care.

Conflict of interest: None

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Original Article

Efficacy of Dexamethasone in Severe Dengue in Khulna Medical College Hospital – A Longitudinal Study

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Abstract

Background: Severe dengue is a major cause of hospitalization in Bangladesh, characterized by plasma leakage, shock, and organ dysfunction. Corticosteroids such as dexamethasone have been proposed to modulate immune-mediated capillary leak, but results are inconsistent.

Objective: To evaluate the efficacy of dexamethasone in clinical outcomes among patients with severe dengue patients admitted to Khulna Medical College Hospital.

Methods: This longitudinal study was conducted between June 2023 and May 2024. One hundred twenty patients meeting WHO 2009 criteria (1) for severe dengue were divided into two groups: Group A (n = 60) received standard management plus intravenous dexamethasone 0.6 mg/kg/day for 3 days, and Group B (n = 60) received standard supportive therapy alone. Data were collected on demographic, clinical, and laboratory parameters. Outcomes assessed included duration of fever, hospital stay, platelet count & shock recovery, and mortality. Statistical analysis was performed using SPSS v26. **Results:** Mean hospital stay was significantly shorter in the dexamethasone group (5.7 ± 1.8 days) compared with controls (7.3 ± 2.4 days, $p < 0.05$). Platelet recovery occurred earlier (4.1 ± 1.2 days vs 5.6 ± 1.7 days, $p < 0.01$). Shock and ICU requirement were lower in the dexamethasone group (8.3% vs 21.7%). Mortality difference was not statistically significant (3.3% vs 5.0%, $p = 0.61$). **Conclusion:** Short-course dexamethasone therapy appears to accelerate clinical recovery and reduce complications in severe dengue without increasing adverse events. Larger randomized trials are warranted.

Keywords: Dengue, Dexamethasone, Corticosteroids, Severe dengue, Bangladesh

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Introduction

Dengue fever, caused by the Dengue virus (Flaviviridae), remains endemic in Bangladesh and Southeast Asia. Severe dengue manifests as plasma leakage, bleeding, and organ dysfunction.¹ Pathogenesis involves a cytokine storm and increased capillary permeability leading to hypovolemic shock². Corticosteroids such as dexamethasone may suppress this exaggerated immune response,

stabilizing endothelial function³. However, international evidence is mixed- some studies report clinical improvement while others find no mortality benefit²⁻⁵. Local data from Bangladesh are limited⁶. This study evaluates the therapeutic efficacy and safety of dexamethasone in severe dengue cases at Khulna medical college hospital, a major tertiary center in southern Bangladesh.

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Materials and Methods

A prospective longitudinal study was performed at Khulna medical college hospital, Department of Medicine, from June 2023 to May 2024. A total of 120 adult patients (13–60 years) diagnosed with severe dengue using WHO 2009 criteria were included.

Inclusion criteria

- Clinically diagnosed & laboratory confirmed dengue case
- Meeting the criteria of severe dengue
 - a. Severe plasma leakage leading to:

Shock (DSS)

Fluid accumulation with respiratory distress

- b. Severe bleeding as evaluated by clinician
- c. Severe organ involvement

Liver: AST or ALT ≥ 1000

CNS: Impaired consciousness

Heart and other organs

Exclusion criteria:

- Pregnancy
- Diabetes
- Chronic liver disease

Patients were divided into two groups

- Group A (Intervention): Intravenous dexamethasone 0.6 mg/kg/day in divided doses for 3 days + standard supportive therapy.
- Group B (Control): Standard supportive therapy only (fluid management, antipyretics, monitoring).

Demographics, clinical findings, hematocrit, platelet count, liver enzymes, complications were recorded at admission and all these parameters were evaluated at 72 hours and outcomes were also recorded. Data were analysed using SPSS v26. $p < 0.05$ was considered as significant.

Results

The baseline characteristics of the study population were comparable between the Dexamethasone and Control groups. There were no significant differences in age, sex distribution, hematocrit, platelet count, or ALT levels (all $p > 0.05$), indicating that both groups were well matched prior to treatment.

