THE IMPACT OF THREE SIBLINGS OF CHILDREN WITH SEVERE OSTEOGENESIS IMPERFECTA ON FINANCIAL BURDEN FOR INPATIENT CARE IN A HOSPITAL

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ABSTRACT

Osteogenesis imperfecta (OI) is a rare genetic skeletal disorder that has a major impact to the financial status of the suffered’s family. We investigated the cost expenditures of three siblings with OI after five admissions in a hospital. Three siblings, two brothers and one young sister aged 1-6 years old were admitted to Methodist Susanna Wesley Hospital in Medan. We reviewed the computerized database capturing a variety of cost expenditures of diagnosis determinant, medical treatment, and hospital charges for each admission. Total charges for five admissions in the hospital were > Rp.31 millions, of which 21.25% was accounted for by diagnosis determinant, 9.38% for treatment, and 69.37% for hospital stay. The mean length of stay was 10 days for five admissions. The findings show that there is enormous financial impact of osteogenesis imperfecta on inpatient care.

Keywords: Financial impact, Family, Osteogenesis imperfecta

INTRODUCTION

Osteogenesis imperfecta (OI), often referred to as "brittle-bone disease," is a heritable genetic disorder characterized in most affected persons by either a reduction in the production of normal type I collagen or the synthesis of abnormal collagen as a result of mutations in the type I collagen genes (Glorieux et al. 1998). Its hallmark feature is bone fragility, (Lang, 2003; Rauch and Glorieux, 2004) with a tendency to fracture from minimal trauma (Marini, 1998) or from the work of bearing weight against gravity.

Improved physician awareness may lead to earlier diagnosis in patients with
previously undiagnosed genetic conditions and improved management of patients with known genetic disorders (Rauch and Glorieux, 2004). Caring of patients with these fractures is also very expensive, and costs will continue to rise as the frequency of fractures increases in the future. To our knowledge, no studies have detailed the cost expenditures of OI patients admitted to the hospital.

Methodist Susanna Wesley Hospital, located in Medan, serves a large diverse disease, either the primary or specialty care. For many families served in the hospital, predominantly the poor family.

SUBJECTS AND METHODS

Between October 2006 and May 2007, three siblings of children (aged 1-6 years) with previously suspected of osteoporosis were admitted to Methodist Susanna Wesley Hospital in Medan for determining the causes of their bone fractures. The clinical, biochemistry, and radiograph examinations were done to confirm the diagnosis.

The computerized database capturing a variety cost expenditures of diagnosis determinant, medical treatment, and hospital charges for each admission was maintained by the financial department at the Methodist Susanna Wesley Hospital. These records formed the total expenditures during the five admissions at a different timeframe at this hospital.

RESULTS

Table 1 shows the gender, ages, height, and biochemistry measurements and the drug (zoledronic acid) used for treating the patients. The initial dose was 0.015 mg/kg, followed by 0.025 mg/kg 12 weeks after the first dose, and the third dose was increased to 0.05mg/kg thereafter every 3 months. The alkaline phosphatase levels were increased, indicating increased bone formation. The urinary type I collagen N-telopeptidase/creatinine was also increased compared to the normal range, suggesting increased bone resorption (Rauch et al. 2003). Taken together, the biochemistry changes confirmed the diagnosis of OI.

Table 2 and Figure 1 show the radiological findings of osteopenia and fractures, the hallmark of severe osteogenesis imperfecta, (Glorieux et al. 1998), probably results from structural abnormalities in bone tissues (Sykes, 1990) and a reduced rate of osteogenesis (Glorieux, 1994).

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**Table 1. The biochemistry characteristics of patients with osteogenesis imperfecta**

<table>
<thead>
<tr>
<th>Patients No</th>
<th>Age (y,m)</th>
<th>Height (cm)</th>
<th>Serum Alkaline Phosphatase (Unit/liter)</th>
<th>Urinary Type I Collagen N-Telopeptidase/creatinine (nMBCE/mM)</th>
<th>Zoledronic Acid Treatment (mg/kg)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Boy</td>
<td>1</td>
<td>5.7</td>
<td>80</td>
<td>552 (115-391)</td>
<td>2890 (13-78)</td>
</tr>
<tr>
<td>Boy</td>
<td>2</td>
<td>3.9</td>
<td>70</td>
<td>514 (115-391)</td>
<td>4221 (13-78)</td>
</tr>
<tr>
<td>Girl</td>
<td>3</td>
<td>1.10</td>
<td>70</td>
<td>417 (115-460)</td>
<td>2387 (14-74)</td>
</tr>
</tbody>
</table>

* Ref. R indicates reference range as established in the accredited laboratory.
# Ref. R indicates reference range as established in the specialty laboratory (Valencia, CA, USA).

