

# Frey Syndrome—An Underreported Complication to Closed Treatment of Mandibular Condyle Fracture? Case Report and Literature Review

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Frey syndrome (auriculotemporal syndrome, gustatory sweating) is characterized by episodes of warmth, flushing, and sweating of the face in the preauricular region initiated by gustatory stimulus.<sup>1</sup> Frey syndrome is a common complication after operations on the parotid gland and the temporomandibular joint.<sup>2,3</sup> The most common hypothesis is that regenerating parasympathetic fibers to salivary glands connect in error with the sweat glands and subcutaneous blood vessels of the skin. The onset has usually been 12 to 18 months after surgery. The most effective treatment has been subcutaneous infiltration of botulinum toxin into the affected area.<sup>4</sup>

Fracture of the mandible is a common injury. The mandibular condyle will sustain a fracture in 10% to 40% of all mandibular fractures, and 19% of condylar fractures will be dislocated.<sup>5,6</sup> The treatment of condylar fractures is still under evaluation, and closed treatment with maxillomandibular fixation has frequently been selected.<sup>7</sup>

Few reports exist on the development of Frey syndrome after acute mechanical trauma to the jaws, and

the frequency after closed treatment of mandibular condyle fracture is unknown.

## Case Report

A 23-year-old woman was referred to the Aarhus University School of Dentistry for an insurance examination after a bicycle accident 18 months earlier. She had had fractures of several teeth, the mandibular body between the lower right lateral and central incisor, the right coronoid process, and both condyles. The right condylar head had been displaced medially (Fig 1A,B). The patient had undergone open reduction and internal fixation of the symphysis and closed treatment with intermaxillary fixation of the coronoid and condylar fractures. She complained of hypoesthesia in the preauricular region and the cheek on the right side and fatigue and pain of both temporomandibular joints. Clinical examination by a temporomandibular joint specialist revealed tender masseter and temporal muscles in both sides and a reduced mouth opening. These problems were all well-known complications to mandibular fractures. The patient had also during the previous couple of months experienced episodes of sweating, blushing, and warmth in the preauricular region and cheek on the right side while eating. At the first clinical examination, this problem was not elicited, because the patient did not connect it to the trauma. She coincidentally confronted one of us (T.W.K.; a medical student at the time) with the problem, and he suspected Frey syndrome. At the second clinical examination some months later, clinical images were taken before and after she had eaten a sandwich. These showed flushing and sweat production at the right side (data not shown). Next, the minor starch-iodine test<sup>8</sup> was performed, confirming sweating on the right side (Fig 2). Thermography was also performed, with the thermograms visualizing heat production. After the patient chewed the sandwich, the temperature increased on average 2.2°C (from 32.5°C to 34.7°C) on the right side and only 1.1°C (from 32.0°C to 33.1°C) on the left side (Fig 3). The patient was diagnosed with Frey syndrome as a complication of the medially dislocated mandibular condyle fracture managed by closed treatment. The patient was pleased by the explanation of her symptoms and did not want any treatment.

## Literature Review

One prospective study and 10 case reports describing the development of Frey syndrome after

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**FIGURE 1.** Three-dimensional CT scan reconstruction of *A*, right side and *B*, left side showing fractures of several teeth, mandibular body between the lower right lateral and central incisor, right coronoid process, left condyle without dislocation, and right condyle with medial dislocation.

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**FIGURE 2.** Clinical image showing minor starch-iodine test. Black dots indicate sweating.

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closed treatment of mandibular condyle fractures were found in the period from 1969 to 2010 using the search criteria in PubMed: “Frey syndrome” combined with “mandibular fracture” or “jaw trauma.” These reports were thoroughly studied.

