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FACTITIOUS HYPOGLYCAEMIA: A CASE REPORT AND LITERATURE REVIEW

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SUMMARY

Background: *Factitious hypoglycaemia is a form of factitious disorder imposed on self with high morbidity and mortality. It is therefore important to be aware of the key demographic and contextual risk factors for factitious hypoglycaemia, as well the investigations and management options available for suspected cases.*

Subjects and methods: *In this article we describe a case report and literature review of factitious hypoglycaemia. The search was conducted using the PubMed database and identified 23 case reports of 31 patients aged 18 or over with insulin-induced factitious hypoglycaemia.*

Results: *The average age of these patients was 33.7 (±13.5) years, the female: male ratio was 4.3:1, 38% had medical occupations or past medical training, 53% had diabetes mellitus, and 41% had a positive psychiatric history. Misdiagnoses were common and often resulted in inappropriate treatment. Very few cases discussed psychiatric management.*

Conclusions: *Factitious hypoglycaemia is more commonly reported in middle-aged females, in a medical profession, with a past medical history of diabetes mellitus and psychiatric illness. However, it may affect a variety of patients and the absence of these features should not discourage a diagnosis. C-peptide levels and insulin assays can help identify factitious hypoglycaemia over other causes of hypoglycaemia, and management should include a greater focus on psychiatric treatment.*

Key words: *factitious disorder – hypoglycaemia - Munchausens syndrome*

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CASE REPORT

A quadragenarian with type 1 diabetes mellitus was admitted to the general hospital with a reduced appetite and frequent hypoglycaemic episodes. Investigations revealed hypoglycaemia (glucose 1.0 mmol/L) with a normal serum concentration of insulin (4 pmol/L) and low C-Peptide concentration (141 pmol/L). HbA1c was within normal limits (40 mmol/mol). An ultrasound of the pancreas showed signs of known chronic pancreatitis.

The patient was known to local mental health services due to his complex psychiatric history, including emotionally unstable personality disorder, episodes of self-harm, and past suicide attempts by poisoning and hanging. He had presented to the emergency department on a number of occasions with thoughts of ending his life. The patient is unmarried and has no children but identifies his siblings and mother as protective factors. He has also been diagnosed with post-traumatic stress disorder which he attributes to the sexual abuse he experienced in childhood, and a road traffic accident he was involved in around the same time.

In addition, the patient has a history of substance abuse, including opioids and alcohol. Whilst he is on replacement therapy for the former, he is still drinking 35-40 units of alcohol daily. This is in spite of the efforts of the local alcohol and drug service. There is a family history of alcoholism and his father passed away from alcohol-related complications. The patient has never wanted to speak about his past in depth, so we do not have detailed information relating to his upbringing, illness behaviors and relationships. We are, however, aware that he has a positive forensic history.

During his stay on the ward there were concerns that the patient was leaving the premises and returning intoxicated, which he consistently denied. After these breaks, he often felt unwell and was found to have a low blood glucose concentration. During one particular episode, the patient collapsed, and three insulin pens fell out of a pouch that he carried. This surprised the medical team who had removed all of his medication upon admission. The patient claimed that his mother had brought these pens into hospital and was adamant that he had never used them during his stay on the ward. He also denied feeling suicidal. The insulin pens were confiscated and following a period of observation, the MDT determined that by leaving the ward, the patient was putting himself at risk. He was placed on a Section 5(2) of the Mental Health Act (MHA) to prevent him leaving the premises, then was detained under Section 2 and admitted to an inpatient psychiatric unit.

During this time, no further hypoglycaemic episodes occured and he was eventually discharged back to the community healthcare services. He was therefore diagnosed retrospectively with factitious hypoglycaemia secondary to insulin misuse.

