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Contemporary Review

Otolaryngology Fantastica: The Ear, Nose, and Throat Manifestations of Munchausen's Syndrome

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Objectives/Hypothesis: Munchausen's syndrome (MS) is a form of severe, chronic, factitious disorder with physical symptoms. Some essential features define MS, such as recurrent, feigned, or simulated illness; peregrination (traveling or wandering); pseudologia fantastica; and drug abuse. Munchausen's syndrome by proxy (MSBP) classically involves a parent or other caregiver who inflicts injury or induces illness in a child. The aim of the present study was to summarize and study the main ear, nose, and throat (ENT) manifestations of MS and MSBP.

Study Design: A systematic literature review carried out in a tertiary university referral center.

Methods: An appropriate string was run on PubMed to retrieve articles dealing with ENT manifestations of MS and MSBP. A double cross-check was performed on citations and full-text articles found using selected inclusion and exclusion criteria.

Results: In total, 24 articles were finally included in the study, describing 30 cases of MS or MSBP involving the ENT region; 15/30 (50%) cases involved the face, most often presenting as facial pain or facial swelling; and 7/30 (23.3%) cases presented with symptoms involving the ear. Six cases out of 30 (20%) were MSBP.

Conclusions: MS and MSBP may present with symptoms involving the head and neck area, particularly the face and external ear canal. The ENT specialist should suspect MS in patients with strange and long-lasting symptoms, so as to avoid misdiagnosis and unnecessary treatments that waste time and money in the healthcare sector.

Key Words: Factitious disorders, Munchausen's syndrome, self-inflicted disease, Munchausen's syndrome by proxy.

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INTRODUCTION

The DSM–IV gives three diagnostic criteria for factitious disorder: 1) the intentional production of physical or psychological signs or symptoms; 2) the motivation to assume the sick role; and 3) the absence of external incentives.[1] The initial differential diagnosis includes a variety of medical and psychiatric conditions. Among the psychiatric diagnoses that should be considered are somatization disorder, malingering, and factitious disorder.[2] In somatization disorder, the patient will frequently offer multiple physical complaints (beginning at a young age and persisting for several years) that result in treatment-seeking or in decreased function. Patients with somatization disorder neither consciously lie about their symptoms nor intentionally cause their illness (e.g., by self-administering medications); they also do not complain of symptoms to receive an external reward.[3] Malingerers[4] intentionally feign physical or psychiatric illness or produce abnormal physical signs for a secondary gain. This gain may include financial benefits, prescriptions for controlled substances, or avoidance of police or military duty. Factitious disorder involves the intentional production of symptoms for a primary gain[2] (i.e., to satisfy a psychological need to maintain the sick role and to receive attention or support). An important facet of this diagnosis that distinguishes it from other syndromes is that the symptoms are consciously produced but do not result in external gain.

Munchausen's syndrome (MS) is a form of severe, chronic, factitious disorder with physical symptoms first characterized by Asher[5] in 1951. He named the syndrome after Baron von Munchausen, an 18th century gentleman who wandered from city to city dramatically embellishing his minor mercenary military adventures during Turk-Russian clashes into intricate self-aggrandizing tales of heroic adventure.

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MS has specific features that distinguish it from other forms of factitious illness. In general, patients with MS have more extreme presentations and a more refractory illness; outcomes are generally worse for those with MS than for patients who have milder forms of a factitious disorder. Folks and Freeman[6] described three essential features of MS: 1) recurrent, feigned, or simulated illness; 2) peregrination (traveling or wandering); and 3) pseudologia fantastica, a form of pathological lying characterized by wildly exaggerated stories told for the seeming benefit of the listener. In most patients affected by MS, drug abuse is reported; they present demanding specific treatment, such as analgesics, or they often have a known drug addiction problem. It is of interest that sudden cessation of therapy with improved clinical signs is seldom followed by withdrawal effects.[7]

