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Conditions with Unilateral Presentation - A Diagnostic Challenge

Dr. Gayathri. V. S, Dr. Saicharan, Dr. Kumari

Ragas Dental College and Hospital, Uthandi, Tamil Nadu, India

ABSTRACT

An appropriate diagnosis of any head and neck condition itself is a challenge in the field of dentistry and when it comes to diagnosing conditions with a unilateral presentation, it becomes a challenge for the diagnostician. Proper knowledge of the head and neck anatomy and the trigeminal system is essential for categorizing unilateral conditions during the formulation of a differential diagnosis. Differential diagnosis of such conditions helps in treatment planning accordingly. This review intends to categorize conditions with a unilateral presentation along with their features for appropriate diagnosis of the same.

KEYWORDS: *Unilateral, head and neck, orofacial, neuralgia, pain, syndrome*

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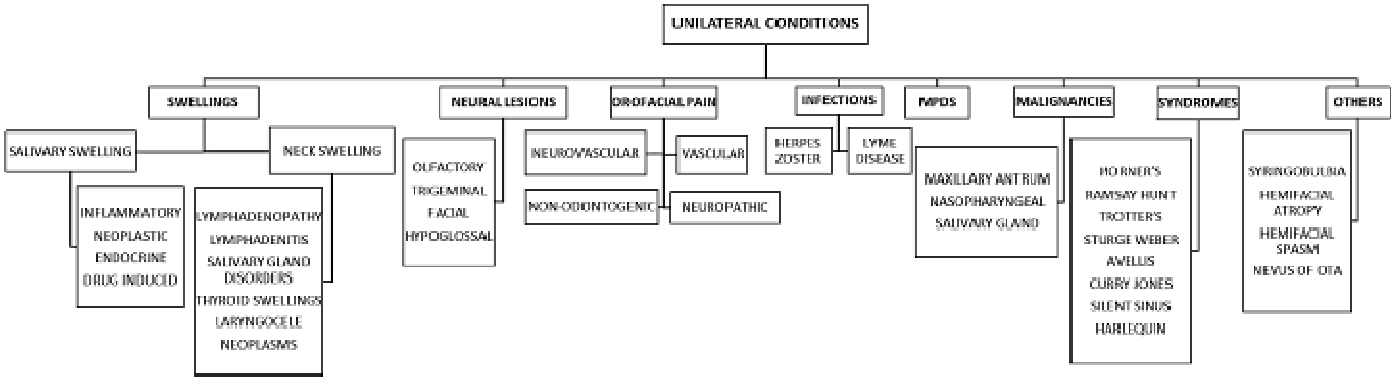
INTRODUCTION

Unilateral conditions are those affecting only one side of the head and neck in particular. It can be swelling, a lesion, an infection, a syndrome, or unilateral pain of the head and neck. The reason for their unilateral presentation, differential diagnosis, and appropriate treatment plan claims knowledge of head and neck anatomy and their neurovascular distributions. This review was performed after gathering a handful of information from the literature and then we were able to sort down the conditions and classify them.

CLASSIFICATION OF CONDITIONS WITH UNILATERAL PRESENTATION:



1. SALIVARY SWELLINGS

Unilateral swelling in the region of the salivary glands may arise from the gland itself or in an intraglandular lymph node, or other tissues. The main causes of salivary gland swellings are obstruction, infection, inflammation, or neoplasm. Ultrasonography, other imaging, and biopsy are often indicated (1).

2. NECK SWELLINGS

Discrete **unilateral swellings** in the neck are often due to lymph node enlargement but may occasionally be caused by disorders in the salivary glands, the thyroid gland & other structures. The location of a lump or swelling in the neck will often give a good indication of the tissue of origin. The age of the patient may suggest the most likely diagnosis. The

duration of the lesion is also relevant, for example, one that has been present since an early age is likely to be of congenital origin, while a lump appearing in later life and persisting may be due to malignancy(2).