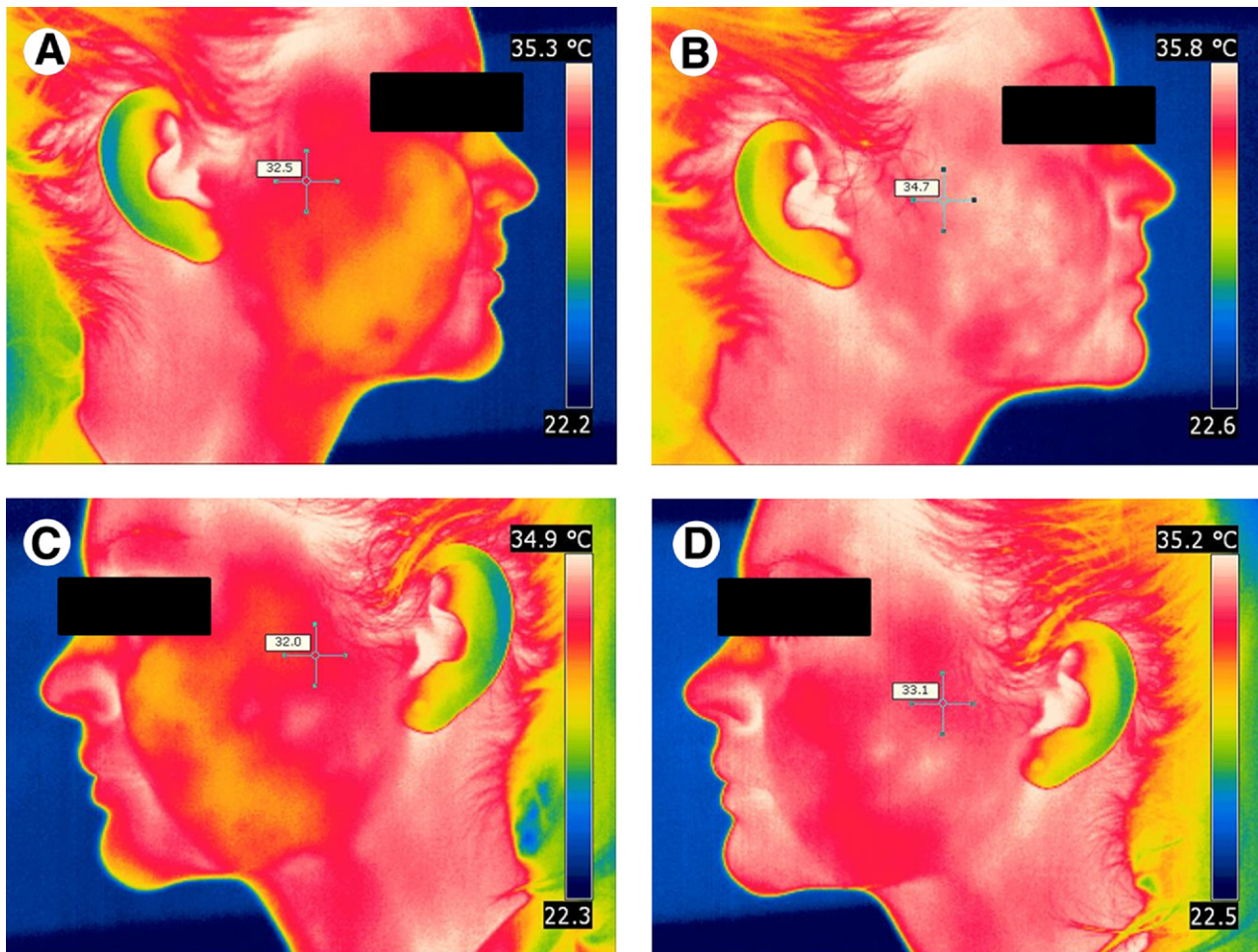
The single prospective study was published in 1977 by Schmidseider and Scheunemann,<sup>9</sup> who reported 2 cases of Frey syndrome (0.8%) in 237 fractures of the mandibular condyle managed by closed treatment from 1971 to 1975. All 10 case reports described patients with Frey syndrome that developed after closed treatment of mandibular condyle fractures.<sup>10-18</sup> Table 1 lists the patient characteristics from each case report. In 9 of the 10 cases, the fractures had resulted from traffic accidents or falls from above ground level. The mandibular condyle was dislocated in 6 cases, and 9 patients had more than 1 mandibular fracture site. In 5 of the 10 cases, the patient had had altered sensibility in the affected area. The latency period before the onset of symptoms ranged from 2 months to 2 years, and only 1 of the 10 patients had had spontaneous remission.

## Discussion

The incidence of Frey syndrome after closed treatment of a condylar fracture in the 1 prospective study was almost 1%.<sup>9</sup> Considering the relatively high frequency of mandibular condyle fractures, it is noteworthy that only 10 cases have been reported in the past 4 decades. This could suggest that the complication might be underreported. Just as occurred in the present patient, the explanations could be that the patient has not connected the symptoms to the trauma or that the symptoms or diagnosis were unknown to the clinicians examining the patient. Frey syndrome might be known to otolaryngologists; however, many patients will primarily be examined by physicians or oral surgeons and radiologists who might not be acquainted with the characteristic symptoms. Also, the long latency and the peculiar association between eating and the development of symptoms might make it difficult for patients to explain the symptoms. It is therefore essential that oral surgeons, temporomandibular joint specialists, and dental-maxillofacial radiologists consider Frey syndrome when examining patients with previous mandibular condyle fracture managed by closed treatment.

The anatomy and pathology of Frey syndrome after closed treatment of condylar fracture has been previously reviewed.<sup>1,17,19</sup> Frey syndrome develops because of nerve injury. The nerve in question is thought to be the auriculotemporal nerve, which consists of parasympathetic fibers to the parotid gland, sympathetic fibers to the sweat glands and subcutaneous vessels of the skin, and sensory fibers to the skin in the preauricular area. The auriculotemporal nerve is a branch of the mandibular division of the trigeminal nerve and passes just posterior to the mandibular condyle. Injury to the auriculotemporal nerve by a fractured and dislocated condyle is thought to be the mechanism leading to the autonomic neuropathies of Frey syndrome after closed treatment of condylar fractures. The gustatory sweating of Frey syndrome can be explained by 2 hypotheses. The most common hypothesis is that the regenerating parasympathetic fibers to salivary glands connect in error with the sweat glands. The other hypothesis is that the sweat glands develop an increased sensitivity after degeneration of sympathetic fibers and are then stimulated by acetylcholine released from adjacent parasympathetic fibers in the parotid gland. The gustatory vasodilation of Frey syndrome causing blushing and warmth is more difficult to elucidate. However, it has mostly been hypothesized to be caused by the misdirected regeneration of parasympathetic fibers to the subcutaneous blood vessels.





