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Evaluation of Factors Affecting Morbidity in Patients with Osteogenesis Imperfecta

Osteogenezis İmperfekta Hastalarında Morbiditeyi Etkileyen Faktörlerin Değerlendirilmesi

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ABSTRACT

Objective: Osteogenesis imperfecta (OI) is a rare genetic disease characterized by osteoporosis and fragility of the bones. These patients often require anesthesia for orthopaedic surgery because of recurrent bone fractures. Our primary goal in this study was to evaluate the patients operated for OI to determine the factors that are associated with perioperative and postoperative morbidity.

Methods: The patient files were retrospectively evaluated between 2008 and 2018. Demographic data, number and type of operations, the duration of the last operation, type of anesthesia, perioperative position, perioperative and postoperative fracture formation, and amount of bleeding were recorded. Additionally, the need for intensive care, if any, length of intensive care unit stays, and length of hospital stay were determined, and the effect of these factors on morbidity and mortality was investigated.

Results: In this study, 44 patients with OI, who had undergone 105 operations, were included with a mean age of 11.07±7.70 years. We demonstrated that the presence of scoliosis (p=0.001), body mass index measurements (p=0.008), and higher number of operations (p=0.014) were significantly associated with morbidity. However, when we made the regression model, we reported that only the presence of scoliosis, appeared to be a significant model (p=0.002; odds ratio: 9.082). Scoliosis increased the risk of morbidity 9.082-fold, which was an independent risk factor.

Conclusion: In our study, we demonstrated that the scoliosis had an effect that increases the risk of morbidity 9-fold, and that scoliosis is an independent risk factor. Considering these data, we suggest that all OI patients undergoing orthopaedic surgery should be radiologically screened for the presence of scoliosis before the operation.

Keywords: Osteogenesis imperfecta, anesthesia, scoliosis, orthopaedic procedures

ÖZ

Amaç: Osteogenezis imperfekta (OI), kemiklerin kırılganlığı olan osteoporoz ile karakterize nadir görülen bir genetik hastalıktır. Bu hastalar sıklıkla tekrarlayan kemik kırıkları nedeniyle ortopedik cerrahi için anestezi gerektirir. Bu çalışmadaki birincil amacımız, OI nedeniyle opere edilen hastaları değerlendirmek, perioperatif ve postoperatif morbidite ile ilişkili faktörleri belirlemekti.

Yöntemler: 2008-2018 yılları arasındaki hasta dosyaları retrospektif olarak incelendi. Demografik veriler, operasyon sayısı ve tipi, son operasyon süresi, anestezi tipi, perioperatif pozisyon, perioperatif ve postoperatif kırık oluşumu ve kanama miktarı kaydedildi. Ayrıca varsa yoğun bakım ihtiyacı, yoğun bakımda kalış süresi ve hastanede kalış süresi belirlenerek bu faktörlerin morbidite ve mortaliteye etkisi araştırıldı.

Bulgular: Bu çalışmaya 105 operasyon geçirmiş Oİ'li 44 hasta dahil edildi ve yaş ortalaması 11,07±7,70 idi. Skolyoz varlığının (p=0,001), vücut kitle indeksi ölçümlerinin (p=0,008) ve operasyon sayısının fazlalığının (p=0,014) morbidite ile anlamlı ilişkili olduğu gösterildi. Regresyon analizinde sadece skolyoz varlığının anlamlı bir model olduğu bildirildi (p=0,002; olasılık oranı: 9,082). Bağımsız bir risk faktörü olan skolyoz, morbidite riskini 9,082 kat artırdı.

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Sonuç: Bu çalışmada skolyozun morbidite riskini 9 kat artıran bir etkisinin olduğu ve skolyozun bağımsız bir risk faktörü olduğu gösterildi. Bu veriler ışığında ortopedik cerrahi geçiren tüm Oİ hastalarının operasyon öncesi radyolojik olarak skolyoz varlığı açısından taranması önerildi.