In terms of clinical outcomes, patients receiving Dexamethasone showed significantly faster recovery compared to the Control group. The mean duration of fever was shorter in the Dexamethasone group (4.5 ± 1.4 days) than in the Control group (5.9 ± 1.8 days, $p < 0.01$). Similarly, the average hospital stay was reduced (5.7 ± 1.8 vs. 7.3 ± 2.4 days, $p < 0.05$), and platelet recovery occurred earlier (4.1 ± 1.2 vs. 5.6 ± 1.7 days, $p < 0.01$). The need for intensive care was also significantly lower in the Dexamethasone group (8.3%) compared to the Control group (21.7%, $p = 0.04$). However, there was no significant difference in mortality between the two groups (3.3% vs. 5.0%, $p = 0.61$).

With regard to complications, the incidence of shock, hepatic dysfunction, and bleeding was lower among patients treated with Dexamethasone compared to controls (10% vs. 23%, 13% vs. 17%, and 15% vs. 20%, respectively). Nonetheless, hyperglycemia was more frequent in the Dexamethasone group (12%) and absent in the Control group, while secondary infection rates were similar between groups (5% vs. 3%).

Overall, Dexamethasone therapy was associated with faster clinical improvement, shorter hospitalization, earlier platelet recovery, and reduced ICU requirement, without an increase in mortality. However, the treatment was accompanied by a higher incidence of hyperglycemia, a known corticosteroid-related adverse effect.

Table I. Baseline Characteristics

Parameter	Dexamethasone (n = 60)	Control (n = 60)	p value
Age (years)	32.8 ± 9.6	33.4 ± 10.1	0.72
Male : Female	35 : 25	33 : 27	0.68
Hematocrit (%)	44.3 ± 5.1	44.7 ± 4.8	0.79
Platelet ($\times 10^9/L$)	49 ± 21	52 ± 18	0.54
ALT (U/L)	83 ± 39	86 ± 43	0.81

Table II. Outcomes

Outcome	Dexamethasone (n = 60)	Control (n = 60)	p value
Duration of fever (days)	4.5 ± 1.4	5.9 ± 1.8	< 0.01
Hospital stay (days)	5.7 ± 1.8	7.3 ± 2.4	< 0.05
Platelet recovery (days)	4.1 ± 1.2	5.6 ± 1.7	< 0.01
ICU requirement	5 (8.3%)	13 (21.7%)	0.04
Mortality	2 (3.3%)	3 (5.0%)	0.61

Table III. Complications

Complication	Dexamethasone (n = 60)	Control (n = 60)
Shock	6 (10%)	14 (23%)
Hepatic dysfunction	8 (13%)	10 (17%)
Bleeding	9 (15%)	12 (20%)
Hyperglycemia	7 (12%)	0 (0%)
Secondary infection	3 (5%)	2 (3%)

Discussion

Pathophysiologically, corticosteroids reduce pro-inflammatory cytokines (IL-6, TNF- α), inhibit T-cell activation, and decrease endothelial permeability⁷. These mechanisms likely explain the shorter hospital stay and faster hematological recovery observed here.

This study demonstrates that short-course dexamethasone accelerates clinical recovery and reduces complications in severe dengue, though mortality reduction was not statistically significant.

Our findings align with Jayashree et al.², who found improved recovery in children with severe dengue treated with corticosteroids. Nguyen et al.⁵ reported similar platelet recovery benefits, while Hossain et al.⁶ observed reduced fluid requirements in Bangladeshi adults.

Conversely, Tam et al.⁴ and Lye et al.⁸ found no survival benefit, possibly due to delayed steroid initiation and heterogeneous disease severity. Meta-analysis by Tran et al.³ concluded that corticosteroids may prevent progression to shock if started early, supporting our results.

Safety outcomes were consistent with many studies 8-10 who found no major infection risk with brief steroid courses, and Lum et al.¹¹, who reported transient hyperglycemia but no mortality impact.

Dengue outbreaks in Bangladesh are increasing^{12, 13} and therapeutic options remain limited. so this study is time demanding.

Limitations

Single-center design, limited sample size, and absence of cytokine or viral load data. Larger multicenter randomized controlled trials are necessary.