3. CRANIAL NERVE LESIONS

OLFACTORY NERVE:

Unilateral anosmia is often unnoticed but causes a loss of ability to smell odors; in practice, the patient may complain of loss of taste rather than of smell. Anosmia is common after head injuries, as the nerves may be torn as they pass through the cribriform plate; this is experienced especially in Le Fort III fractures of the middle third of the facial skeleton. Olfactory neuroblastoma is a rare tumor linked tentatively to dental staff. An olfactory lesion is confirmed by the inability to smell substances, such as orange, soap, coffee, cloves, or peppermint oil(3).

TRIGEMINAL NERVE:

Trigeminal motor neuropathy may be possibly related to a viral infection. Trigeminal motor neuropathy is frequently associated with trigeminal sensory neuropathy or due to lesions affecting the motor division of the trigeminal nerve when there are usually other cranial nerve deficits, sometimes caused by an intracranial tumor. Weakness may develop, sometimes with masseter and temporalis wasting. Damage to the motor part of the trigeminal can be difficult to establish; it is usually asymptomatic if unilateral, but the jaw may deviate towards the affected side on opening. It is easier to detect motor weakness by asking the patient to open the jaw against resistance, rather than by trying to test the strength of closure(4).

FACIAL NERVE:

The motor neurons supplying the lower face receive upper motor neurons (UMNs) from the contralateral motor cortex, whereas the neurons to the upper face receive bilateral UMN innervation. Thus, a UMN lesion causes unilateral facial palsy with some sparing of the frontalis and orbicularis oculi muscles. In contrast, lower motor neuron (LMN) facial palsy is characterized by **unilateral paralysis** of all muscles of facial expression – for both voluntary and emotional responses(4).

BELL'S PALSY:

Bell's palsy is **unilateral** facial paralysis for which no local or systemic cause can be identified, though it is now recognized to be a lower motor neuron palsy and mainly due to infection with the herpes simplex virus. There appears to be inflammation of the facial nerve in the stylomastoid canal, sometimes with demyelination(5).

ASSOCIATIONS: Hypertension, lymphoma, Melkersson–Rosenthal syndrome, Crohn's disease, orofacial granulomatosis, sarcoidosis, connective tissue disorders, acoustic neuroma, Guillain–Barré syndrome, multiple sclerosis, and a variety of other disorders.

HYPOGLOSSAL NERVE:

LMN lesions of the twelfth cranial nerve lead to **unilateral tongue weakness**, wasting, and fasciculation. When protruded, the tongue deviates towards the weaker side. The cause may be within the brainstem, at the skull base, or within the neck(6).

4. NEUROVASCULAR OROFACIAL PAINS

Neurovascular pains are a group of visceral pain disorders that are generally characterized by episodic pains accompanied by neurologic, gastrointestinal, and psychological changes. These pains are commonly **unilateral** with varying duration and can greatly debilitate the patient(7).

1. MIGRAINE
2. TRIGEMINAL AUTONOMIC CEPHALGIA
   - CLUSTER HEADACHE
   - PAROXYSMAL HEMICRANIA
   - SHORT-LASTING UNILATERAL NEURALGIFORM HEADACHE ATTACK (SUNCT)
   - HEMICRANIA CONTINUA

1. MIGRAINE:

Migraine is a recurrent **Unilateral headache** combined with autonomic disturbances, affecting over 14% of women and 7% of men.

PRODROME: Diminished cerebral blood flow, possibly the result of a neuronal trigger mechanism in the trigeminovascular nucleus, but the headache itself is usually associated with increased cerebral and extracranial blood flow. This causes the trigeminal nerve to release neuropeptides; the combination of serotonin and bradykinin may cause blood vessels to become dilated and inflamed. Migraines can be with or without aura.

CLINICAL FEATURES OF CLASSIC MIGRAINE:

Unilateral headache preceded by an aura experienced 10–30min before the headache. Most auras are visual (bright shimmering lights around objects or at the edges of the field of vision or zigzag lines, castles, wavy images, or hallucinations). Other patients have temporary vision loss.