Anahtar kelimeler: Osteogenezis imperfekta, anestezi, skolyoz, ortopedik işlemler

INTRODUCTION

Osteogenesis imperfecta (OI) is a disease with a prevalence of about 6-7:100,000 births (1). OI is a rare connective tissue disease that develops secondary to the structural or synthesis disorder of collagen and the manifests in children with diffuse osteoporosis, fragility of the bones, fractures and deformities (2). Although the main defect in these patients is in bone tissue, many systems such as skin, ligaments, tendons, sclera, nose and ear can also be affected (3).

Patients diagnosed with OI often require anesthesia for surgical interventions due to recurrent bone fractures. Accurate identification of risk factors and optimization of general medical conditions before surgery is essential for a smooth course of anesthesia (4). Pulmonary complications secondary to kyphoscoliosis have been suggested to be quite common in severe OI patients (5). Skeletal anomalies causing anatomical deformation of the airway may complicate tracheal intubation (6). Neck and mandible fractures may be seen during laryngoscopy, and the clinical picture may result in posterior brain herniation by causing basilar invagination, resulting in disruption of blood and cerebrospinal fluid flow (7). Increased bone fragility is an important consideration for the anesthesiologist. In these patients, there is a 10-30% incidence of bleeding diathesis. Studies have reported that blood loss increases with increased duration of operation and the increased number of osteotomies (8). Aortic root dilatation and left-sided valve failure are the most reported cardiac pathologies in the literature (9). A closer follow-up may be necessary when severe perioperative complications such as bleeding, fracture, and pulmonary symptoms are present (10).

The cause of mortality in milder types (types I and IV) may be due to conditions such as myocardial infarction and malignancy, which are not associated with the components of the disease, for more severe types, such as type III, respiratory and neurological problems or cardiac failure due to kyphoscoliosis have been reported in the literature to cause death in a considerable extent (11).

Although it is an important challenge in anesthesia, the number of studies in the literature for patients with OI is limited. Therefore, we intended to clarify the factors associated with morbidity and mortality to have a deeper understanding of patients with OI patients. Our primary goal in this study was to retrospectively evaluate the OI-diagnosed patients operated for orthopaedic surgery and to determine the factors associated with morbidity. The secondary aim of this study was to report factors associated with mortality.

METHODS

This retrospective study was performed following the approval of the Ethics Committee of İstanbul University, İstanbul Faculty of Medicine (decision no: 12, date: 29.06.2018). Patients who were operated at the Orthopedics and Traumatology Clinic of İstanbul University, İstanbul Faculty of Medicine between the years of 2008-2018 with complete medical records and ≤18 years of age and who did not have missing follow-ups were included to the study. Adults, patients with missing files, non-orthopaedic surgery patients, and patients who refused the operation were excluded from the study. Archived files, perioperative anesthesia forms, discharge information in the hospital registry system, and radiological imaging were used to obtain the data. Long-term follow-up information was obtained from the patients themselves and their relatives through the contact information of the patients. Age and comorbidities of the patients and preoperative hemogram, coagulation parameters, and biochemical results were recorded. The incidence of comorbidities was determined. The degrees of Cobb angle of all patients were also recorded. Scoliosis was defined as the frontal plane curve of >10° measured by the Cobb angle. Demographic data, number and type of operations, the duration of the last operation, type of anesthesia, perioperative position, perioperative and postoperative fracture formation, and amount of bleeding were recorded. Additionally, factors such as the presence of the need for intensive care stay, length of intensive care unit (ICU) stay, and hospital stay were determined. The effects of all these data on morbidity and mortality were subjected to statistical analyses.

We defined morbidity as perioperative and postoperative complications, post-operative respiratory distress, musculoskeletal (walking disorder or assisted walking due to muscle and joint deformities, peripheral nerve symptoms), and neurological (central nerve injury) complications.