Conflicte of Interest : None

Conclusion

Dexamethasone given early in severe dengue appears to reduce complication rates and hospitalization time without increasing mortality or severe adverse events. Its use as an adjunct to standard care could be beneficial in resource-limited settings, but confirmation through larger trials is essential. A low-cost adjunct like dexamethasone may be valuable, particularly in tertiary hospitals with high caseloads.

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Original Article

Comparison of Carbetocin and Oxytocin in Active Management of the Third Stage of Labour Following Vaginal Delivery at Jashore CMH

Karim R^{1*}, Islam MA², Nargis M³, Sarmen S⁴**Abstract**

Background: Postpartum hemorrhage (PPH) is the foremost global cause of maternal mortality, especially in resource-limited settings. Preventing PPH relies on actively managing labor's third stage (AMTSL). While oxytocin is standard, its heat sensitivity is problematic. Carbetocin, a stable, long-acting synthetic analog, presents a potentially superior alternative.

Objective: To assess the efficacy of a single intravenous dose of carbetocin with 10 IU oxytocin in preventing PPH in women having a vaginal delivery with AMTSL.

Methods: This randomized controlled trial was carried out in the Department of Obstetrics and Gynaecology at CMH Jashore between October 2024 and July 2025. One hundred eligible full-term, singleton pregnant women were randomly assigned to receive intravenous carbetocin (100 mcg, Group A) or intravenous oxytocin (10 IU, Group B). Primary endpoints assessed were average blood loss and PPH occurrence (blood loss exceeding 500 mL). Secondary outcomes measured significant hemorrhage, additional uterotonic requirements, transfusion rates, and adverse events.

Results: No cases of postpartum hemorrhage (PPH) occurred in patients administered carbetocin. Conversely, PPH developed in 8.2% of the oxytocin group. Average blood loss was 350 mL with carbetocin, significantly less than the 450 mL observed with oxytocin. Requirement for supplemental uterotonics, uterine massage, or transfusions was exclusive to the oxytocin group. Adverse events were minimal and similar between the cohorts.

Conclusion: Carbetocin demonstrates superior efficacy in minimizing postpartum hemorrhage (PPH) and decreasing blood loss compared to oxytocin. Its extended duration of effect and thermostability offer a pragmatic advantage for active labor management, particularly in settings with limited medical resources.

Keywords: Postpartum haemorrhage, AMTSL, carbetocin, oxytocin, randomized trial.

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Introduction

Postpartum haemorrhage (PPH) remains a principal contributor to maternal death worldwide, disproportionately impacting low-resource settings.¹ Even with modern obstetric practices, the third stage of labour constitutes a critical window for potential severe haemorrhage.² The active management of this stage (AMTSL) is a cornerstone strategy for PPH prevention, employing the prompt administration of a uterotonic drug, controlled traction of the umbilical cord, and immediate cord clamping.³

The current gold-standard uterotonic, oxytocin, has well-documented limitations. Its brief therapeutic activity and susceptibility to degradation at elevated temperatures can compromise efficacy, especially in environments where consistent refrigeration is unreliable.^{4,5} This presents a significant practical barrier in many healthcare facilities, potentially undermining the benefits of established AMTSL protocols. Carbetocin, a synthetic long-acting oxytocin

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analogue with inherent heat stability, has emerged as a potentially more reliable agent for routine AMTSL.⁶ Its pharmacological profile suggests it could offer a more sustained uterine contraction and a practical logistical advantage, particularly in settings with constrained resources or challenging storage conditions.

This investigation, conducted at Jashore Combined Military Hospital, directly compares the clinical performance of intravenous carbetocin versus oxytocin in the context of AMTSL for preventing excessive blood loss after vaginal birth. The study aims to determine if the theoretical advantages of carbetocin translate into superior real-world outcomes by measuring key indicators such as mean blood loss, the incidence of PPH, and the need for additional interventions. Evaluating these two agents within a specific clinical setting is crucial for informing local and regional guidelines, optimizing resource allocation, and ultimately strengthening efforts to reduce preventable maternal morbidity and mortality associated with postpartum bleeding.

Methodology

This randomized controlled trial was conducted in the Department of Obstetrics and Gynaecology at CMH Jashore between October 2024 and July 2025.

