



A Case Report of Recurrent unilateral Bell's Palsy in young female

Khlood Mohammad Aldossary*

Department of Pharmacy Practice, College of Pharmacy, Princess Nourah bint Abdulrahman University, Riyadh, Saudi Arabia



Article History:

Received on: 01 Apr 2020
Revised on: 30 Apr 2020
Accepted on: 20 May 2020

Keywords:

Facial nerve,
Facial paralysis,
Recurrent Bell's palsy

ABSTRACT

Most facial palsies comprise of lower motor neuron type or Bell's palsy. The mean incidence of recurrence of Bell's palsy is 6.5% and can recur at any age. Although it was first reported in the year 1871, most of its etiology, pathology, management and progression are still neither fully described nor reported. We reported a case of Bell's palsy in a young Saudi female who was presented with a unilateral recurrent episode of Bell's palsy. Patient came to the emergency department with facial asymmetry, severe neck pain, loss of sensation on the left side of face and headache, while she was also not responding to oral medications. She was admitted to the hospital for an intravenous treatment and for further evaluation. Her serologic investigation was unremarkable. Similar episode had occurred five years ago as well, which was completely resolved after treatment. Her brain MRI localized T2 and T2 weighted sequence showed hyperintense lesion at the superior anterior aspect of left orbit and could represent lipoma versus hemangioma. Her presenting complaint responded well on IV treatment and patient was discharged on 11th day. She was also referred for plastic and neurosurgery services for the treatment of underlying cause. Recurrent Bell's palsy is infrequent with etiology not fully elucidated. However, our case emphasizes that the timely medical treatment with regular follow-up is necessary for better prognosis and identification and treatment of underlying cause.

*Corresponding Author

Name: Khlood Mohammad Aldossary
Phone:
Email: Kmalossary@pnu.edu.sa

ISSN: 0975-7538

DOI: <https://doi.org/10.26452/ijrps.v11i3.2546>

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INTRODUCTION

An acute, asymmetrical and idiopathic paralysis of the VII (facial) nerve is known as Bell's palsy (Gilden, 2004). Sir Charles Bell (1774-1842) defined paralysis of cranial VII nerve along with its anatomical features and function and hence the disease was named

after him as Bell's palsy (Greco *et al.*, 2012). Also known as idiopathic facial paralysis (IFP), around 60-80% of all the lower motor neuron facial palsies are Bell's palsy (Kubik *et al.*, 2012). Of all the facial palsies, around 4-7% cases are of recurrent Bell's palsy (Pitts *et al.*, 1988). The mean incidence of recurrent Bell's palsy, i.e. in patients who were affected previously as well, is 6.5% (Dong *et al.*, 2019). It was first reported in 1871 but its etiology, pathogenesis, management and prognosis are still not clearly reported (Dong *et al.*, 2019). The exact cause of recurrent idiopathic Bell's palsy in an individual is not well studied, although some studies have reported its association with malignant hypertension, diabetes, autoimmune disease and pregnancy (Scola *et al.*, 2004). In literature, it has been categorized as a unilateral recurrent or non-recurrent, simultaneous or alternating or recurrent bilateral. Usually, it recurs after more than one-year interval, whereas two or more relapses in a patient

are less common. There is a very rare chance of more than four relapses in a single patient (Pitts *et al.*, 1988). Unilateral recurrent type of Bell's palsy is rare and requires further evaluation to rule out the causes like malignancy (schwannoma) (Pitts *et al.*, 1988).

CASE REPORT

History

A 22 years old young Saudi woman presented at the emergency department in February 2019 with a complaint of severe neck pain and stiffness with an inability to move her neck, loss of sensation and facial drop for two hours. These symptoms developed suddenly after she took a bath. Upon examination, she was vitally stable and conscious, had torticollis, left-sided facial palsy, deviation of the mouth to the left with a loss of nasolabial fold and weakness of closure of the left eye. She was diagnosed with Bell's palsy and received intravenous diphenhydramine, benzotropine, paracetamol and discharged with oral medication Prednisolone 60mg and Acyclovir 400mg. She came to the emergency department again the very next day with the same complaint, and she could not tolerate the oral medication along with left-sided headache and ear pain with tinnitus. However, her headache responded to analgesics. She was vitally stable and was then admitted to the hospital for further investigations. She had a history of similar complains five years ago, but back then it resolved completely the same day after she had received treatment. She also had a history of infantile hemangioma on the left side of the face that was treated with laser and Steroid injections 10 years ago. She had an amblyopic left eye since early childhood. She also had a history of left blepharoplasty (10 years ago) and cosmetic surgery for nasal fold (8 months ago). She was found to be non-diabetic, non-hypertensive and had no other chronic disease and no history of any rash. Serum investigations have been presented in Table 1.