BACKGROUND

Nomenclature

'Munchausen's syndrome' was coined in 1951 as a way of describing patients that exaggerate or produce their own symptoms (Asher 1951). It compares sufferers to the literary character Baron Munchausen, renowned for his outlandish lies and use of exaggeration (Munchausen 1992). However, given the origin of the word 'Munchausen's', medical professionals have long argued that it is a disrespectful term (Fisher 2006). It does not appreciate the seriousness of these disorders, which can result in death (Schober et al. 2008, Unal et al. 2017). Medical authorities including the American Psychiatric Association and World Health Organisation are therefore moving away from this term, instead opting to use 'factitious disorder' in their latest criteria (the DSM-5 & ICD-10 respectively) (DSM-5 2013, ICD-10 2016). Interestingly, there has also been some critique over the use of the word 'factitious' but this has gained comparatively little traction (Bauman et al. 2018). The result is that in modern practice, Munchausen's syndrome may be instead referred to as factitious disorder imposed on self (FD), whilst Munchausen's by proxy is being phased out in favour of factitious disorder imposed on another. Contrastingly, the media continues to publicise anglicised versions of these old eponyms through platforms such as television (Gajanan 2019). It is therefore important that clinicians are aware that they may encounter both terms in their day-to-day practice.

Diagnostic criteria

This article focused on FD, in which patients exaggerate or create symptoms of illness in themselves to seek attention, examination, treatment and/or sympathy from medical personnel. This is a compulsive behaviour that patients find hard to ignore even when the risks of their actions are known or complications arise (DSM-5 2013). It has been suggested to have an unconscious component that cannot be fully controlled; it is only voluntary in the sense that is deliberate and purposeful (DSM-5 2013). It is important to differentiate FD in which there is personal gain due to mental illness (e.g. to assume the sick role), from malingering, in which the patients action result in external gain (e.g. financial compensation). Similarly, it is distinct from hypochondriasis in which symptoms are not self-induced but there is instead an exaggerated self-awareness or hypervigilance towards illness. The latest diagnostic criteria for FD are presented below (DSM-5 2013):

- Falsification of psychological or physical signs or symptoms, or induction of disease or injury associated with identified deception;
- The individual presents to others as injured, ill or impaired;
- The deceptive behaviour is apparent even in the absence of external incentives;
- The behaviour is not better explained by another mental disorder.

Notably, there are many variants of FD. For example, patients may produce signs by misusing bodily fluids such as blood and faeces, or by abusing medications (Naqvi et al. 2017, Iwanaga et al. 2019). There are many cases of the latter described in the literature, including the surreptitious use of thyroxine, steroids and even adrenaline (reviewed in Kinns et al. 2013). However, the classic example is that of insulin misuse which remains the focus of this paper. This has been appropriately dubbed 'factitious hypoglycaemia' (hereby referred to as FH). Importantly, we realise that with the advent and increasing use of novel hypoglycaemic drugs such as sulfonylureas, other means of FH are now becoming popular. Considering the above, it is therefore essential to keep an open mind when dealing with patients suffering from any suspected factitious disorder.

Importance & Aims

FH is dangerous for a multitude of reasons. Firstly, the means by which patients produce symptoms often result in severe complications, including coma, brain damage and death (Brett et al. 2016, Grunberger et al. 1988). Moreover, the investigations and treatments they are subjected to also come with significant morbidity and mortality. For example, there have been numerous cases of unnecessary pancreatectomies and laparotomies in patients later found to have insulin-induced FH (Gil et al. 2011, Berkowitz et al. 1971). However, these risks do not deter patients and they are 'willing to undergo incredible hardship to perpetuate the masquerade' (DSM-5 2013). It follows that FH often results in repeated admissions, prolonged costly hospital stays, and further stress on resource-limited healthcare systems. The literature review has been inspired by the aforementioned case, and has three primary aims:

- To highlight the key demographics, features and risk factors these patients present with so that we can identify them more readily in future;
- To review and clarify what investigations are available for suspected cases of FH;
- To explore what management options exist and the outcomes/long term prognosis in these patients.

SUBJECTS AND METHODS

We searched the PubMed database using different combinations of the following keywords: factitious, Munchausen, hypoglycaemia and insulin. This identified 123 papers of which 50 were fully accessible in English and are referenced throughout this study. The data was then further processed to produce Results and Table 1. Only case reports of insulin-induced factitious hypoglycaemia over the age of 18 were included. Other diagnoses and paediatric patients were excluded. The following information was extracted from qualifying reports: age, sex, occupation, diagnosis of diabetes, past medical history, psychiatric history, background and risk factors.