Study population

A cohort of 113 women with full-term (≥ 37 weeks), singleton pregnancies, scheduled for vaginal birth, was initially enrolled. Following screening, thirteen participants were excluded for the following indications: pre-eclampsia (4 cases), eclampsia (3 cases), multifetal gestation (3 cases), or significant anemia (3 cases). Consequently, the final study population comprised 100 parturients, who were randomly allocated to one of two treatment arms:

- Group A (Carbetocin): 50 women received carbetocin 100 mcg IV, single dose after delivery.
- Group B (Oxytocin): 50 women received oxytocin 10 IU IV after delivery.

Inclusion criteria

- Women with singleton pregnancies at ≥ 37 weeks
- Planned vaginal delivery
- Informed consent obtained

Exclusion criteria

- Placenta previa or placental abruption
- Multiple pregnancy
- Hypertensive disorders (preeclampsia/eclampsia)
- Cardiac, renal, hepatic disease, epilepsy
- Moderate to severe anemia
- Refusal to participate

Outcome measures

Primary outcome: Blood loss within 24 hours postpartum (quantified using pre-weighed sanitary pads and calibrated MacIntosh sheets).

Secondary outcomes: Incidence of PPH, massive blood loss, requirement for additional uterotonics, need for blood transfusion, and adverse effects.

Data analysis

Statistical analysis was performed with SPSS (version 16). Continuous variables are presented as mean $\pm$ standard deviation (SD). Group comparisons for categorical data were conducted using the Chi-square test or Fisher's exact test, as appropriate. A result was deemed statistically significant if the p-value was below 0.05. The study protocol received ethical clearance from the Institutional Ethical Review Committee of CMH, Jashore.

Results

Baseline characteristics

The average maternal age was 23.9 years (± 3.2 SD) for participants receiving carbetocin and 23.3 years (± 3.2 SD) for those administered oxytocin. Gestational age at the time of delivery did not differ significantly, with means of 38.01 (± 1.1) weeks and 38.09 (± 1.7) weeks, respectively. Pre-delivery measurements, including hemoglobin concentration, blood pressure values, and the rate of anemia, were equivalent between the two study cohorts.

Table-I: Baseline characteristics of study participants (n=100).

Variable	Carbetocin group(n=50)	Oxytocin group(n=50)	p-value
Mean age (years)	23.9 $\pm$ 3.2	23.3 $\pm$ 3.2	0.42
Mean gestational age (weeks)	38-40	38-40	
Mild anemia (%)	17 (36.1%)	20 (42.6%)	0.55
Mean systolic BP (mmHg)	112 $\pm$ 5.6	110 $\pm$ 1.7	0.09
Mean diastolic BP (mmHg)	75 $\pm$ 4.3	72 $\pm$ 11.5	0.11

Outcomes of AMTSL

Women who received carbetocin experienced no instances of massive hemorrhage, postpartum hemorrhage (PPH), or the requirement for supplementary medical interventions. Conversely, the group administered oxytocin demonstrated a greater frequency of these undesirable clinical outcomes (Table-II).

Table-II: Outcomes of the third stage of labour (n=100).

Outcome	Carbetocin group(n=50)	Oxytocin group(n=50)	p-value
Massive blood loss (%)	0 (0.0)	3 (6.2%)	<0.05
Fundal massage needful (%)	0 (0.0%)	4 (8.2%)	<0.05
Blood transfusion needed (%)	0 (0.0%)	5 (10.4%)	<0.05
Additional uterotonics needed (%)	0 (0.0%)	5 (10.6%)	<0.05
Primary PPH (%)	0 (0.0%)	4 (8.2%)	<0.05

Table-III: Adverse effects observed within 24 hours (n=100).