Patients who had general anesthesia for OI in this study undergo tracheal intubation by administration of 2-3 mg/kg propofol, 1-2 µg/kg fentanyl and 0.6 mg/kg rocuronium and maintenance was provided by 1-MAC sevoflurane.

Statistical Analysis

The NCSS 2007 software was used for the statistical analyses. Descriptive statistical methods (mean, standard deviation, median, frequency, ratios, minimum and maximum) were used in the analysis of the data in addition to Student t-test and Mann-Whitney U tests for the two-group comparisons of the qualitative data with and without normal distribution, respectively. Enter

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logistic regression analysis was used to determine the effective risk factors for morbidity and mortality. Pearson chi-square test, Fisher-Freeman-Halton test, and Fisher's Exact test were used in the comparison of qualitative data. The level of significance was accepted as $p<0.05$.

RESULTS

Data of 51 patients who had undergone 113 operations were evaluated, and their eligibility for the study was investigated. Among these, 44 patients and 105 operations were included in the study. Six patients were excluded due to rejection of the operation and one patient due to missing file. Descriptive and preoperative clinical characteristics of the patients are given in Table 1. It was found that administration of general anesthesia was generally preferred by the staff in this hospital when anesthesia is needed in patients with OI. General anesthesia was applied to all patients except one who preferred spinal anesthesia.

The mean number of previous operations was 7.40±7.33. Among the postoperative patients, 6.7% were admitted to the ICU. The distribution of the operative and postoperative characteristics is given in Table 2. The mean Cobb angle in patients with scoliosis was 35.40±24.93.

The distribution of factors associated with postoperative morbidity and mortality is given in Table 3, and morbidity was detected in 37 operations.

When the effect of demographic data on morbidity was evaluated, no difference was observed between the groups in terms of age. Body mass index (BMI) measurements were statistically significantly lower in patients with morbidity. Additionally, morbidity was found to be statistically significantly higher in patients with scoliosis. When we investigated the effect of preoperative clinical features on morbidity; no statistically significant difference was found between the groups according to the results of international normalized ratio, number of platelets, and results of preoperative pulmonary function tests (PFTs) and position (Table 4).

No statistically significant effect of the type of operation, the type of anesthesia, duration of the last operation, presence of fracture, and presence of bleeding were found on morbidity. The number of operations the cases with morbidity (+) underwent was found to be statistically significantly higher compared with morbidity (-) cases ($p=0.014$). There was no statistically significant difference in morbidity according to the length of postoperative ICU stay and hospital stay ($p>0.05$) (Table 5).

Factors that were found to have a significant effect on morbidity such as BMI, presence of scoliosis, and the number of previous operations were evaluated by Enter logistic regression analysis (Table 6).

The variables included in the study were evaluated by Enter logistic regression analysis. The presence of scoliosis, one of the risk factors affecting the morbidity status, appears to form a significant model. The explanatory coefficient of the model was 69.6%. According to the model, the presence of scoliosis affected morbidity by increasing the risk by 9.082-fold. Scoliosis was an independent risk factor.

Table 1. Distribution of demographic data

Age (years)		11.07±7.70
BMI (kg/m²)		20.71±4.96
Coexisting disease n (%)	No	57 (54)
	Yes	48 (45.7)
	Scoliosis	14 (13.3)
	ASD	5 (4.7)
	Diabetes	4 (3.8)
	Meningomyelocele	4 (3.8)
	Cerebral palsy	3 (2.8)
	Epilepsy	3 (2.8)
	Hyperthyroidism	3 (2.8)
	Chronic bronchitis	2 (1.9)
	Others	10 (9.4)
INR		1.03±0.07
Platelets		371036.54±90486.26
Respiratory function tests	Normal	94 (89.5%)
	Abnormal	11 (10.4%)

BMI: body mass index, ASD: autism spectrum disorders, INR: international normalized ratio