Table 1. Data from Literature Review

Paper	Age	Sex	Occupation	Diagnosis of diabetes	Past medical history	Psychiatric history	Family history	Background and other risk factors
Whelton MJ et al. 1968	23	F	Biochemistry technician	T1DM	No Info	Personality Disorder	No info	No info
Berkowitz S et al. 1971	44	M	Postal worker	IDDM	No Info	No Info	No info	No Info
Scarlett JA et al. 1977	20	F	Nurse	No	No info	No info	No info	No info
Scarlett JA et al. 1977	41	F	Nurse's aide	No	No info	No info	No info	No info
Scarlett JA et al. 1977	20	F	Medical technician	No	No info	Substance abuse	No info	No info
Scarlett JA et al. 1977	24	F	Nurse	No	No info	No info	No info	No info
Scarlett JA et al. 1977	27	F	Unemployed	T1DM	No info	No info	No info	No info
Dimitriadis G et al. 1980	32	F	Nurse	IDDM	Obesity	Hysterical personality	No info	No Info
Schade DS et al. 1985	21	F	No info	T1DM	Obesity	No info	No info	No info
Schade DS et al. 1985	30	F	Nurse	T1DM	No info	No info	No info	No info
Schade DS et al. 1985	28	F	Nurse	T1DM	No info	No info	No info	Refused to live with roommate or parents
Roberts I et al. 1985	16	F	No info	No	No Info	No Info	Father & sister have IDDM	No info
Walfish PG et al. 1987	28	F	Hospital ward clerk	No	Polycystic ovarian syndrome, headaches	No Info	Aunt has diabetes	Friend had been injecting her with illicit drugs
Johnson RG Jr et al. 1987	24	F	Nurse	No	No Info	No significant psychiatric Hx	No info	Boyfriend and husband of best-friend are diabetics
Schuler G et al. 1989	46	F	No info	IDDM	No significant medical Hx	Abnormal personality	No info	No info
Schuler G et al. 1989	72	M	No info	IDDM	Minor myocardial infarction	Abnormal personality	No info	No info
Lebowitz MR, Blumenthal SA. 1993	22	F	Emergency medical technician	No info	No info	Suicide attempts (two), substance misuse	No info	Abused by father
Lebowitz MR, Blumenthal SA. 1993	66	F	No info	No info	Hypothyroidism	Depression	No info	No info
Roy M, Roy A. 1995	32	F	Unemployed	No info	No info	Depression, suicidal ideation (past overdoses)	No info	Family issues, marital issues

Total number of cases: 32;. No info = no information (unspecified); T1DM = type 1 diabetes mellitus; T2DM = type 2 diabetes mellitus; IDDM = insulin-dependent diabetes mellitus; Hx = history

Table 1. Continues

Paper	Age	Sex	Occupation	Diagnosis of diabetes	Past medical history	Psychiatric history	Family history	Background and other risk factors
Jermendy G. 1996	43	F	No info	T2DM	Hypertension	No Info	No info	Desired to remain in hospital despite leaving a child at home
Waickus CM et al. 1999	39	F	No info	T1DM	No Info	Depression, no suicidal ideation	No info	No info
Bretz SW, Richards JR. 2000	25	M	Hospital technician	No	Migraine	Substance abuse	No info	Father was a doctor, mother was a nurse
Ameh V, Speak N. 2008	20	F	No info	No	No info	No info	No info	Psychosocial issues
Donegan D et al. 2012	34	F	No info	T1DM	Pancreas-kidney transplant	Anorexia nervosa	No info	No info
Vega Guedes B et al. 2014	29	F	Unemployed	T2DM	No significant medical Hx	No info	Hypertension	No info
Loh TP et al. 2014	39	M	No info	T1DM	Neuropathy, hypertension, dyslipidaemia, mitral valve prolapse	No info	No info	Several significant social stressors
Joshi T et al. 2016	24	F	No info	T1DM	No info	No info	No info	Significant sexual, emotional & physical abuse, required foster care
Brett F et al. 2016	53	F	No info	No	No significant medical Hx	No info	No info	No info
Chemmanam J et al. 2017	48	F	No info	T2DM	Hypercholesterolaemia, chronic back pain, gastro-oesophageal reflux	Pseudo seizures	No info	No info
Akbari M et al. 2018	46	M	Elevator company	No	No significant medical Hx	No significant psychiatric Hx	Mother has diabetes	Marital disputes, attention-seeking behaviour, past medical training
Patel A, Daniels G. 2018	19	F	No info	No	Acne	No significant psychiatric Hx	No info	Foster care, severe psychological stress, diabetic girlfriend
Case reported here	40s	M	Unemployed	IDDM	Chronic pancreatitis, hepatitis C	PTSD, emotionally unstable personality disorder depression with self-harm & suicidal attempts, substance abuse	No info	Foster care, childhood abuse