Adverse effect	Carbetocin group (n=50)	Oxytocin group (n=50)	p-value
Nausea (%)	2 (4.0%)	3 (6.0%)	0.64
Vomiting (%)	1 (2.0%)	2 (4.0%)	0.56
Headache (%)	1 (2.0%)	1 (2.0%)	1.00
Tremor (%)	0 (0.0%)	1 (2.0%)	0.31
Hypotension (%)	0 (0.0%)	1 (2.0%)	0.31
Pruritus (%)	1 (2.0%)	1 (2.0%)	1.00
Abdominal pain (%)	1 (2.0%)	2 (4.0%)	0.56
Fever (%)	0 (0.0%)	1 (2.0%)	0.31
Arrhythmia / Pulmonary edema	0 (0.0%)	0 (0.0%)	—

No major adverse effects were reported in either group. Minor side effects were comparable between groups. No statistically significant differences were observed ($p>0.05$).

Table-IV: Mean blood loss within 24 hours postpartum (n=100).

Group	Mean blood loss (mL)	p-value
Carbetocin (n=50)	350 mL	<0.05
Oxytocin (n=50)	450 mL	

Mean blood loss within 24 hours was significantly lower in the carbetocin group.

Table-V: Incidence of primary postpartum hemorrhage (PPH) (n=100).

Group	PPH (%)	p-value
Carbetocin (n=50)	0.0%	<0.05
Oxytocin (n=50)	4(8.2%)	

Discussion

The study population presented with a mean age of 23.9 years (± 3.2 SD) in the carbetocin cohort and 23.3 years (± 3.2 SD) in the oxytocin group. This demographic profile is younger than that reported in the study by Laciprete et al.⁵ Preoperative systolic and diastolic blood pressure measurements were within the normal range for participants in both arms, a finding consistent with observations made by Debbie-Lynuy and Nelindac Atherinep et al.⁷

The results indicate that carbetocin confers superior protection against postpartum hemorrhage (PPH) when utilized in active management of the third stage of labor (AMTSL). This is evidenced by significantly reduced measured blood loss and a marked decrease in the necessity for additional therapeutic interventions. Notably, no participant receiving carbetocin required a blood transfusion, whereas this intervention was needed for 10.4% of the oxytocin group. This pattern aligns with the work of Manal M. E. Behery et al., who reported transfusion rates of 0% and 15.5% in their respective carbetocin and oxytocin groups.⁹ Furthermore, the requirement for supplemental uterotonic agents was exclusive to the oxytocin group, affecting 10.6% of its participants, a rate lower than the 34.3% noted by Debbie-Lynuy et al.⁷

The need for therapeutic fundal massage was absent in the carbetocin arm but was required for 8.2% of patients administered oxytocin. This trend, though less pronounced, is directionally consistent with the substantial disparity (10% vs. 83%) reported by Agnes P. Monteo-Fenix et al. in a study of high-risk patients.⁸ The mean blood loss difference of 100 mL (350 mL vs. 450 mL for carbetocin and oxytocin, respectively) observed in the present study represents a clinically meaningful margin, corroborating the findings of Sergio Rosales-Ortiz et al. and Ahmed Mohamed Maged, who documented similar volumetric advantages for carbetocin.^{10, 11}

The observed reduction in blood loss is a critical determinant of clinical outcomes. Minimizing hemorrhage directly correlates with a lower incidence of severe PPH, decreased requirements for blood transfusions, and a reduced need for invasive rescue interventions such as uterine artery embolization or emergency hysterectomy.¹²

This preservation of maternal blood volume is essential for preventing related sequelae, including hemodynamic instability, coagulopathy, and end-organ dysfunction, thereby significantly lowering maternal morbidity.¹³ The consistent demonstration of carbetocin's superior hemostatic effect across diverse study populations strengthens the evidence for its role as a more reliable first-line uterotonic.

The significant blood loss reduction with carbetocin is attributable to its distinct pharmacodynamic profile. Unlike oxytocin, which has a short half-life and often requires repeat dosing or continuous infusion to maintain uterine tone, carbetocin provides a sustained and potent contraction for over an hour.¹⁴ This prolonged action more effectively counteracts uterine atony—the leading cause of PPH—during the most vulnerable period immediately following placental delivery.¹⁵ Furthermore, its proven stability at ambient temperatures ensures full pharmacological potency is delivered in real-world settings where cold chain maintenance for oxytocin can be unreliable, translating theoretical efficacy into guaranteed clinical performance.¹⁶

It is important to acknowledge the constraints of this investigation, including its single-center design and limited sample size. Future research employing larger, multicenter trials would be invaluable in confirming these findings and enhancing their generalizability to broader obstetric populations.