Neurological examination

Her neurological examination showed facial asymmetry, absent forehead wrinkles, weakness of closure of the left eye, deviation of the mouth to the left with a loss of left nasolabial fold and was not able to clench her teeth. Her sensory examination showed a sensory deficit on the left facial side (beyond fifth nerve distribution) but no limb hemisensory. Extraocular eye movements were full, with no nystagmus. She has divergent strabismus (since early childhood). The remaining neurological examination was unremarkable.

Radiological examination

Her brain's CT with contrast showed heterogeneity and foci of low density involving the left parotid gland superficial lobe. The rest of the scan was unremarkable. All other labs were within normal ranges.

Her tympanogram showed normal results bilaterally. Pure tone audiometry showed normal hearing threshold at 250-8000 Hz in the right ear while the left ear has profound sensorineural hearing loss at 250-8000 Hz.

Ultrasound of the parotid gland showed minimal enlargement of left parotid as compared to the right parotid with no edema or hypervascularity or no parotid masses or lymphadenopathy detected.

Magnetic Resonance Imaging (MRI) the brain and Internal Auditory Canal (IAC) + Internal Auditory Meatus (IAM) reported multiple 2 hyperintense foci involving the lateral aspect of left parotid gland superficial lobe with suppression of the signal on FAT SAT sequence and could be related to lipomatosis versus hemangioma. Localized T2 and T2 weighted sequence showed hyperintense lesion at the superior anterior aspect of left orbit and could represent lipoma versus hemangioma.

TREATMENT

The patient received intravenous (IV) Methylprednisolone 60mg once daily, Ketorolac IV 30mg three times a day and Esomeprazole (Nexium) IV 40mg once daily during the hospital stay. She admitted in hospital for 11 days and responded well to IV medications and was vitally stable and medically fit at the time of discharge. She was also advised to seek plastic and neurosurgery services for the treatment and further evaluation of the underlying lipoma or hemangioma.

DISCUSSION

Bell's palsy's incidence is around 11-40 cases per hundred thousand persons. It can occur at any age, but the peak incidence has usually been reported around the age of 30 to 45 years (De *et al.*, 2005; Zhao *et al.*, 2017). The reported lifetime risk of Bell's palsy is 1 in 60 individuals (De *et al.*, 2005). It occurs equally across both genders, though pregnant women and the elderly are reported to have a higher incidence (Holland and Weiner, 2004). Recent studies of Bell's palsy showed that most of the recurrence cases were reported in females between the age of 17 and 30 years, and predominantly involved the right side (Cirpaci and Goanta, 2014). Common problems associated with it were

Table 1: Blood Investigation results of Recurrent Bell's Palsy Case

Test	Results
Complete Blood Count (CBC)	
Hemoglobin (Hb)	13.2 g/dL
Platelets	345 x 10 ⁹ /L
White Blood Cells (WBCs)	8.6 x 10 ⁹ /L
Renal Function Test	
Urea	48 mg/dL
Creatinine	3.1 mg/dL
Sodium	136 mEq/L
Potassium	3.4 mEq/L
Erythrocyte Sedimentation Rate (ESR)	7 mm/hr
Vitamin D	19 ng/mL