Total number of cases: 32;. No info = no information (unspecified); T1DM = type 1 diabetes mellitus; T2DM = type 2 diabetes mellitus; IDDM = insulin-dependent diabetes mellitus; Hx = history

RESULTS

Our search identified 31 patients (or 23 papers) that met the inclusion criteria in Section 4. Methods. Including our own case, these 32 patients were then analysed with regards to the variables above. The average age was 33.7 years + 13.5 standard deviations (range 19-72). There were 26 female cases and 6 male cases (a ratio of 4.3:1). 4 patients were unemployed (12.5%), 12 had medical occupations or past medical training (37.5%), 14 were unspecified (43.8%) and the remainder had other vocational backgrounds (9.4%). 17 had a diagnosis of diabetes (53.1%) of which 9 were explicitly stated to be 'type 1 diabetes mellitus' (28.1%), 5 were 'insulin-dependent diabetes mellitus' (15.6%) and 3 were 'type 2 diabetes mellitus (9.4%). However, there were 3 patients for which no information was available (9.4%) and 12 had no previous history of diabetes (37.5%). 16 (50%) patients had no information regarding past medical history, whilst 4 (12.5%) were stated to have no significant past history and the remaining cases represented a mix of conditions (given in Table 1). 12 patients had a positive psychiatric history (40.6%) which can be broken down into: 4 cases of personality problems (15.6%), 3 of substance misuse (9.4%), 5 of depression (15.6%), 1 anorexia (3.1%) and 1 pseudoseizure (3.1%). 3 patients had no significant psychiatric history (9.4%) and no information was available for 16 cases (50%). In terms of family history, 28 had no information available (87.5%) and 3 had diabetes (9.4%). Background and other risk factors were described in 20 cases (62.5%). This included 2 patients with diabetic friends (6.3%), 2 with ties to medicine e.g. training or through a parent's occupation (6.3%) and 3 reports of abuse growing up (9.4%). The remainder was a mix of findings (given in Table 1).

DISCUSSION

Demographics

The classical patient with FD has been described as being a middle-aged female of Caucasian ethnicity, with diabetes, a past psychiatric history and ties to the medical profession (Grunberger et al. 1988, Jaghab et al. 2006, Awad and Ilahi 2019, Savino and Fordtran 2006). Interestingly, our case agreed with some of these characteristics and challenged others; he was a quadragenarian of Asian ethnicity with diabetes, a past psychiatric history, but no obvious ties to medicine. Our search showed that there are indeed a number of reports that conflict with the current 'paradigm'. This again, highlights the need to think widely when it comes to FH.

Age & Sex

Although FH has been described in a variety of ages, for example from as young as 8 to as old as 72, our review of 32 patients revealed that the average age amongst adults was indeed 33.7 years or middle-aged (Libow 2000, Schuler et al. 1989). In terms of sex, we found that 26 out of 32 cases of FH were female – a ratio of 4.3:1. Whilst we do not fully understand why there is a higher incidence of FH in women than men, it has been reported that women in general have a higher rate of psychiatric illness (Riecher-Rössler 2010). There are many possible reasons for this, perhaps relating to differences in sex hormones, exposures to abuse or discrimination and differing familial and societal stresses (reviewed in Riecher-Rössler 2010).