Non-visual auras include motor weakness, speech or language abnormalities, dizziness, vertigo, and tingling or numbness of the face, tongue, or extremities(8).

2. TRIGEMINAL AUTONOMIC CEPHALGIA:

Trigeminal autonomic cephalalgias are a group of headache disorders that are characterized by not only a headache but also prominent cranial parasympathetic autonomic features(9).

A. CLUSTER HEADACHE

Paroxysms of stabbing, ice prick–like pains in the periorbital region that last for a few seconds and may occur once or several times in rapid succession. The pain often commences quickly, without warning, and reaches a crescendo within 2 to 15 minutes. The pain usually begins in, around, or above one eye or the temple. It is **always unilateral** and generally affects the same side in every episode, with a predominance on the right side. The onset of severe pain at night (classically at around 2.00 a.m.)(9)

ACCOMPANYING AUTONOMIC SYMPTOMS:

Flushing and/or sweating unilaterally on the affected side of the face, Lacrimation, Conjunctival injection, Nasal congestion, sometimes with Horner syndrome.

B. PAROXYSMAL HEMICRANIA

Characterized by frequent short-lasting attacks of severe **unilateral pain**. In orbital, supraorbital or temporal regions Paroxysmal hemicrania typically last minutes only. The attack frequency usually ranges from 5–40 attacks per day. Associated with autonomic symptoms(10).

C. SHORT LASTING UNILATERAL NEURALGIFORM HEADACHE ATTACK

Mainly seen in males above 50 years, short-lasting unilateral attacks of intermittent, moderate to severe burning, piercing or throbbing daytime pain, **strictly unilateral**. Pain is around the eye & temple lasting 5 seconds to 5 minutes per episode. Systolic blood pressure may rise during the attacks. Movement of the neck may trigger these headaches. Associated with autonomic symptoms(11).

D. HEMICRANIA CONTINUA

Characterized by a continuous, **unilateral headache** that varies in intensity. Waxing and waning without disappearing completely. More severe than the inter-paroxysmal pain of the other TACs. The exacerbations in hemicrania continua are longer than the paroxysms of the other TACs. Associated with autonomic symptoms(12).

5. VASCULAR OROFACIAL PAINS

Vascular pains are those pains that originate from the tissues that make up the vessel walls. These pain conditions are far less common than neurovascular pains. There are many different types of vascular conditions that can produce head and neck pain. Some of these conditions are arterial hypertension, cerebral venous thrombosis, ischemic stroke, vascular malformation, and intracranial hematoma. But only, two conditions produce pain **unilaterally** in the orofacial region. They are:

1. CRANIAL ARTERITIS
2. CAROTIDYNIA

CRANIAL ARTERITIS:

The term cranial arteritis suggests that an artery in the cranial region has become inflamed. The most common artery to be involved in the temporal artery; inflammation of this artery is therefore referred to as temporal arteritis. Temporal arteritis commonly presents as a severe headache in the **unilateral temporal region.** Pain is often throbbing, boring, burning pain with a superimposed lancinating quality. Palpation of the temporal region will usually find a prominent, tortuous, very tender, and enlarged temporal artery(13).

CAROTIDYNIA:

Carotidynia is a painful condition that arises from the cervical carotid artery. **Unilateral neck pain** frequently radiates to the ipsilateral face and ear and sometimes to the head. The presence of facial pain, carotid artery tenderness, and overlying soft tissue swelling are classic features. If the thumbs are placed on the CCA just below the bifurcation and the structures are pressed back against the transverse cervical processes with a rolling movement, severe pain is produced. Pain is referred to the eye, deep in the malar region, and spreads back to the ear(14).