In 1977, Roy Meadow,[8] a pediatric nephrologist, first described a condition he subsequently termed Munchausen's syndrome by proxy (MSBP). The classic form involves a parent or other caregiver who inflicts injury or induces illness in a child, deceives the treating physician with fictitious or exaggerated information, and perpetrates the trickery for months or years. A related form of pathology is more insidious and more common but also damaging. It involves parents who fabricate or exaggerate symptoms of illness in children, causing overly aggressive medical evaluations and interventions.[9] Differential diagnosis in these cases must be made with child abuse.[10] MSBP should be suspected whenever persistent and recurrent unexplained illness afflicts a child.[11]

Using a systematic literature review, the aim of the present study was to summarize the main ear, nose, and throat (ENT) manifestations of MS or MSBP. This review is also intended to contribute to the awareness among otolaryngologists of MS as a differential diagnosis of some chronic and inexplicable ENT pathologies. It is important that every health professional, even specialists, suspect this kind of pathology in selected cases, as a misinterpretation of MS may prove very wasteful for the healthcare sector in terms of time and money. Failure to reach a definite diagnosis can be frustrating for healthcare professionals and possibly harmful to the patients.

MATERIALS AND METHODS

PRISMA 2009 guidelines were considered and applied whenever possible in this systematic review. The following search string was run on PubMed: ("Munchausen Syndrome"[Mesh] OR "Munchausen Syndrome by Proxy"[Mesh] OR "Factitious Disorders"[Mesh]) AND ("Ear"[Mesh] OR "Ear Canal"[Mesh] OR "Ear Auricle"[Mesh] OR "Nose"[Mesh] OR "Nose Diseases"[Mesh] OR "Epistaxis"[Mesh] OR "Pharynx"[Mesh] OR "Pharyngitis"[Mesh] OR "Neck"[Mesh] OR "Neck Pain"[Mesh] OR "Neck Injuries"[Mesh] OR "Face"[Mesh] OR "Facial Pain"[Mesh] OR "Larynx"[Mesh] OR "Laryngeal Diseases"[Mesh] OR "Tracheostomy"[Mesh] OR "Paranasal Sinuses"[Mesh] OR "Submandibular Gland"[Mesh] OR "Submandibular Gland Diseases"[Mesh] OR "Parotid Gland"[Mesh] OR "Parotid Diseases"[Mesh] OR "Parotid Region"[Mesh] OR "Tongue"[Mesh] OR "Tongue Diseases"[Mesh] OR "Palatine Tonsil"[Mesh] OR "Adenoids"[Mesh] OR "Vocal Cords"[Mesh] OR "Lip"[Mesh] OR "Lip Diseases"[Mesh] OR "Palate, Soft"[Mesh] OR "Head"[Mesh] OR "Head Injuries, Closed"[Mesh] OR "Head Injuries, Penetrating"[Mesh] OR "Mouth"[Mesh] OR "Palate"[Mesh] OR "Palate, Hard"[Mesh] OR "Deglutition Disorders"[Mesh]). Further searches were included using the option "titles in your search terms."

After running the above search string, abstracts and titles obtained were screened independently by two of the authors (M.A.C. and M.R.), who subsequently met and discussed disagreements on citation inclusion. Inclusion criteria for citations were diagnosis of Munchausen; description of cases with ENT manifestations; and English, Spanish, Italian, and French language. Exclusion criteria were clear unrelated pathologies and Munchausen syndrome involving areas other than ENT.

Then the full texts of the articles identified were obtained for a second screening again by the same two authors, who met and discussed disagreements on article inclusion. Inclusion criteria for full text articles identified were diagnosis of Munchausen and description of cases with ENT manifestations. Exclusion criteria were lack of definite diagnostic criteria of Munchausen; suspicion of diagnosis of malingering, simulations, hypochondria, or other factitious disorders; and lack of sufficient clinical data.

A further manual check of the references included in the articles was performed. The final number of articles included in the present review was identified, and the main information was extracted and summarized.