Ethnicity

Ethnicity was less frequently reported than age or sex. However, from the few papers that mentioned it, it seemed to be very variable and included Caucasian, Afro-Caribbean and even Hispanic patients (Lebowitz and Blumenthal 1993, Huddle et al. 1984, Patel and Daniels 2018). We thought to therefore look at the where the paper was written as a surrogate of ethnicity where it is was unspecified. This showed a worldwide distribution with the majority of papers in the USA and Europe, and few in Asia and the Middle East (data not shown). Doctors across the globe should therefore be aware of FH. These findings are to be taken with caution though, as the ability to produce scientific papers depends on many factors within a country such as research funds or resources, as well as publishing practises. It could therefore be publication bias that Caucasian populations simply report FH more often, or other populations report it less often (rather than a true difference in incidence). It remains that classically associating FH with the Caucasian population may be misleading, so we urge medical professionals to avoid this stereotype.

Occupation

As outlined above, FH is commonly associated with ties to the medical profession. Our results corroborate this; we found that 37.5% of patients had medical occupations, most commonly as nurses, or past medical training (Table 1). In one case the link was less direct, as it instead involved his parents (his father was a doctor and his mother a nurse (Bretz & Richards 2000). We postulate that the reasons behind this are three fold: access to insulin, medical knowledge on how to administer insulin and the effects it produces and increased exposure (and so desire) to be in the sick role (DSM-5 2013). The sick role enables patients to escape the ordinary constraints of working life, receive attention and gain sympathy from others (Parsons & Turner 2005).

Occupation may also be important from a socio-economic stand-point as our study identified a high incidence of FH in unemployed patients dependent on benefits (12.5%). Financial strain is undoubtedly a major stressor in many people. It remains to be elucidated whether parental occupation or socioeconomic status growing up relates to FH.

Medical & Psychiatric history

53.1% of patients reviewed were reported as having diabetes mellitus which may explain the ease of access to insulin in these cases. On the other hand, 12 had no previous history of diabetes (37.5%). A minority of these may be accounted for by having other diabetics in the family (9.4%) or friends with diabetes (6.3%). Moreover, many of the cases which did not have diabetes were medical professionals (see above) providing an alternate means of access. It remains that whilst FH is usually a condition seen in diabetics, it is not unique to this population. Aside from this, 16 (50%) patients had no information regarding past medical history, whilst 4 (12.5%) were stated to have no significant past history. The former represents an unfortunate problem in patient reporting; without a decent number of papers documenting a full history, we cannot determine whether any other medical conditions are commonly co-morbid in FH.

In line with the classical history of FH, we found that 12 patients had a positive psychiatric history (40.6%) which can be broken down into three four main categories: personality problems (15.6%), substance misuse (9.4%), depression (15.6%) and other (6.3%). The finding that many cases are co-morbid with depression is especially important as this has been linked to an increased number of attempts or completed suicide (Schober et al. 2008, Roy & Roy 1995). The same association has been made with non-insulin substance abuse (Cassidy et al. 1999). In addition, there have even been reports of euphoria regarding self-injection in one case (Jordan et al. 1977). Our literature review also revealed one case in which the patient reported feeling ashamed of his diabetes, which may have contributed to his surreptitious use of insulin (Berkowitz et al. 1971). Furthermore, 3 patients had no significant psychiatric history (9.4%) and no information was available for 16 cases (50%). The latter may represent either a failure to elicit a thorough psychiatric history in these cases, or omissions by the authors.

Other risk factors

The background of these patients and other risk factors were described in 20 cases (62.5%). Some of the most common themes that arose from this was instability within the home with involvement of marital disputes (6.3%), childhood abuse (9.4%) and foster care (9.4%). These may precipitate FH by inducing stress, or through the desire to avoid stressors. The latter two categories are intrinsically linked, and it has long been recognised that childhood abuse increases the incidence of a variety of psychiatric problems in adulthood. More specifically, it has been suggested that many cases of factitious disorder actually start in early adolescence before they are diagnosed in adulthood, and may even predispose to other conditions such as factitious disorder imposed on another (Sheridan 2003). This raises the question as to whether an early 'cause' in childhood may be responsible. It also emphasises the importance of long-term follow-up in these patients.