Conclusion

Carbetocin demonstrates greater efficacy than oxytocin for preventing postpartum hemorrhage (PPH) and minimizing total blood loss after vaginal birth. Its extended pharmacological effect and heat-stable properties render it a suitable agent for the routine active management of the third stage of labour (AMTSL), especially in healthcare environments with constrained or unreliable refrigeration capabilities. Further investigation through larger clinical trials and formal cost-benefit assessments is advised to substantiate its broader adoption into clinical practice.

Conflict of interest: None

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Case Report**Bilateral Hirayama Disease Presenting with Long-Standing Weakness and Wasting of the Hands and Forearms: A Rare Case Report**Sadeque ABMZ¹, Islam MA², Arman M³**Abstract**

Hirayama disease (HD), or juvenile muscular atrophy of the distal upper extremity, is an uncommon motor neuron disorder, usually presenting with unilateral hand and forearm wasting in young males. Bilateral involvement is rare and may mimic motor neuron disease.

A 34-year-old Bangladeshi male presented with weakness and wasting of both hands and forearms, which began insidiously at age 17, progressed for six months, and then stabilized for 17 years. Symptoms worsened during cold weather, impairing fine motor activities. Neurological examination showed bilateral distal atrophy, weakness of intrinsic hand muscles, reduced grip strength, and preserved reflexes and sensation. Electromyography demonstrated chronic denervation in C7–T1 myotomes. Dynamic cervical MRI revealed anterior displacement of the posterior dura and lower cervical cord compression, diagnostic of Hirayama disease. The patient was managed with a multidisciplinary rehabilitation program, including neuromuscular stimulation, hand exercises, occupational therapy, orthoses, and vocational reintegration. This case emphasizes the importance of considering Hirayama disease in young males presenting with distal upper limb wasting, even when bilateral. Early recognition with dynamic MRI and multidisciplinary rehabilitation can optimize functional outcomes and prevent misdiagnosis.

Keywords: Hirayama disease, monomelic amyotrophy, distal upper limb atrophy, dynamic MRI, rehabilitation.

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Introduction

Hirayama disease (HD) is a rare, benign motor neuron disorder that primarily affects adolescent and young adult males. It is characterized by insidious onset of weakness and wasting of the hand and forearm muscles, usually unilaterally, with relative sparing of the brachioradialis, leading to the so-called “oblique amyotrophy” appearance^{1,2}. First described in 1959 by Hirayama¹, the condition is most commonly reported in Asian countries such as Japan, India, and China^{2,3}, although sporadic cases have been described worldwide¹¹.

The proposed pathophysiology involves disproportionate growth between the spinal canal and dura mater during adolescence, leading to forward displacement of the posterior dura during neck flexion and subsequent ischemia of the lower cervical anterior horn cells^{5,9}. This dynamic pathogenesis explains why flexion MRI is crucial for diagnosis^{6–8,14}.

Although most cases are unilateral and stabilize within 3–5 years (2), bilateral presentations have been reported in up to

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20% of patients¹⁰. These atypical cases are frequently misdiagnosed as amyotrophic lateral sclerosis (ALS), cervical spondylotic myelopathy, or peripheral neuropathy¹³. We report a case of long-standing bilateral Hirayama disease in a Bangladeshi male, managed with a comprehensive rehabilitation program.

Case Presentation

A 34-year-old right-handed male farmer presented with weakness and wasting of both hands and forearms for 17 years. Symptoms began at age 17, progressing gradually over six months before stabilizing. He reported worsening of weakness during cold exposure. Functional impairment included difficulty in eating, grooming, dressing, writing, and gripping tools.

There was no history of trauma, sensory symptoms, cranial nerve involvement, bulbar dysfunction, or systemic illness. Family history was Unremarkable.