RESULTS

Running the above search string in PubMed, 87 articles were identified. Further references were included by using the option "titles in your search terms." After an initial check, full-text retrieval, and manual cross-checking of references included in the articles, 24 articles (describing a total of 30 cases overall) were finally included for qualitative synthesis in the study (Fig. 1). Of these, 18/30 (60%) were MS, whereas 6/30 (20%) were MSBP. The mean age of patients was 29.9 years (±7.6 standard deviation [SD]) for MS and 7.3 years for MSBP. In total, 16/30 patients (53.3%) were male and 8/30 (26.7%) were female. The mean duration of symptoms was 86.4 (±81.1 SD) months, ranging from a few weeks to 240 months (20 years).

In total, 15/30 (50%) cases involved the face, most often presenting as facial pain or facial swelling, whereas 7/30 (23.3%) cases presented with symptoms involving the ear. This percentage drastically increased in the case of MSBP, in which more than 80% of cases came to the attention of the ENT specialists through some ear problems (5/6, 83.3%). In 10/30 (33.3%) cases, the symptoms were simulated, whereas in the remainder symptoms were directly provoked causing real lesions. In 7/30 (23.3%) cases, ENT manifestations were associated with symptoms involving other areas. The maximum of known surgical procedures that a patient underwent was 18, and 37 hospital admissions were reported in another patient. The case summaries are shown in Table I.

DISCUSSION

There have been no studies that have specifically assessed the prevalence of MS, and the prevalence of