**FIGURE 3.** Thermograms showing mean temperature at affected region A, before and B, after patient chewed a sandwich and at the unaffected region C, before and D, after patient chewed a sandwich. White indicates areas with highest temperature.

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Under these circumstances, gustatory stimuli will lead, not only to stimulation of the parotid gland, but also to stimulation of the sweat glands and subcutaneous blood vessels. The symptoms of these autonomic neuropathies are episodes of warmth, flushing, and sweating of the face in the preauricular region initiated by gustatory stimulus. The changed sensibility associated with Frey syndrome can be explained by the injury to the auriculotemporal nerve or smaller nerves in the area.

In the present case, and in most of the previous case reports, Frey syndrome after closed treatment of a mandibular fracture developed after a traffic accident. Another characteristic was the presence of more than 1 fracture of the mandible. This could suggest that injury of the auriculotemporal nerve requires trauma with a considerable impact. In more than one half of the cases, the mandibular condyle was dislocated. Dislocation of the condyle could also be a risk factor for developing Frey syndrome, considering the anatomy of the auriculotemporal nerve.

Altered sensibility was also present in one half of the cases and in our patient and might also be associated with the development of Frey syndrome, because this would indicate injury to the auriculotemporal nerve. The difference in the latency period could be explained by the different hypotheses of the pathology of Frey syndrome.<sup>17</sup> The theory of regenerating nerves explains well the cases of the syndrome developing after 6 months or longer. The hypothesis of degenerating nerves could explain the cases with a faster onset of symptoms. The only patient with spontaneous remission had had a fast onset, which might suggest that late onset predicts a worse prognosis.

The diagnosis of Frey syndrome is based on the characteristic symptoms. In addition, the minor starch-iodine test is a sensitive diagnostic tool.<sup>8</sup> In the present case, thermography displayed the heat production characteristic of Frey syndrome. Thermography has previously been suggested as a valid method to document the temperature differences between the 2 sides of the face

**Table 1. PATIENT CHARACTERISTICS FROM PREVIOUS CASE REPORTS**

Investigator	Trauma	Other Fractures	Condylar Dislocation	Changed Sensibility	Latency	Course
Laws et al <sup>12</sup>	Fall from a scaffold	Ascending ramus	Medial dislocation	Hypersensitivity in the preauricular region	6 mo	Remission after resection of the temporomandibular nerve
Laws et al <sup>12</sup>	Bicycle accident	None	None	None	>6 yr	Chronic
Martis et al <sup>13</sup>	Car accident	Body, zygomatic arch	None	Hypersensitivity in preauricular region	1 mo	Spontaneous remission
Storrs et al <sup>16</sup>	Traffic accident	Symphysis, angle, other condyle	Posterior dislocation	Paresthesia in front of ear	2 yr	Chronic
Olson et al <sup>15</sup>	Motorcycle accident	Symphysis, coronoid, other condyle	None	None	3 mo	Chronic
Goodman et al <sup>11</sup>	Motorcycle accident	Parasymphyseal, other condyle	Not stated	None	Years	Not stated
Zoller et al <sup>17</sup>	Car accident	Symphysis, midface, skull base	Medial	Hypoesthesia in preauricular region	2-3 mo	Not stated
Dhaif et al <sup>1</sup>	Traffic accident	Parasymphyseal	Dislocated	None	3 mo	Not stated
Mellor et al <sup>14</sup>	Fall after fainting	Parasymphyseal, other condyle	Open through auditory meatus	Paresthesia in preauricular region	1 yr	Chronic
Gerbino et al <sup>10</sup>	Jump from 3rd floor	Symphysis, other condyle, Le Fort I	Medial dislocation	None	6-7 mo	Chronic

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(when only 1 side is affected).<sup>20</sup> This can be an aid in insurance cases, when the degree of permanent injury must be reported.

The treatment of Frey syndrome has been previously reviewed.<sup>4</sup> The condition can cause considerable social embarrassment and social incapacity because of the profuse flushing and sweating when eating, and the diagnosis should have an effect on the insurance benefits for the patient. Importantly, however, only a few patients with Frey syndrome will need treatment other than an explanation of the condition and reassurance. If treatment is required, the most effective treatment has been subcutaneous infiltration of botulinum toxin to the affected area.

In conclusion, the present case report has described the development of Frey syndrome after a dislocated mandibular condyle fracture managed by closed treatment. The findings from the literature review suggest that the complication might be underreported. The risk factors seem to be trauma with a considerable impact, more than 1 mandibular fracture site, dislocation of the mandibular condyle, and altered sensibility in the preauricular region. The minor starch-iodine test and thermographic imaging can supplement the clinical examination.

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