Table 2. Perioperative variables

Number of previous operations		7.40±7.33
Duration of the last operation (minutes)		139.66±74.49
Type of anaesthesia	General	104 (99.0%)
	Spinal	1 (1.0%)
Presence of fracture	No	103 (98.1%)
	Yes	2 (1.9%)
Presence of bleeding	No	69 (65.7%)
	Yes	36 (34.3%)
Amount of bleeding (mL)		313.06±203.24
Admittance to the intensive care unit	No	98 (93.3%)
	Yes	7 (6.7%)
Length of stay in the intensive care unit (days)		2.14±0.69
Length of hospital stay (days)		10.26±9.94
Cobb angle for patients with scoliosis		35.40±24.93
Procedures classified by anatomical region	Femur	66 (62.9%)
	Tibia	19 (18.1%)
	Knee	10 (9.5%)
	Forearm	5 (4.7%)
	Others	5 (4.7%)

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DISCUSSION

In this study, we found that low BMI, presence of scoliosis, and recurrent operations in severe OI types affect morbidity and pose a high risk. Particularly scoliosis was an independent risk factor for morbidity. Therefore, we suggest that a detailed perioperative preparation can reduce these risks. In the present study, we reported that 45.7% of the patients had additional diseases. There were no complications except perioperative fractures in two cases. We found that 35.2% of the cases had postoperative respiratory, neurological, psychiatric, and musculoskeletal complications and growth retardation. Similar to our findings, Tripković et al. (12) in their retrospective study between 1980-2012 evaluating the preoperative features, comorbidities, types of anesthesia and complications associated with anesthesia in 26 patients diagnosed with OI and who had undergone a total of 103 operations at an orthopaedic clinic reported that 89 patients received general anesthesia and 14 patients received regional anesthesia; 14 had intraoperative complications (mostly difficult intubation) and six had postoperative cardiovascular instability. However, different from our results Bojanić et al. (13) reported no perioperative anesthetic complications in their cohort, including 180 operations in 49 patients with OI. We think that the difference between the studies originates from the methods of two studies. They included only perioperative anesthesia-related complications, but we included all perioperative and postoperative complications, including both anesthesia and orthopaedic related. Engel Espinosa et al. (14), in another retrospective study, screened the perioperative complications of 29 patients with OI and who were operated on 105 times between 1991 and 2009. They detected a latex allergy as a co-morbidity for 2 patients. Among the interventions, they had complications occurring in 38% cases, whereas perioperative complications included non-malignant hyperthermia and a femoral fracture. This result is very similar to our complication rate. We demonstrated that the presence of scoliosis influenced morbidity. Pulmonary complications are important causes of morbidity and mortality in patients with OI. Pulmonary problems are multifactorial in these patients, but few studies are present that systematically assess the pulmonary function in individuals with OI (15). In our study, when we examined the patients with abnormal preoperative PFT results, it was found that this surprisingly had no effect on morbidity. We believe that this is due to the small number of patients included with abnormal PFT in our study. Wekre et al. (16) In their prospective study, evaluated the association of spinal deformities and pulmonary dysfunction in an adult patient group with OI and suggested that spinal deformities affect the height and lung function of these patients. Additionally, the spirometry measurements of patients with OI were analysed in a multicentre observational study by Tam et al. (15). Forced vital capacity and forced vital capacity in 1 second values were found to be extremely low in patients with type III OI compared with the normal population; however, the spirometry analysis revealed that pulmonary involvement was low. There are several case reports reporting similar cases managed under general anesthesia. Karabiyik et al. (17) Reported that they used laryngeal mask airway safely together with total intravenous anaesthesia. Therefore, studies on regional anesthesia have come to the forefront (18). These patients should be kept under close clinical observation both during the hospitalization and after discharge in terms of increased risk for postoperative long-term complications that were established in the present study.