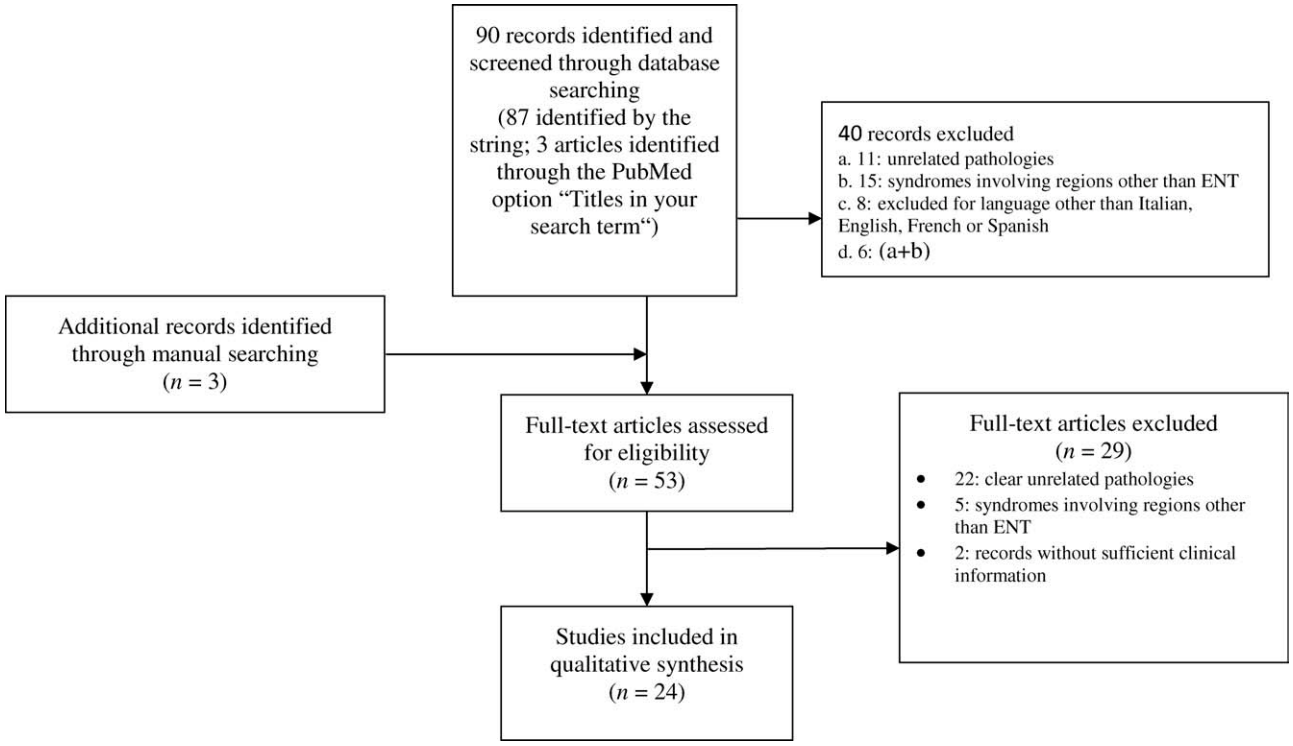

Fig. 1. Flow diagram for the study. ENT = ear, nose, and throat.

factitious disorder and MS might be underreported.[2] One reason for this may be that physicians in other medical specialties are often unaware of the psychiatric nomenclature used for these syndromes; they may recognize that the patient is consciously deceiving others to maintain the sick role, but may not know the formal difference between malingering, factitious disorder, and somatoform disorders.[2]

In general, those with MS tend to be men, to have a lower than average socioeconomic status, and to appear socially isolated (both by history and by a notable lack of visitors during their hospitalizations).[12] Also in our review, a slightly higher prevalence of males was noticed (16/30; 53.3%).

More often than not, individuals with MS have some medical training and/or medical sophistication on which the simulation and fabrication of symptoms is based. Such patients often use medical terminology to intimidate physicians.[2] Also in the present review, in at least two articles reviewed, patients presented with a fluent medical terminology, although these data could have been underestimated.

The mean duration of symptoms in our review was approximately 7 years, ranging from a few weeks to 240 months (20 years), whereas the maximum of known surgical procedures that a patient underwent was 18, with 37 hospital admissions reported in another patient. The reader should certainly reflect on the importance of reducing the enormous costs that such conditions could demand and be aware of the condition so that a specialist in the healthcare sector does not waste time and money.

From our review, two main ENT regions were involved: in one half of the cases the face was involved, most often presenting as facial pain or facial swelling, whereas many patients presented with symptoms involving the ear (23.3%). Altogether, the face and ear represented almost 75% of ENT areas involved in MS. These data should be kept in mind because they might aid in the diagnosis of MS. It is worth underlining that in one out of three cases, the symptoms were simulated, whereas in the remainder, symptoms were directly provoked, causing real lesions. A curious finding is that, very often in our review, a deep injection of air or other substances was reported or suspected of producing symptoms, leading to emphysema or abscess formation.

MS has generally been associated with abdominal procedures (appendectomy, cholecystectomy, adnexectomy); however, it can also present during the treatment of post-traumatic or surgical wounds.[13] Based on our results, ENT manifestations can also be associated with symptoms involving other areas, and in fact some of the patients underwent abdominal procedures, although several other conditions can be associated with ENT manifestations, as shown in Table I.

In cases where MS is suspected, patients should be evaluated by a psychiatrist who would possibly make a definite diagnosis. The most important role of the psychiatrist in the treatment of MS is to help the primary treatment team manage the patient in the safest and most appropriate way.[2,6,12] Such patients tend to produce strong countertransference feelings and have comorbid borderline personality disorder; physicians typically react intensely to such patients. The patient's

TABLE I.
Clinical Summary of Study.