6. NON-ODONTOGENIC TOOTHACHES

Toothaches that do not have their origin in the dental structures are called non-odontogenic toothaches. Neurovascular pain conditions are unique pain disorders only felt in the craniofacial structures. The intracranial vessels of the brain are primarily innervated by the trigeminal nerve and thus are called the trigeminovascular system. Migraine and trigeminal autonomic cephalgia arise from trigeminovascular systems which produce pain in teeth(8).

7. NEUROPATHIC PAINS

Bureau et al provided evidence that six sensory adjectives were significantly more frequently chosen by patients with neuropathic pain. The six adjectives were **Electric shock, Burning, Cold, Pricking, Tingling,** and **Itching(15)**.

1. EPISODIC NEUROPATHIC PAIN can be either neurovascular or neuralgic.
2. CONTINUOUS NEUROPATHIC PAIN can be peripherally mediated neuropathy, centrally mediated neuropathy, or metabolic polyneuropathy.

EPISODIC NEUROPATHIC PAIN:

Episodic neuropathic pain is associated with Neuralgia. Neuralgic pain is characterized by sudden volleys of paroxysmal electric-like pain. This paroxysmal pain is quite characteristic of neuralgias, assisting greatly in diagnosis(16).

- Trigeminal Neuralgia
- Glossopharyngeal Neuralgia
- Postherpetic Neuralgia
- Superior laryngeal Neuralgia
- Raeder paratrigeminal Neuralgia

TRIGEMINAL NEURALGIA:

Pain is confined to the trigeminal area of **one side**, usually the maxillary or mandibular division, or occasionally both. Infraorbital or lower lip/lower jaw pain is common, and is severe and sharply stabbing, of only a few seconds duration, but paroxysms may follow in quick succession. A patient seen crying with pain during an attack is not easily forgotten. Trigger zones are usually within the trigeminal area. The triggers can be shaving, stroking or touching the face, eating, drinking a hot or cold liquid, brushing teeth, talking, putting on cosmetics, encountering a cold breeze, or walking into an air-conditioned room(17).

GLOSSOPHARYNGEAL NEURALGIA:

Glossopharyngeal neuralgia is rare and frequently idiopathic, possibly caused by an abnormal intracranial blood vessel, as in trigeminal neuralgia. The pain is like that of TN but affects the unilateral throat, especially the tonsillar region. The triggers are swallowing and coughing. Pain may be felt within the ipsilateral ear and simulate neuralgia of the nervus intermedius. Sometimes, along with lesions of the vagus and accessory nerves. Hoarseness, dysphagia, palatal deviation to the intact side, anesthesia of the posterior pharyngeal wall, and weakness of the sternomastoid and upper trapezius are seen(18).

POSTHERPETIC NEURALGIA:

In some instances, acute herpes zoster persists, which ends in a condition referred to as postherpetic neuralgia. If Pain persists over 3 months after the rash has healed, may be a debilitating and difficult-to-manage consequence of Herpes Zoster. Repeated exposure to the virus, stress, fatigue, immunosuppression and Hodgkin's disease can be predisposing factors. An intractable, chronic, burning pain is felt superficially within the area littered with the acute attack. If it lasts longer than a year, it's likely to persist indefinitely and be proof against all therapies. Other symptoms include dysesthesia, hypoesthesia, and hyperesthesia within the cutaneous distribution of the affected nerve(19).

SUPERIOR LARYNGEAL NEURALGIA:

The superior laryngeal nerve is a branch of the vagus and innervates the cricothyroid muscle of the larynx, which stretches, tenses, and adducts the plica vocalis. The Pain of superior laryngeal neuralgia is periodic, **unilateral submandibular** pain radiating through the ear, eye, or shoulder, lasting. It is sometimes difficult to differentiate this condition from glossopharyngeal neuralgia. The patient may report an irresistible urge to swallow. Swallowing, straining the voice, turning the head, coughing, sneezing, yawning, or blowing the nose can trigger pain. The trigger point is found just superior and lateral to the thyroid cartilage(20).

