

Sheehan's syndrome: A descriptive case series from a developing country

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Abstract

BACKGROUND AND OBJECTIVES OF THE STUDY

Sheehan's syndrome refers to pituitary ischaemic necrosis due to massive postpartum haemorrhage. It is rarely reported from sub-Saharan Africa despite high incidence of postpartum haemorrhage. The objective of the study was to describe the clinical characteristics of the cases seen by the Endocrinology unit in a tertiary hospital over a period of two years.

SUBJECTS, MATERIALS AND METHODS

Five cases of Sheehan's syndrome were seen between March, 2016 and March, 2018. Clinical data was retrieved from the case records and analyzed using SPSS-version 20.

RESULTS

The age at diagnosis was 36 ± 8.67 years. The median interval between the obstetric haemorrhage and diagnosis was 11 years. Amenorrhoea/oligomenorrhoea, lactation failure, weight loss, excessive tiredness and cold intolerance were found in all the cases. 80% had loss of libido while 60% had loss of hair. 40% had fainting spells. 60% had hypotension. All the patients had anaemia and secondary hypothyroidism. 80% had hypocortisolemia. Hyponatremia was found in 20%. Empty sella was found in 2 out of the 3 (67%) cases that did cranial magnetic resonance imaging.

CONCLUSION

Sheehan's syndrome is still rarely seen and diagnosed late as the symptoms are mostly non-specific despite the fact that postpartum haemorrhage is quite common. Menstrual irregularities, lactation failure weight loss and hypothyroidism are common in Nigerian patients.

Keywords: Sheehan's syndrome, case series, descriptive, developing country

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Introduction

Sheehan's syndrome refers to pituitary ischaemic necrosis due to massive postpartum haemorrhage [1]. It may however be rarely seen without massive bleeding or after normal delivery [2]. Patients with Sheehan's syndrome have varying degrees of anterior pituitary hormone deficiency [3]. It is a rare cause of hypopituitarism in developed countries due to advances in obstetric care although it is still relatively frequent in developing countries [4]. The higher prevalence in developing countries is ascribed to poor obstetric practices, home deliveries and unavailability of rapid blood transfusion in several health facilities [2].

There are scanty data on the prevalence and clinical characteristics of patients with Sheehan's syndrome in sub-Saharan Africa. Over a five year period, 11 cases of Sheehan's syndrome were seen in the Endocrinology Unit of a teaching hospital in Ibadan, Nigeria [5]. The prevalence of Sheehan's syndrome in India is estimated to be 2.7–3.9% among parous women older than 20 years [6]. In an international database (KIMS database) containing 1034 patients with GH deficiency (GHD), Sheehan's syndrome is the cause in 3.1% of the cases [7]. Also, in a Spanish cross-sectional study, Sheehan's syndrome is responsible for 6-10 cases of growth hormone deficiency cases [8].

Enlargement of pituitary gland, small sella size, disseminated intravascular coagulation and autoimmunity have been suggested to play a role in the pathogenesis of Sheehan's syndrome in women who suffer from severe postpartum hemorrhage [5]. In addition, Cakir *et al.* found protein S deficiency in 2 out of 12 patients with Sheehan's syndrome [9].

The patients may seek medical advice because of various presentations ranging from non-specific symptoms to coma and the clinical manifestations vary from one patient to another. Although a small percentage of patients with Sheehan's syndrome may cause abrupt onset severe hypopituitarism immediately after delivery, most patients have a mild disease and go undiagnosed and untreated for a long time [5]. These myriads of symptoms may make the women present to different specialties. So, a high index of suspicion would be required to make a diagnosis [10]. The most important clues for diagnosis of Sheehan's syndrome are lack of lactation and failure of menstrual resumption after a delivery complicated with severe hemorrhage [11].

Methodology

This is a retrospective study of the prevalence and clinical characteristics of Sheehan's syndrome in patients attending the endocrinology clinic of the University College hospital, Ibadan, Nigeria. The hospital is an over 800-bedded academic tertiary health institution and a key referral centre from the southwest zone of Nigeria. Demographic and clinical data was retrieved from the case records of patients seen between March, 2016 and March, 2018.

Ethical approval for this study was obtained from the Joint University of Ibadan/University College Hospital Institutional Review Board (IRB).

Data on obstetric history, mode of presentation, laboratory investigations and radiographic test results were collected. Results of hormonal assays were also obtained from the patients' records. All diagnostic testing was done in a standardized manner at that time. Continuous variables summarized as mean or median as appropriate and categorical variables summarized as frequency (percentage).