ipsilateral ear complains and facial paresthesia (Circiaciu and Goanta, 2014). It can be congenital or acquired (Spencer and Irving, 2016). Acquired conditions are caused by a wide range of risk factors like infections (mostly caused by herpes virus) or otitis media, trauma (surgical or after head injury), vascular ischemia, neurologic (multiple sclerosis, Guillain-Barré syndrome), autoimmune diseases, immunologic or inflammatory disorders and sometime idiopathic Bell's palsy (Stjernquist-Desatnik *et al.*, 2006; Zhao *et al.*, 2017). Some studies also suggest migraine as an associated risk factor for Bell's palsy (Kim *et al.*, 2019; Peng *et al.*, 2015). Additionally, diabetes mellitus and some psychological disorders are also common comorbidities (Tseng *et al.*, 2017). Some studies, although controversial, noted seasonal and epidemiological clustering (Eviston *et al.*, 2015; Rowlands *et al.*, 2002). Likewise, the pathogenesis of the disease is still unidentified. Some suggested that inflammation and edema of facial nerve play a role which is also acceptable and the basis of decompression surgical procedure of nerve (Baugh *et al.*, 2013).

Facial asymmetry and weakness can be both peripheral and central and it will be differentiated by history and neurological examination of the patient. There is no definitive diagnostic marker for facial palsy, therefore clinical examination is vital. To evaluate the central cause, usually Magnetic Resonance Imaging (MRI) of the brain is done. Other tests include a cerebrospinal fluid examination and serological tests. In Bell's palsy, the cause of facial weakness is usually peripheral and in most cases no apparent cause will be found (Gilden, 2004). In this case, the cause is idiopathic, and the patient showed good prognosis after both the episodes.

The first line of treatment, recommended in most

of the countries, is the use of steroids whereas in some places antiviral is also used in combination with steroids (Almeida *et al.*, 2014; Baugh *et al.*, 2013). Some studies also reported that the use of a combination therapy is more effective as compared to steroids alone (Sullivan *et al.*, 2016; de Almeida *et al.*, 2009). In our reported case, the medical treatment (steroid) was given to the patient and the patient had mostly recovered by the time of discharge. Literature showed no benefits after surgical decompression of facial nerve in the prevention of recurrence (Swami *et al.*, 2010). Timing of surgery, route and extent of decompression all are still debatable. Nevertheless, (Dutt *et al.*, 2000) have reported recurrence of facial palsy after decompression surgery (Dutt *et al.*, 2000).

Almost 70% of the patients have a good prognosis if they receive treatment within the first 72 hours of onset (Holland and Weiner, 2004). However, untreated and 30% of patients who have received an early treatment may progress towards a poor recovery, continuing facial asymmetry, psychological distress and facial pain (Peitersen, 2002; Neely *et al.*, 1996). Factors involving poor prognosis are old age (De *et al.*, 2005), combined disease like hypertensive or diabetes (Fujiwara *et al.*, 2014), impaired taste (Diamant *et al.*, 1972), pain beside earache and weakness of whole face (Adour and Wingerd, 1974). Approximately 2/3 of the Bell's palsy patients recover completely, whereas prognosis is lower in cases of recurrence as compared to primary Bell's palsy (Dong *et al.*, 2019). However, there is a lot of disparity reported in the prognosis of recurrent Bell's palsy. Some studies noted that as compared to non-recurrent palsies, unilateral recurrent palsies showed poor prognosis (Ralli and Magliulo, 1988). Meanwhile, in another study,

the researchers did not observe any poor prognosis regardless of the affected side (Pitts *et al.*, 1988). It was also reported that the probability of recurrence is directly related to the age of the patient; younger the patient higher the chances of recurrences. Similarly, probability also increases with a total occurrence count (Swami *et al.*, 2010).

CONCLUSIONS

Recent studies of Bell's palsy showed that most of the recurrence cases were reported in young females, predominantly involved the right side while common problems associated with it were ipsilateral ear complains and facial paresthesia. There is a high chance of relapse during the early two years from the initial episode. As in the case of our patient, she is young and had history of left side cosmetic surgery and came with recurrent Bell's palsy. Therefore, regular follow-ups of younger patients and children after every year from the first episode are highly suggested. This might reduce the recurrent episode and will improve the prognosis. There is also a need for further detailed and longitudinal controlled studies to find out the epidemiology of recurrent Bell's palsy.

ACKNOWLEDGMENT

This research was funded by the Deanship of Scientific Research at Princess Nourah bint Abdulrahman University through the Fast-track Research Funding Program.

Declaration of Conflict interests

The authors declare that there is no conflict of interests.

Patient Consent

An informed consent has been taken to publish this case study.

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