RAEDER PARATRIGEMINAL NEURALGIA

Raeder paratrigeminal syndrome is characterized by severe, **unilateral facial pain** and **headache** within the distribution of the ophthalmic division of the fifth cranial nerve. Combination with ipsilateral oculosympathetic palsy or Horner syndrome. Head trauma, hypertension, vasculitis, migraine headaches, para sellar mass lesions, and internal carotid artery dissections(21).

8. INFECTIONS

HERPES ZOSTER/SHINGLES/ZONA:

An acute infectious viral disease of extremely painful and incapacitating nature, characterized by inflammation of dorsal root ganglion, related to vesicular eruptions of skin and mucosa of the area supplied by the affected nerve. Trauma, malignancy, radiation, and immunosuppressive therapy are a few predisposing factors. Zoster of any division of the fifth cranial nerve may cause a facial rash, pain, and oral ulceration which is **unilateral and doesn't cross the midline**. Usually preceded by aching, burning, stabbing, or shock-like pain of varying severities. The rash is initially erythematous and maculopapular but progresses to coalescing clusters of clear vesicles over several days and later evolves through pustular, ulcer, and crust stages over 7–10 days, with complete healing within 2–4 weeks(22).

LYME DISEASE:

Lyme disease is caused by the Borrelia burgdorferi, transmitted mainly by insects. The chance of Lyme disease is a principle in people who live or work near areas surrounded by tick-infested woods or overgrown brush. Red 'bull's-eye' rash that spreads outwards from the location of the hurt. Tiredness, fever, headache, stiff neck, and muscle aches are common. Arthritis may develop within the acute phase and be transient, or later persistent, most typically affecting the knees severely. Neurological involvement ends up in **unilateral facial palsy(23).**

9. MALIGNANT NEOPLASMS

MAXILLARY ANTRAL CARCINOMA:

Antral carcinoma could be a rare neoplasm of unknown etiology, seen mainly in older people. It presents with severe maxillary pain, intraoral alveolar swelling; ulceration of the palate or buccal sulcus; swelling of the cheek; **unilateral nasal obstruction,** often related to a blood-stained discharge; obstruction of the nasolacrimal duct with consequent epiphora; hypoesthesia or **unilateral anesthesia of the cheek** within the infraorbital nerve distribution; proptosis and ophthalmoplegia(24).

NASOPHARYNGEAL CARCINOMA:

Nasopharyngeal carcinoma, being a rare neoplasm, may be associated with Epstein–Barr virus and dietary nitrosamines. Often remains asymptomatic for some time, because it rarely obstructs the nasopharynx. **Unilateral** cervical lymphadenopathy, **unilateral** conductive deafness & blocked nose, sixth cranial nerve palsy, palate palsy and immobility, and ipsilateral pain, sometimes with anesthesia(25).

SALIVARY NEOPLASM:

Neoplasms within the major salivary glands are most commonly pleomorphic adenomas, monomorphic adenomas, mucoepidermoid tumors, or acinic cell tumors. Neoplasms within the minor salivary glands are most commonly pleomorphic adenomas but carcinomas, particularly adenoid cystic carcinomas, account for about 50%.

BENIGN – Long history of **unilateral** salivary gland swelling

MALIGNANT – **Unilateral** Pain, Facial palsy, rapid growth, change in growth pattern(26).

10. SYNDROMES

HORNER SYNDROME:

Caused by interruption of sympathetic nerve fibers peripherally. Maybe due to trauma to the neck, or lung carcinoma infiltrating the superior cervical sympathetic ganglion. Unilateral presentation of symptoms like miosis, ptosis, loss of sweating of the ipsilateral face, and enophthalmos(27).

RAMSAY HUNT SYNDROME:

Ramsay Hunt syndrome is a herpes zoster of the nervus intermedius, the sensory component of the facial nerve. **Unilateral** neuritic pain, ipsilateral facial paralysis, superficial herpetic lesions in the external ear, auditory canal, mastoid area, and even on the tympanic membrane. Intraorally, the heterotopic pain, and herpetic lesions affect the fauces, palate, and anterior tongue(28).