Features

Hypoglycaemia, irrespective of cause, presents with a variety of organic symptoms including: fatigue, hunger, sweating, paraesthesia, tremor, dizziness, palpitations, irritability and altered mental state. As the blood glucose concentration falls, these symptoms increase in incidence, severity and duration. In severe cases, patients can even suffer complications such as seizures, brain damage, coma and death. This topic has been a subject of multiple reviews (Kittah and Vella 2017, Freeland 2017). However, in FH patients may also exhibit additional features which may distinguish it from organic causes (Table 2).

Table 2. Additional Features of Factitious Hypoglycaemia

Category	Features
Features of substance abuse or addiction	Excessive bruising, infections, lipohypertrophy
	Use of other drugs, addiction
Features of psychiatric illness, trauma or abuse	Self-neglect, depression, suicidal ideation/attempts, lability
	Repeated admissions, lengthy medical notes, many surgical scars
Features of poorly managed diabetes (if diabetic)	Neuropathy, gastroparesis, repeated diabetic ketoacidosis, diabetic retinopathy, diabetic nephropathy, polyuria, polydipsia, lethargy

Differentials

Classically, the findings of Whipple's triad: symptoms of hypoglycaemia, hypoglycaemia (blood glucose level <50 mg/dL), and relief of symptoms following ingestion of glucose, are associated with an underlying diagnosis of insulinoma (Whipple 1938). However, these criteria are terribly non-specific and hold true for many other differentials including factitious hypoglycaemia. It may also be seen for example, in cases of neisidioblastosis, now termed 'congenital hyperinsulinism' (Schuler et al. 1989). This is a condition in which overactivity of the pancreatic beta cells leads to hyperinsulinaemia and hypoglycaemia. More recently, It has been discovered that recurrent hypoglycaemia occurs in some patients due to anti-insulin antibodies.

One must also consider more simply the complications of diabetes, which is often co-morbid in FH and provide patients with a means of obtaining insulin and/or hypoglycaemia drugs. It has been postulated that severe gastroparesis can cause a mismatch in the timing between insulin release and digestion such that hypoglycaemia ensues. Moreover, lipohypertrophy at injection sites may interfere with insulin administration. These patients may inject additional doses to try and correct their hyperglycaemia and overshoot, resulting in accidental hypoglycaemia. (Loh et al. 2014)

One other important differential to consider is factitious disorder imposed on another (Munchausen's by proxy). The victims of this disorder can present very

similarly to FH and may even have identical investigations. The only distinction is that somebody other than the patient is physically injecting the insulin.

Investigations

Investigating a patient for FH is often a long, grueling process that is only initiated after several episodes or repeat admissions. Often patients will avoid the same centres or doctors, such that it is hard to piece together the puzzle. This and a lack of awareness amongst doctors with a limited exposure to psychiatry, have led to a long time to diagnosis. Indeed, there are papers where it has taken several months and even decades to realise this diagnosis (Schuler et al. 1989). It could also be that physicians are cautious about labelling patients with FH as it is a stigmatising diagnosis with social, legal and clinical implications (Loh et al. 2014). Alternatively, or in addition, many people do not like confrontation which is necessitated in suspected cases of FH. Whilst many patients react positively to this by stopping insulin misuse, others may become violent (Bretz and Richards 2000). Lastly, we suggest that doctors may be misled by previous improper diagnoses such as 'brittle' or 'labile diabetes', such that the underlying, true FH has gone undiagnosed. Whatever the reasons may be, medical professions need to be better able to recognise, investigate and treat FH