**Table 2. The radiological findings of patients with osteogenesis imperfecta**

<table>
<thead>
<tr>
<th>Patient</th>
<th>Spine Abnormalities</th>
<th>Osteopenia</th>
<th>Deformity</th>
</tr>
</thead>
<tbody>
<tr>
<td>Boy</td>
<td>Biconcave vertebra</td>
<td>Yes</td>
<td>Bowing extremities, Bulbous distal</td>
</tr>
<tr>
<td>Boy</td>
<td>Biconcave vertebra</td>
<td>Yes</td>
<td>Bowing extremities, fractures</td>
</tr>
</tbody>
</table>
Table 3. Costs of expenditure of patients with OI in hospital stay

<table>
<thead>
<tr>
<th>Types of service</th>
<th>1st Adm (6 d)</th>
<th>2nd Adm (13 d)</th>
<th>3rd Adm (10 d)</th>
<th>4th Adm (9 d)</th>
<th>5th Adm (12 d)</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Inpatient Hospital</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>19,636,800</td>
</tr>
<tr>
<td>Diagnostic Radiology</td>
<td>2,365,000</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>2,365,000</td>
</tr>
<tr>
<td>Biochemical Measurements</td>
<td>3,600,000</td>
<td>1,960,000</td>
<td>360,000</td>
<td></td>
<td></td>
<td>5,920,000</td>
</tr>
<tr>
<td>Physician Visits</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Medications</td>
<td>126,200</td>
<td>132,300</td>
<td>921,200</td>
<td>1,081,700</td>
<td>1,163,400</td>
<td>3,424,800</td>
</tr>
</tbody>
</table>

Abbreviations: Adm = admission; d = day; Rp = rupiah.

Table 3 shows the total charges of the five admissions in the hospital were Rp. 31,346,600, in which Rp. 8,285,000 (26.43%) was accounted for by diagnosis determinant (diagnostic radiology and biochemical measurements), Rp. 3,424,800 (10.92%) for medications, and Rp. 19,636,800 (62.65%) for hospital stay. The mean length of hospital stay in five admissions was 10 days.

DISCUSSION

This study begins to quantify the enormous impact of the osteogenesis imperfecta on financial burden in inpatient care. We found that, in five admissions, the total cost expenditures in the hospital were substantially high for the poor family of six children with the last three affected this genetic bone disease. The total cost would be higher than that calculated if the hospital did not provide the social beds for the poor. This study also confirms what Hall et al, found >25 years ago—that the cost to individual and society for treating genetic disorders is high (Hall, et al. 1978).

The first admission was to improve the general conditions and to confirm diagnosis of the patients. The second admission were intended to improve the
whole health status and giving supplemental calcium prior to given intravenous antiresorptive drugs. The third to fifth admissions were given zoledronic acid, the antiresorptive agent, for strengthening the bone by reducing bone pain and fracture. Not surprisingly, the longer hospital stay was similar to that reported for children who are admitted for genetic disorders. (Hall, et al. 1978; McCandless et al. 2004). To our knowledge, this is the first report regarding financial burden for three siblings with severe osteogenesis imperfecta. Further studies should extend on a larger sample on diverse genetic bone disorders concerning socioeconomic burden in families.

There are several drawbacks to the current study. First, only three siblings of children in a single family were analyzed. Second, the financial burden was imposed by a generous hospital; and third, the billings were paid by a foundation. In spite of these drawbacks, the findings are consistent with those of Hall et al. in an entirely different hospital setting and provide a unique case in a single family.

Osteogenesis imperfecta is a rare genetic skeletal disease, so that the healthcare provider could miss the diagnosis. The fourth child was undiagnosed in inpatient care in a hospital in Medan when his parents had noted the spontaneous femoral fracture while he was lifted up. Subsequently, the fifth and sixth children were born with the same symptoms related to osteogenesis imperfecta. The previous undiagnosed genetic conditions is also encountered in the primary even the tertiary provided health care hospital (Kumar et al. 2001).

CONCLUSION

The financial burden of the osteogenesis imperfecta in inpatient care is high and has a longer hospital stay. The data set were based and analyzed by capturing a variety cost expenditures of diagnosis determinant, medical treatment, and hospital charges for each admission at the financial department of the Methodist Susanna Wesley Hospital in Medan.

SUGGESTION

Improved physician awareness may lead to earlier diagnosis of osteogenesis imperfecta, which leads to improved management of patients with known genetic disorders. Every person providing care to children will need to be competent to explain these complexities to anxious public and to discuss their implications with affected families due to enormous impact on their families in terms of financial burden and their subsequent risks of giving birth of genetic disorders.

ACKNOWLEDGEMENT

The authors would like to thank the Yayasan Lestari Indo Makmur (Marga LIM) in Medan for funding the hospitalization of the three siblings of children at the Methodist Susanna Wesley Hospital, Medan.

REFERENCES

