

to hemorrhage due to the size and vascularity of the fetal adrenal gland. (4)

Congenital neuroblastoma is an important differential diagnosis. There are no pathognomonic sonographic features of neonatal hemorrhage but serial ultrasound examinations can help to differentiate between these two conditions. A hematoma decreases in size within one to two weeks whereas neuroblastoma tends to retain its echogenicity and does not change in size. Urinary catecholamines are elevated in the majority of cases of neuroblastoma. Colour coded Doppler sonography is useful to differentiate between congenital neuroblastoma and adrenal haemorrhage. In neuroblastoma it shows a network of microscopic vessels that invade the tumour and provide blood supply essential for its growth. This network gives rise to characteristic high velocity doppler shifts. In contrast to neuroblastoma, adrenal haemorrhage is characterized by diminished or absent blood flow. (3) Investigations include imaging in form of ultrasound, CT scan and MRI. MRI can determine the age of the hematoma. (2)

There are no reports in literature whether adrenal insufficiency is transient or chronic following neonatal adrenal hemorrhage. In our patient it was transient

and recovery occurred within 4 months. A short ACTH stimulation test has to be done at regular intervals for monitoring adrenal recovery.

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## CASE REPORT

### MARGINAL MANDIBULAR NERVE PALSY AS AN UNUSUAL COMPLICATION IN LUDWIG'S ANGINA

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#### Abstract

Ludwig's angina is a rapidly spreading cellulitis of the floor of the mouth characterized by firm induration and elevation of the tongue. The cellulitis may spread and cause life-threatening complication by obstructing the airway. Several other complications have been reported secondary to the progress of cellulitis in different anatomical plains. We describe a case of a two year old girl with Ludwig's angina who developed left sided marginal mandibular nerve palsy. This complication has never been reported before.

**Keywords:** Marginal mandibular nerve palsy, Ludwig's angina, complication

#### Introduction

Ludwig's angina is a potentially life-threatening infection of submandibular space characterized by hardness of floor of mouth (1). It presents most often with fever, neck mass or swelling and has a varying degree of systemic toxic symptoms. The spreading cellulitis may cause complications like life threatening obstruction of airway. Cellulitis may progress in different anatomical spaces extending to the lateral pharyngeal space, retropharyngeal space and may even descend to the mediastinum. We present here a case report of a two year old child with Ludwig's angina who had an

unusual complication of paresis of marginal mandibular branch of facial nerve.

#### Case Report

A two year old female child presented with fever for last 4 days along with swelling of the lower part of the face which was progressively increasing in size. She had mild cough and occasional vomiting. She was not taking any oral feeds and was quite irritable. She was the only child of her parents. She was born by normal delivery and her immediate neonatal period was uneventful. She was vaccinated with BCG, OPV, DPT and MMR. Her development was normal and she had not had any significant illness before. On examination she was irritable, febrile and had pallor. She had a brawny, indurated and markedly tender significant swelling in the submandibular region extending both sides up to the parotid region. The floor of the mouth was raised. There was no pharyngeal or tonsillar inflammation. There were multiple enlarged lymph nodes in the neck region. She also had palsy of the left lower part of the face as her angle of mouth was getting deviated to the right side while crying indicating involvement of left marginal mandibular nerve, a branch of cervicofacial division of facial nerve (Figure 1). A clinical diagnosis of Ludwig's Angina was made.

**Figure 1: Illustration showing swelling in the submandibular region with deviation of angle of mouth towards right side.**



On complete blood examination, hemoglobin was 9.2 g/dl and total WBC count was elevated to 13600/cumm with predominant leucocytosis. CRP was positive and her random blood sugar was normal. An ultrasound of neck showed soft tissue swelling of the neck region with diffuse enlargement of submental lymph nodes along with diffuse bulkiness of parotid glands. No necrotic focus was seen. She was treated with IV Ampicillin, Cloxacillin, Amikacin, dexamethasone, maintenance IV fluids, oral anti-inflammatory and analgesics. Over next 2 days, the fever was less but the swelling, tenderness and the left lower facial paresis persisted. She was unable to eat or drink and continued to be on IV fluids along with IV antibiotics and analgesic. After two days she was operated by maxillofacial surgeons and 25cc pus was drained from bilateral submandibular space and submental space and through & through communication between spaces was achieved by dissection. Corrugated rubber drains were placed and secured in place with the help of silk suture to achieve continuous drainage. Irrigation was done every day and dressing was placed. The pus culture did not show any growth. The child became afebrile within next 24 hours and the paresis of left lower part of the face improved. She had a gradual recovery over next 3-4 days and she was discharged home after 5 days with oral antibiotics. The paresis resolved completely before discharge.