Source	MS/ MSBP	Sex	Age	ENT Manifestations	Associated Manifestations/ Clinical History	Time From First Symptoms to Diagnosis	Notes/Comments
Oldham[14]	MS	M	26	Facial pain, swollen face	Bilateral conjunctivitis, weight loss, left facial hypoaesthesia, right body hypoaesthesia, right-sided weakness	4 yr	Six different jobs, nine hospital admissions, medical assistant on a ship, simulated symptoms of the disease
Patterson et al.[15]	MS	F	33	Acute dyspnea and stridor	Tachypnoeic	5 yr	15 hospital admissions, simulated symptoms of the disease
Rhys Evans[16]	MS	M	24	Neck pain, increasing dysphagia, blood-stained saliva	Previous admissions with acute abdominal symptoms	6 yr	37 hospital admissions, simulated fish bone ingestion (laryngoscopy and lateral neck x-ray: negative)
Pender and Pender[17]	MS	M	22	Airway distress and hemoptysis	Previous history of epilepsy, previous tracheotomy in childhood	7 yr	Patient under arrest, dishonorable discharge from Marines, simulated symptoms of the disease
Mahler and Ginath[18]	MS	M	31	Unpleasant facial appearance (referred by patient)	Disturbance in self- and sexual identity	14 yr	10 surgical procedures on his nose and chin, 13 plastic surgeons consulted
Winans et al.[19]	MS	F	23	Recurrent facial swelling and emphysema	Proptosis of the right eyeball, blindness	10 yr	Several admissions for emphysema in other parts of the body, suspected self-injection of air (syringes in her room)
Bourchier[20]	MSBP	M	2.5	Recurrent otorrhagia	Poor weight gain, hematemesis, melaena, cough, unsteady gait	2 yr	Her mother introduced her blood into the child's ears (ear blood and mother's: same blood group)
Michalowski[21]	MS	F	20	Bleeding from the lip, cheilitis glandularis	Thrombocytopenia	13 yr	12 hospital admissions, self-inflicted lip wound
Zohar et al.[22]	MSBP	M	5	Bilateral chronic external otitis	Several bilateral paracenteses, bilateral mastoidectomy	2 yr	Divorced mother's victim, inflicted ear canal lesions by his mother
	MS	F	25	Recurrent acute external otitis	Foreign body (soil clay) in the external meatus	—	Divorced woman, suspected self-contamination
	MSBP	M	5	Recurrent external otitis	Chalk in external meatus	—	Suspected chalk introduction in son's ears by father, father with history of Munchausen's syndrome

(*Continued*)

TABLE 1.
(*Continued*).

Source	MS/ MSBP	Sex	Age	ENT Manifestations	Associated Manifestations/ Clinical History	Time From First Symptoms to Diagnosis	Notes/Comments
	MS	F	42	Facial swelling	—	20 yr	25 hospital admissions, silicon ointment self-injected by needles
Mastrocola et al.[23]	MS	M	41	Left-sided facial pain	Dental caries	9 yr	More than 12 oral and ENT surgeries, simulated symptoms of the disease
	MS	M	45	Right-sided facial pain	Toothache	—	Five endodontic treatments, simulated symptoms of the disease
Thomas[7]	MS	M	26	Recurrent facial swelling and pain, trismus, dysphagia	Swinging pyrexia	8 yr	18 surgical treatments in 8 years, admitted self-introduction of needle in his cheek
Fishbain et al.[24]	MS	M	36	Chronic neck pain	Low-back pain	3 yr	Divorced, patchy job history, suicide attempts, simulated symptoms of the disease
	MS	F	35	Facial pain	—	—	12 previous surgeries, patchy job history, suicide attempts, simulated symptoms of the disease
Cirigliano et al.[25]	MS	F	22	Recurrent right-sided facial lesions	Neuralgic pain, periorbital edema, regional adenopathy	18 yr	Several hospital admissions, admitted self-infliction of lesions by graphic art tools (art student)
Magnay et al.[26]	MSBP	F	7 mo	Excoriated lesions around the nostrils	Excoriated perianal lesions, diarrhoea, bloody stools, thrive failure, foot ulcers	3 mo	Mother pinched and scratched child's skin, self-infliction referred by the mother (with MS)
Paar[27]	MS	F	35	Bilateral external otitis	—	16 yr	Fear of AIDS, sexual abuse during a homeopathic consultation (reported), suspected self-inflicted lesions
Scully et al.[28]	MS	M	31	Left maxillary pain	—	4.5 yr	Several endodontic treatments, simulated symptoms of the disease
	MS	F	35	Right submandibular gland chronic pain, right glossopharyngeal neuralgia	—	15 yr	Fluent in medical terminology, simulated symptoms of the disease
Tyler et al.[29]	MS	F	20	Recurrent aphthous stomatitis, geographic tongue	Mandibular subluxation, periorbital ecchymoses	4 yr	Painful folds in the cheeks referred, suspected self-induced injuries

(*Continued*)

TABLE 1.
(*Continued*).