TROTTER SYNDROME:

Occurs because of the invasion of the nasopharynx and trigeminal nerve by a malignant tumor. **Unilateral deafness,** pain in the mandibular division of the nerve, ipsilateral palatal immobility, and trismus are classic features(29).

STURGE-WEBER SYNDROME:

A congenital hamartomatous angioma of the unilateral upper face, oral mucosa, and underlying bone. Ipsilateral glaucoma was noted in 1/3rd of cases. Extending intracranially causes convulsions, contralateral hemiplegia, intracerebral calcifications, and sometimes learning disorders(29).

SILENT SINUS SYNDROME:

Silent sinus syndrome occurs due to hypoventilation of the maxillary sinus after osteomeatal complex obstruction. Clinical features are **Unilateral** enophthalmos, hypoglobus secondary to thinning of the maxillary sinus roof, and absence of sinonasal inflammatory disease(29).

CURRY JONES SYNDROME:

Caused due to mutations in genes in the Sonic Hedgehog pathway. Clinical features include **Unilateral** coronal synostosis, Plagiocephaly Microphthalmia Craniofacial asymmetry Iris coloboma Broad thumbs Hand syndactyly, foot polydactyly Skin lesions Gastrointestinal abnormalities(30).

HARLEQUIN SYNDROME:

Harlequin syndrome is caused by a **unilateral blockade** of the sympathetic innervation of the face which results in an inability of the facial vasculature to dilate in response to normal stimuli. Unilateral facial flushing, unilateral facial sweating, and contralateral anhidrosis are classic features. These features are induced by Exercise, Heat & Emotions(31).

11. TEMPOROMANDIBULAR JOINT PAIN DYSFUNCTION SYNDROME

Triad of **unilateral** joint symptoms:

1. Recurrent clicking in the TMJ
2. Jaw locking or limitation of movement
3. Pain in the joint and/or surrounding muscles and elsewhere ipsilaterally.

12. OTHERS

SYRINGOBULBIA:

Syringomyelia is cavitation of the spinal cord, of unknown etiology, disrupting pain and temperature neurons of the anterior commissure. Syringobulbia is the term used if it affects the brainstem. Presents with facial or oral sensory changes or paralyzes, unilateral palatal and vocal cord palsies, nystagmus, weakness and atrophy of the tongue, dysphagia, and dysarthria(32).

HEMIFACIAL SPASM:

Hemifacial spasm is a neuromuscular disorder characterized by frequent involuntary contractions of the muscles on one side of the face. Unilateral spasm of the angle of the mouth or the eyelid, worse towards evening. Usually idiopathic, it may be caused by a vascular anomaly affecting the vertebrobasilar vessels causing pressure on the facial nerve(33).

HEMIFACIAL ATROPHY:

A rare form of lipodystrophy is the facial hemiatrophy of Romberg, starting typically in adolescence and females. Progressive unilateral disappearance of facial fat, mimicking facial paralysis. Patients appear to be otherwise well. Plastic surgery is the only treatment(34).

NEVUS OF OTA:

The unilateral proliferation of dermal melanocytes in the distribution of the first and second branches of the trigeminal nerve. The lesion is most common in Japanese people, and approximately 80% of affected individuals are female. In about half of the cases, the lesions are present at birth. The lesions tend to be macular; to be brown, bluish, or slate to black; and have ill-defined margins. The Ocular and Oral mucosa, Auditory canal, Tympanic membrane, and Skin may be involved(35).

CONCLUSION:

Diagnosing & managing unilateral conditions can be challenging and the clinician should be aware of different etiologies & characteristics of the conditions occurring unilaterally in the head & neck region. Understanding the neurobiology of the Trigeminal system is the key to better diagnosis & management.

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