### Discussion

Ludwig's angina is a rapidly progressing cellulitis involving the submandibular neck space. It is characterized by brawny induration of the submental region and floor of mouth, bearing the potential for rapid airway obstruction. It was formally described by von Ludwig in 1836, and criteria for diagnosis were developed by Grodinsky in 1939 (2). Patients with Ludwig's angina classically present with progressive bilateral submandibular and submental neck swelling, firm induration of the floor of the mouth, and edematous posterior and superior displacement of the tongue. The systemic symptoms generally are fever, chills, malaise, dehydration from decreased oral intake, and a generalized toxic ill-appearance. Other symptoms may include tongue and throat pain, dysphagia, trismus,

dysphonia, and drooling (3) Infants less than 9 months old most often present with a neck mass or swelling, lymphadenopathy, fever, rhinorrhea, poor oral intake, and cough(4).

The cellulitis rapidly involves the sublingual space with subsequent expansion of the floor of the mouth. This results in displacement of both the oral tongue superiorly and the base of the tongue posteriorly, producing a potentially life-threatening obstruction of the airway at the level of the oral cavity and oropharynx (3). Various complications may arise secondary to progress of the cellulitis in different anatomical plains. Those include mediastinitis and mediastinal abscess formation, subphrenic abscess formation (5), pneumothorax(6), pericardial or pleural effusion, necrotizing fasciitis, jugular venous thrombosis, rupture of the inominate artery (7), empyema (8), and mandibular or cervical osteomyelitis (9). Besides these complications, a generalized sepsis syndrome (10, 11) or aspiration pneumonia (7) also have been reported. But unilateral palsy of any branch of facial nerve secondary to Ludwig's angina has never been reported before in literature. This complication can also be explained on anatomical basis (1,12). Kissig et al (2006) described clinical findings from 35 cases of paralysis of distinct branches of facial nerve and out of those marginal branch of mandibular nerve was involved in 21 cases. The various causes described were iatrogenic, tumour, trauma, congenital, idiopathic and inflammation (13). In this particular case the complication could have arisen due to involvement of the marginal mandibular nerve secondary to the inflammatory process. This particular complication of paresis of marginal mandibular branch of facial nerve secondary to Ludwig's angina has never been reported before.

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## CASE REPORT

### PROTEUS SYNDROME

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#### Abstract

Proteus syndrome is a rare hamartomatous condition with multisystem involvement and great clinical variability. A 7yr old girl presented with recurrent falls with progressive maculopapular lesions over face, trunk and limbs, asymmetry of face and calf, scoliosis, lower limb discrepancy and lipoma over lower back suggestive of Proteus syndrome. She is planned for lipoma excision and laser treatment.

#### Introduction

Proteus syndrome is a rare hamartomatous condition with multisystem involvement and great clinical variability with prevalence of less than 1 per 1,000,000 live births. (1) First reported in 1979 and named Proteus syndrome after Greek god Proteus who could change his shape at will to avoid capture. (2)

#### Case Report

A term appropriate for gestational age female baby weighing 3.2 kgs was born to a booked immunized multigravida by normal vaginal delivery. The mother and father were non consanguineous and were aged 23 and 25 years respectively. The baby was detected to have a lump on right lower back with an approximate size 2x2cm with no associated tuft of hair or watery discharge. Mother also noticed there was facial asymmetry with a deviation of angle of mouth to left side with no impaired closure of eyes or drooling of saliva from angles of mouth. There were multiple hyperpigmented lesions over face, trunk and upper limbs. On D2 of life baby developed neonatal jaundice and was managed by double surface phototherapy for 24 hrs. Developmentally child had achieved all milestones in time.

Now at the age of 7yrs the child is brought to our hospital with history of recurrent falls while

running and progressive nature of cutaneous lesions. The examination revealed height, weight and head circumference appropriate for age and sex. The cardiopulmonary status was stable. There were multiple hyperpigmented maculopapular lesions of various sizes over face, trunk and limbs which increase in size on crying (picture1). There was asymmetry of face with deviation of angle of mouth to left. There was lipoma on thoracolumbar region measuring 12cm x 10cm .There was asymmetry of calf muscles (left>right), lower limb length discrepancy (left>right by 3.5 cm) and macrodactaly. There was scoliosis to left; however other neurological examination was essentially normal.

X-ray spine revealed scoliosis to left and soft tissue swelling at thoracolumbar region, CECT (chest) was normal; MRI (brain) did not reveal any abnormality. Presently child is under follow up in orthopedic and skin OPD, LASER treatment is planned for cutaneous lesion later on. The lipoma on back is planned for excision.

**Picture 1: Multiple maculopapular lesion over trunk and limbs**