Source	MS/ MSBP	Sex	Age	ENT Manifestations	Associated Manifestations/ Clinical History	Time From First Symptoms to Diagnosis	Notes/Comments
Gadre et al.[30]	MS	F	36	Recurrent left-sided facial swelling	Decreased vision, left eye proptosis, fever, persistent pneumothorax	—	Fluent in medical terminology, syringes and needle possession: suspected self-injection
DiBiase et al.[31]	MSBP	M	3	Chronic left otorrhea	Fever, hypotension	—	Mother injected fecal fluid into child's central line during the hospitalization
Somani[32]	MSBP	M	28	Burning sensation over left cheek and ear	—	10 d	Adult victim, acid pouring during the night by his wife
Solomon and Lipton[33]	MS	M	34	Head and facial pain	Migraine, cluster headache, trigeminal neuralgia	4 yr	Simulated symptoms of the disease
Juaneza and Isaac[34]	MS	F	35	Hematemesis, epistaxis	Tachycardia, lower extremity ecchymoses, vaginal bleeding, elbow and back pain	11 yr	Suspected rat poison (warfarin) ingestion, history of sexual abuse (referred)
Tosun et al.[35]	MS	M	21	Recurrent subcutaneous cervicofacial emphysema	Dyspnea during swelling attacks	4 mo	Drug abuser, suspected self-injection of air subcutaneously
Hojjati et al.[36]	MS	F	20	Facial subcutaneous emphysema	—	2 yr	Patient found self-injecting air into her face with a syringe

MS = Munchausen's syndrome; MSBP = Munchausen's syndrome by proxy; ENT = ear, nose, and throat; M = male; F = female.

demands and ability to shame the physician can lead the physician to order unnecessary procedures, to dispense addictive or dangerous treatments, or to make errors or omissions in medical care.

A treatment strategy has been proposed that suggests these patients be blacklisted by hospital associations. Trials have been reported with hypnosis, insulin coma, electroconvulsive therapy, and lobotomy but without long-term success.[2,6,12] In general, the prognosis for patients with MS appears to be poor from a psychiatric point of view. Flexible and creative approaches that emphasize consistency and regular outpatient psychiatric care have been associated with the most success.[2,6,12]

Six out of 30 (20%) cases in our series were MSBP. One case of an adult victim of MSBP was retrieved. The mean age of victims of MSBP was 7.3 years, and compared to MS, the percentage of ear manifestations drastically increased in the case of MSBP, where more than 80% of cases came to the attention of the ENT specialist through ear problems (5/6, 83.3%).

Concerning the victims of MSBP, by recognizing that this problem is a form of child abuse taking place in a medical setting, the medical team (psychiatrist or clinicians) should contact child protective services agencies that are mandated to keep children who are abused (sexually, physically, or psychologically) safe regardless of whether the abuse occurs in the home or the hospital.[10] It is also important to underline the importance of awareness not only of MS, but also of MSBP, rarer and only recently recognized by the scientific community.[8] We believe that physicians can recognize caregiver-induced illness cases of MSBP if they maintain a reasonable awareness of the condition. When a child's disease is chronic, recurring, confusing, incomprehensible, and unresponsive to typical treatments, MSBP should receive consideration.[9]

CONCLUSION

MS and MSBP may present with symptoms involving the head and neck area. The ENT specialist should suspect MS in patients with strange and long-lasting symptoms, particularly involving the face and external ear canal, so as to avoid misdiagnosis and unnecessary treatments that waste time and money in the healthcare sector. MSBP should be suspected whenever persistent and recurrent unexplained illness affects a child, particularly involving the external ear canal.

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