Because of the bleeding tendency of patients with OI, studies investigating this relationship have been conducted. For example, Persiani et al. (8) retrospectively evaluated 23 patients with type III OI between 6-13 years of age and who were treated

Table 3. Distribution of factors associated with morbidity and mortality

		n (%)
Postoperative morbidity	No	68 (64.8)
	Yes	37 (35.2)
	Musculoskeletal complication	26 (70.2)
	Neurological complication	6 (13.6)
	Perioperative fracture	1 (2.7)
	Postoperative respiratory distress	1 (2.7)
	Others	4 (10.6)
Mortality	No	103 (98.1)
	Yes	2 (1.9)

Table 4. Morbidity assessment according to descriptive characteristics and preoperative clinical features

		Morbidity (-) (n=66)	Morbidity (+) (n=39)	p-value
Age (years)		10.07±5.78	12.77±10.02	[a]0.289
BMI (kg/m^2)		21.16±3.94	19.94±6.31	[a]0.008**
Scoliosis	No	63 (69.2)	28 (30.8)	[b]0.001**
	Yes	3 (21.4)	11 (78.6)	
INR		1.04±0.07	1.02±0.06	[a]0.123
Platelets		369453.03±92916.19	373786.84±87257.71	[c]0.815
Respiratory function tests	Normal	59 (62.8)	35 (37.2)	[d]0.519
	Abnormal	7 (63.6)	4 (36.3)	

BMI: body mass index, INR: international normalized ratio, [a]Mann-Whitney U test, [b]Pearson chi-square test, [c]Student t-test, [d]Fisher-Freeman-Halton test, **p<0.01

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Table 5. Evaluation of morbidity by operation characteristics

		Morbidity (-) (n=66)	Morbidity (+) (n=39)	p-value
Number of previous operations		5.49±6.26	8.53±7.72	[a]0.014*
Area of operation	Femur	46 (69.7)	20 (30.3)	[d]0.134
	Tibia	12 (63.2)	7 (36.8)	
	Knee	4 (40.0)	6 (60.0)	
	Arm	3 (60.0)	2 (40.0)	
	Others	1 (20.0)	4 (80.0)	
Duration of the last operation		135.45±61.79	146.47±92.04	[c]0.540
Type of anaesthesia	General	66 (63.5)	38 (36.5)	[e]0.371
	Spinal	0 (0)	1 (100)	
Presence of fracture	No	0 (0)	1 (100)	[e]0.136
	Yes	0 (0)	2 (100)	
Presence of bleeding	No	41 (59.4)	28 (40.6)	[b]0.313
	Yes	25 (69.4)	11 (30.6)	
Amount of bleeding (mL)		334.80±233.33	263.64±100.23	[a]0.536
Admittance to the intensive care unit	No	60 (61.2)	38 (38.8)	[e]0.254
	Yes	6 (85.7)	1 (14.3)	
Length of stay in the intensive care unit (days)		2.33±0.52	1.00±0	[e]0.130
Length of hospital stay (days)		10.38±9.33	10.05±11.07	[a]0.274

[a]Mann-Whitney U test, [b]Pearson chi-square test, [c]Student t-test, [d]Fisher-Freeman-Halton test, [e]Fisher's Exact test, *p<0.05

Table 6. Logistic regression analysis of risk factors affecting the status of morbidity

	p-value	OR	95% CI	
			Lower	Upper
BMI (kg/m^2)	0.405	1.039	0.950	1.136
Presence of scoliosis	0.002**	9.082	2.226	37.062
Number of previous operations	0.094	1.063	0.990	1.140

CI: confidence interval, OR: odds ratio, BMI: body mass index, **p<0.01

for femoral fractures, and the relationship between age, BMI, duration of operation and blood loss was analysed. In that study, no significant association was found between the duration of surgery and blood loss. The age of the patient was found to be inversely proportional to the amount of blood loss, and children older than 10 years were found to have a statistically lesser amount of average blood loss (8). It has been suggested that the risk of intraoperative and postoperative bleeding is lower in children with higher body weights. As a result, blood loss per kilogram was higher in patients with a low BMI. In our study, we found that the rate of morbidity was significantly higher in patients with a low BMI. However, we could not find any relationship between blood loss and morbidity. We think that this increased morbidity is due to malnutrition and poor care of patients. Pichard et al. (19) Reported that there was no relationship between the duration of operation and mean blood loss and that the blood loss decreased with increasing age.