On Examination, the patient was of below-average build with a BMI 24.2kg/m². Vitals sign were within normal limit. Examination revealed bilateral wasting of distal forearm and intrinsic hand muscles (Fig 1a, b) exhibiting the characteristics of 'Oblique amyotrophy' pattern. Grip strength measured by Modified Sphygmomanometer (MST) Test was markedly reduced (right 10 mmHg, left 20 mmHg). Weakness was most pronounced in finger flexion/extension and thumb movements (MRC grade 2–3/5 distally, 4/5 at wrist, 5/5 proximally). Deep Tendon reflexes were preserved, and plantar responses were flexor, sensation intact, and coordination normal. Other systems findings were normal.



Fig 1a : Wasting of hand muscle



Fig 1b : Wasting of hand muscle

Routine laboratory investigations including Hematology and biochemistry parameters within normal limits, Creatine kinase: 55 U/L. NCS/EMG (Fig 3) finding was chronic denervation in C7–T1 myotomes, consistent with anterior horn cell disorder. MRI of cervical spine (neutral and flexion) findings were forward displacement of posterior dura, venous engorgement, and lower cervical cord atrophy (Fig 2) diagnostic of Hirayama disease. X-ray cervical spine finding was unremarkable.

The patient was independently ambulant with modified independence in ADLs. Fine motor activities such as writing and buttoning were significantly impaired. Functional Independence Measure (FIM) score was 120.

The patient was managed with a multidisciplinary rehabilitation team approach. Education & counseling: Regarding benign, non-progressive nature of disease. Physiotherapy: Range-of-motion, stretching, and resistance hand exercises; tendon-gliding and elastic-band training. Neuromuscular electrical stimulation (NMEST): Over finger flexors and intrinsic hand muscles. Occupational therapy: Task-oriented training for ADLs, fine motor retraining, and activity modification. Orthoses & assistive devices: MCP hyperextension restriction splint and wrist-hand orthosis. Vocational rehabilitation: Adaptations for farming. Psychological support: Counseling to address anxiety and reduce unnecessary medical consultations.

Management

Collar therapy is effective in the early progressive phase by preventing neck flexion (15). Surgical interventions (anterior decompression, posterior fixation) are reserved for

rapidly progressive cases (16). For stable long-term cases, rehabilitation is the cornerstone. Rehabilitation strategies — including ROM, resistive exercises, electrical stimulation, orthoses, and vocational training — can maintain independence and quality of life^{17,18}.

Our patient, with a 17-year stable course, was managed conservatively with rehabilitation. His functional goals focused on ADL independence, fine motor improvement, and vocational reintegration.

Discussion

HD is often mistaken for more aggressive motor neuron diseases, especially when bilateral^{10,13}. In contrast to ALS, HD stabilizes after a few years and lacks upper motor neuron signs or bulbar involvement^{2,9}.

Typical onset occurs during adolescence, with a male predominance (male: female $\approx$ 20:1)². Unilateral distal upper limb weakness and wasting are hallmarks^{1,3}, but bilateral cases such as ours account for 10–20%¹⁰. Cold paresis, reported by our patient, is another supportive feature².

Electrodiagnostic findings include denervation in C7–T1 myotomes with preserved sensory conduction^{3,6}. Dynamic MRI is diagnostic, showing forward displacement of the posterior dura, loss of dural attachment, venous plexus engorgement, and focal cord atrophy^{6–8,14}. Our patient's MRI demonstrated classic findings.