Results

Five cases of Sheehan's syndrome were seen between March, 2016 and March, 2018. The age at diagnosis was 36 ± 8.67 years. The median interval between the obstetric haemorrhage and diagnosis was 11 years. None of the patients delivered at the University College Hospital, Ibadan, Nigeria or any other tertiary hospital in Nigeria. Three of the patients received blood transfusion. Two patients had emergency Caesarean section. All the patients had menstrual disturbance (60% had amenorrhoea whereas 40% had oligomenorrhoea). In addition, all the patients

had lactation failure, weight loss, excessive tiredness and cold intolerance. 80% had loss of libido and 60% had loss of hair and hypotension. Visual disturbance and fainting spells were found in 40%. Headache and facial coarseness were found only in 20% of the patients. Anaemia was found in all the patients while hyponatremia was found in 20%. Only 3 of the patients were able to afford pituitary magnetic resonance imaging and complete empty sella was seen in 2 (67%) of them.

Below shows the summary of the various pituitary axes affected from the hormonal assays done.

All the patients are on replacement prednisolone and levothyroxine therapy. 40% had successful pregnancies after the index postpartum haemorrhage.

Discussion

Five patients were seen at the Endocrinology clinic of the University College Hospital, a tertiary hospital in Nigeria over a period of 2 years. This gives a total number of 2 to 3 cases per year. Famuyiwa *et al* reported 11 cases of Sheehan's syndrome over a period of 5 years in the Endocrinology Unit of a teaching hospital in Ibadan, Nigeria [5]. This gives a total of 2 cases per year which is similar to what was found in this study. Ramindranoa *et al* reported one case each year in a population study done in France over 31 years [12]. This is unsurprising as the incidence of Sheehan's syndrome is said to be less prevalent in developed countries.

In this study, the mean age at diagnosis was 36 years. This finding is similar to the findings of other researchers such as Famuyiwa *et al* who reported an average age of diagnosis of 35 years [5]. Kristjansdottir *et al* reported an average age of 37 years [1] while Mokta *et al* reported 38 years as the mean age at diagnosis [13]. According to the findings in this study, the median interval between the obstetric haemorrhage and diagnosis was 11 years. Sanyal *et al* also reported a lag period of 15 years between the postpartum haemorrhage and diagnosis of Sheehan's syndrome [14]. In a study of 60 patients, the average time between the previous obstetric event and diagnosis of Sheehan's syndrome was 13 years [15].

60% of the patients in this study had classic amenorrhoea. This is similar to the findings of Sayyal *et al* who reported 60% classic amenorrhoea in their study [10]. Shivasprasad had stated that the absence of the classic amenorrhoea does not rule out Sheehan's syndrome [3]. All the patients had lactation failure. In a case series done by Dash *et al*, he also found the incidence of lactation failure in Sheehan's syndrome to be 100% [16].

Anaemia was found in all the patients while hyponatremia was found in 20% in this study. In their study, Laway *et al* also reported that the frequency of anemia was significantly higher in

Table 1: summary of the various pituitary axes affected from the hormonal assays done.

Pituitary axis	Number of patients who had hormonal assays	Percentage affected of the patients assayed
Secondary hypogonadism	3	100%
Secondary hypothyroidism	5	100%
Low a.m. Cortisol	5	80%
Growth hormone deficiency	None	-

the patients with Sheehan's syndrome compared to controls [17]. Gokalp *et al* also reported that Sheehan's syndrome patients had significantly higher rates of anaemia compared to controls [18].

Magnetic resonance imaging done for the three patients that could afford the imaging showed complete empty sella. In a case series, Laway *et al* reported that the hypophysial magnetic resonance imaging (MRI) confirmed empty sella in all the cases [17]. However, Fleckman *et al* reported that only 55% of the cases in their study had complete empty sella [19]. This could be due to the fact that Fleckman *et al* used computerized tomography imaging which is less sensitive compared to magnetic resonance imaging.

It is worthy of note that 2 (40%) patients had successful pregnancies following the index postpartum haemorrhage leading to the diagnosis of Sheehan's syndrome. Chaieb *et al* also reported 2 cases of spontaneous pregnancies in patients with Sheehan's syndrome [20].

Limitation

The case notes of the patient did not contain details about the obstetric events leading to the diagnosis of Sheehan's syndrome. Also, the patients could not afford all the necessary hormonal assays or dynamic testing.

Conclusion

Sheehan's syndrome is still rarely seen and diagnosed late as the symptoms are mostly non-specific despite the fact that postpartum haemorrhage is quite common. Menstrual irregularities, lactation failure weight loss and hypothyroidism are common in Nigerian patients.

Conflict of interest

none

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