Changing practice

In the past, investigating suspected cases of factitious hypoglycaemia was comparatively easy. Investigators had to simply test whether the hyperinsulinaemia was animal (bovine) or human in origin. With the introduction of human insulin analogues, doctors instead began radio-labelling vials of exogenous insulin to distinguish it from endogenous insulin. These supply vials were discreetly replaced in anticipation of surreptitious use and the patient's urine radioactivity measured (Berkowitz et al. 1971, Whelton et al. 1968). Meanwhile, anti-insulin antibody assays became increasingly popular (Scarlet et al. 1977). Unfortunately, other teams favoured a more invasive approach including proceeding to perform multiple exploratory laparotomies and pancreatic resection (Berkowitz et al. 1971). Fortunately, the rush to do these risky procedures has now been largely abolished by the advent of C-peptide (Melani et al. 1970). C-peptide is a component of pro-insulin that is released alongside active insulin by the pancreatic beta cell, such that endogenous release and exogenous administration can be distinguished. Whilst C-peptide remains a useful test today, its role in diagnosing factitious hypoglycaemia is dwindling as alternative means of induction (such as sulfonylureas) increase in prevalence. For instance, in sulfonylurea induced FH the C-peptide level will appear congruent with the insulin level as it causes endogenous insulin release (rather than exogenous administration). Centres have therefore begun to use terbutaline testing and sulfonylurea urine assays (Walfish & Kashyap 1975). In addition, with the increasing use of insulin pumps, especially in children, we must also interrogate the device output and even its priming history (Osipoff et al. 2010). Clearly this is an evolving area and it is likely that the standard work-up for FH will soon be updated yet again.

Aside from medical investigations, one should also perform a thorough psychiatric and environmental assessment. Although the literature reports a variety of weird and wonderful hiding places for insulin, it remains that many patients simply hide extra insulin in their pockets or within hospital rooms (Whelton et al. 1968, Giordano and Rainwater 1986, Schade et al. 1985). This is recognised by the DSM5 which stats that the identification of a factitious disorder is usually made in one of four ways: 1. The patient is accidentally discovered in the act, 2. Incriminating items are found, 3. Laboratory values suggest non-organic aetiology, OR 4. The diagnosis is made by exclusion (DSM-5 2013).

Management

In the acute setting all hypoglycaemia patients, including those with FH should be given glucose to correct the hypoglycaemia. This step in medical management is ubiquitous. However, comparatively few papers were identified that focused on the need for psychiatric treatment. Psychotherapy is currently recommended to treat factitious disorder, and it has been shown that combining behaviour modification and dynamic therapy can be successful (Eastwood & Bisson 2008, Yassa 1978). Unfortunately, not all patients engage with this though (Jermendy 1996). Family support may also play a key role in management (Walfish 1987). Equally, it is important to manage any co-morbidities and risk factors as described in Section 6.1: (Demographics), as these may exacerbate the condition. This is especially true of depression and suicide risk as a significant number go on to commit suicide. Ultimately though, it seems that confrontation with irrefutable evidence is the best 'curative' measure, though should be done in a controlled manner with caution (Bretz & Richards 2000, Marchetti et al. 1988). In terms of outcome, there is a lack of reporting of long-term follow up in these patients. When it has been reported, the results have been unpredictable. For example, in one study patients were followed-up for 15 years and only 3 returned to a productive life. Unfortunately, 2 committed suicide (Gruberger et al. 1988).

CONCLUSION

FH is associated with significant morbidity, mortality and strain on healthcare systems. It is therefore important to keep an open mind as to which patients it may affect. The current investigations for a patient with recurrent hypoglycaemia have evolved over time, and if FH is suspected these patients should also undergo a thorough psychiatric assessment. Psychotherapy is currently recommended as the treatment of choice in

these cases. Integrated provision of services is paramount to ensure patients are supported well from both a physical and a mental health point of view. Currently, there is limited follow-up data to inform robust management of FH and there is a clear need for further research. We hope to see several advances in this field in future.

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Contribution of individual authors:

Harry Kyriacou & Maria Filippidou came up with the idea of this manuscript.

Maria Filippidou provided the case report.

The literature search was equally divided, performed and analysed by Harry Kyriacou, William Duggleby, Adam Hatoum, Tahir Khan & George Manley.

Harry Kyriacou wrote the draft of the manuscript and all authors were involved in editing.

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