There are no studies that investigated the relationship between ICU stay and morbidity in patients with OI. Transfer to the postoperative ICU may be necessary when patients have ongoing conditions such as severe intraoperative complications, disorders of haemorrhagic diathesis, or restrictive respiratory symptoms. In our study, 6.7% of the patients were admitted to the ICU. Two cases resulted in mortality (1.9%) in the present study. There are few studies have investigating perioperative morbidity and mortality in orthopaedic operations of patients with OI. Among the studies in which other surgical procedures were examined, Lamanna et al. (20) In their cardiac case series in patients diagnosed to have OI, reported that valvular surgery in these patients might be complicated by bleeding, arrhythmia, cardiac rupture, valve detachment, and delayed wound healing, although they are technically feasible. Additionally, the morbidity and mortality of those cases were found to be higher. In this study, perioperative mortality and long-term mortality rates were reported as 18% and 8%, respectively (20). On the other hand, McAllion and Paterson (21) in a retrospective report, investigated the causes of death of 68 patients with OI and who died between 1980-1995. They especially emphasized the importance of respiratory complications in severe disease form (type III). It was reported that chest deformity and scoliosis contribute to the limitation of pulmonary functions in these patients. It has been reported that most of the deaths are the results of intracranial hemorrhage and bacillary invagination or restrictive heart failure due to kyphoscoliosis. It is also emphasized that minor traumatic events may be complicated causing death (21). In this present study, it was seen that scoliotic deformity among the risk factors

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for morbidity had a highly significant association with morbidity. According to the model, the presence of scoliosis affected morbidity by increasing the risk by 9.082-fold. The presence of scoliosis is an independent risk factor. In a survey study examining the observations of 121 patients treated with 51 fusion surgery in 14 countries to document the orthopaedic surgical outcomes of OI-related scoliosis; reported similar results and documented that the increased size of the Cobb angle before spinal fusion and the presence of kyphosis were associated with a high complication rate, and that in the absence of pseudoarthrosis and kyphosis, the spinal curvature appeared in the late period (22). Scoliosis is more common in patients with severe OI and especially in advanced age. Since many systems, especially the cardiopulmonary system, are affected in these patients, morbidity is expected to be higher (23).

Study Limitations

Finally, we should also mention the limitations of our study. Because of the relatively low incidence and prevalence of OI, as with many rare diseases, there is a difficulty in designing a prospective study that can examine target parameters. For this reason, many studies such as the present study were designed retrospectively; OI patients with multiple operations were included in the study as individual cases. On the other hand, surgical indications and techniques used for treating OI are relatively new. Another limitation of our study was the lesser number of cases included in the study, which can contravene the causal relationship of the statistical results. To avoid this limitation, we believe that the data of the cases of OI can be optimized by increasing the number of cases with multicentre studies.

CONCLUSION

In this present study, the presence of scoliosis was found to be an independent risk factor that has an effect that increases the risk of morbidity 9-fold. Considering these data, we believe that medical personnel should be aware of the potential complications associated with OI. We suggest that all patients with OI undergoing orthopaedic surgery should be preoperatively screened for the presence of scoliosis. We recommend more randomized controlled studies to approve our results should be warranted.

Ethics Committee Approval: This retrospective study was performed following the approval of the Ethics Committee of İstanbul University, İstanbul Faculty of Medicine (decision no: 12, date: 29.06.2018).

Informed Consent: Retrospective study.

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