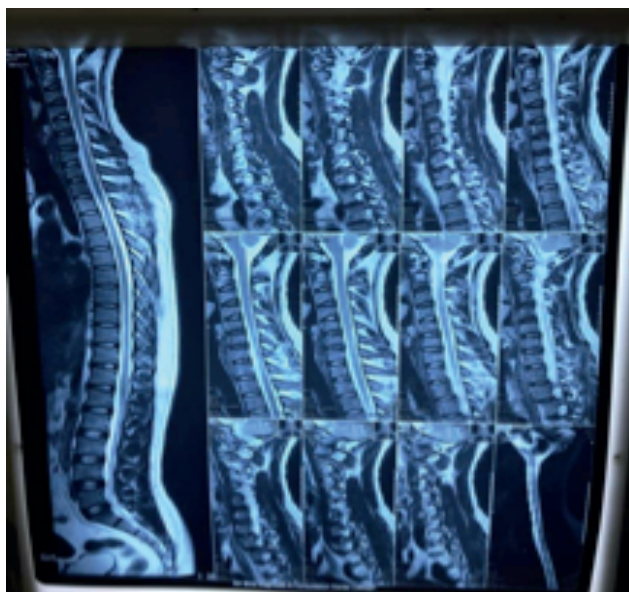


Fig 2 : MRI of spine

Patient Information

ID	M2886	In/Out Patient	Out
Name	Mr. Ataur Rahman	Refer. Dept.	B.S.M.M.U.
Date of Birth		Physician	
Age	30 Y	Examination Date	5/11/2019
Sex	Male	Examination No.	
Height		Examined by	
Weight			

Motor Nerve Conduction Study

Site	Latency (ms)	Amplitude	Area	Segment	Distance (mm)	Interval (ms)	NCV (m/s)	NCV N.D.
Median, L								
Wrist	4.34ms	9.62mV	23.23mVms	Wrist		4.34ms		
Elbow	9.06ms	9.14mV	22.96mVms	Wrist - Elbow	230mm	4.72ms	48.7m/s	
Ulnar, L								
Wrist	2.7ms	1.22mV	1.91mVms	Wrist		2.70ms		
Elbow	2.26ms	1.38mV	3.77mVms	Wrist - Elbow	270mm	5.26ms	51.3m/s	
Median, R								
Wrist	6.32ms	2.53mV	6.89mVms	Wrist		6.32ms		
Elbow	0.46ms	2.85mV	7.84mVms	Wrist - Elbow	230mm	4.14ms	55.6m/s	
Ulnar, R								
Wrist								
Elbow								

F-wave Study

Nerve	Stim. Site	F-Lat.	F-Lat. N.D.	M Lat.	F-M Lat.	F-Occurs.	Distance	FWCV	N.D.
Median, L	L Wrist	32.9ms				37.4%			
Ulnar, L	L Wrist	29.75ms				27.2%			
Median, R	R Wrist	29ms				216.13%			
Ulnar, R	R Wrist					314.14%			

Sensory Nerve Conduction Study

Site	Latency (ms)	Amplitude	Area	Segment	Distance (mm)	Interval (ms)	NCV (m/s)	NCV N.D.
Median, L								
Wrist	2.94ms	50.10uV	5.56uVms	Wrist		2.94ms		
Ulnar, L								
Median, R	2.96ms	32.50uV	1.52uVms	Wrist		2.96ms		
Ulnar, R								
	2.88ms	38.40uV	1.39uVms	Wrist		2.88ms		

EMG Findings Summary

Muscle-Side	Int. Act.	Fibs.	Pos. Wave	Fasc.	MYO Disch.	Normal MUP	Poly	Low Amp.	High Amp.	Dur.	Recruit	Int. Pat.
Extm. Carpi Radialis L										Normal		
Flex. Carpi Radialis L										Normal		
Flex. Carpi Radialis R										Normal		
Triceps L									+1	Long	Reduce	
Extm. Indicia Pro L									+1	Long	Reduce	
Extm. Indicia Pro R									+1	Long	Disrec	
Abduc. Pol. Brevis R									+1	Long	Disrec	
Abduc. Pol. Brevis L									+1	Long	Disrec	
1st Dorsal Inter. L									+1	Long	Disrec	
1st Dorsal Inter. R									+1	Long	Disrec	

Fig 3 : NCS/EMG Findings

Differential diagnosis include: Cervical spondylotic myelopathy: Associated with sensory involvement and UMN signs. Syringomyelia: Cape-like sensory loss. Ulnar/median neuropathy: Sensory deficits and focal nerve tenderness. ALS: Progressive, diffuse wasting with UMN signs (13). The absence of sensory or pyramidal signs favored HD.

Conclusion

This case demonstrates an atypical bilateral presentation of Hirayama disease with long-standing stability. It underscores the importance of dynamic MRI in diagnosis and differentiating from motor neuron disease. Early recognition prevents misdiagnosis and inappropriate treatment, while multidisciplinary rehabilitation is essential in managing chronic deficits and improving quality of life.

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