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Troubles spécifiques de la reconnaissance musicale chez des personnes avec ou sans lésion cérébrale

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Troubles spécifiques de la reconnaissance musicale chez des personnes avec ou sans lésion cérébrale

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Résumé

L'objectif de cette thèse est de dégager des arguments supplémentaires en faveur de l'autonomie du système musical par rapport aux autres fonctions cognitives. Pour ce faire, nous avons étudié des personnes, avec ou sans lésion cérébrale connue, manifestant un trouble de reconnaissance sélectif au domaine musical (agnosie musicale). Dans la première étude, nous avons vérifié si une rupture d'anévrisme situé à l'artère cérébrale moyenne pouvait causer des troubles de reconnaissance musicale. Nous avons donc évalué, avec un ensemble de tests mesurant la plupart des habiletés musicales en jeu dans la reconnaissance musicale, 20 patients ayant subi une chirurgie visant à ligaturer un anévrisme situé soit sur l'artère cérébrale moyenne gauche, sur la droite ou sur les deux, et 20 sujets contrôles neurologiquement intacts. L'étude démontre que ce type de chirurgie peut conduire à des déficits sélectifs dans le traitement auditif de la musique. Plus spécifiquement, les résultats montrent que les patients avec une chirurgie à l'artère cérébrale moyenne gauche sont plus perturbés que le groupe contrôle dans toutes les tâches où interviennent la mémoire musicale. Cette étude a également permis de découvrir deux nouveaux cas d'agnosie musicale apperceptive, comme en témoigne leur performance déficiente dans toutes les tâches de mémoire musicale et tous les tests de perception musicale. L'analyse neuroradiologique indique des lésions aux niveaux du lobe temporal supérieur et de l'insula de l'hémisphère droit. Les résultats concordent avec l'hypothèse voulant que l'agnosie apperceptive résulte de dommages aux structures de l'hémisphère droit, tandis que l'agnosie associative résulterait de dommages à l'hémisphère gauche. Dans la deuxième étude, nous avons recruté des personnes, sans lésion cérébrale connue, manifestant un désordre spécifique au domaine musical. Bien que décrite depuis plus d'un siècle, cette condition, que l'on appelle amusie congénitale, a peu de base empirique. Nous avons procédé à l'évaluation de 11 adultes amusiques congénitaux à l'aide d'une série de tests évaluant la présence et la spécificité des désordres musicaux. Les résultats révèlent que l'amusie congénitale est reliée à des déficiences sévères dans le traitement des variations de hauteur. Les problèmes musicaux s'étendent également à la mémoire et la reconnaissance musicales, de même qu'aux habiletés à chanter et à frapper au rythme de la musique. De plus, le désordre est spécifique au domaine musical car les amusiques congénitaux traitent et reconnaissent le langage (incluant la prosodie), les sons environnementaux et les voix humaines aussi bien que les sujets contrôles. Ainsi, cette étude démontre de façon convaincante l'existence de l'amusie congénitale comme un nouveau type de trouble d'apprentissage affectant sélectivement les habiletés musicales. Dans la troisième étude, les résultats obtenus lors des tests psychophysiques chez l'un des sujets amusiques congénitaux (Monica) indiquent des problèmes sévères pour détecter des changements de hauteur dans une séquence sonore. Ils suggèrent que les difficultés musicales des amusiques congénitaux pourraient découler de problèmes dans la discrimination fine des hauteurs, de la même façon que certains problèmes de langage pourraient provenir de déficiences dans le traitement temporel rapide.

Mots clés: agnosie musicale, amusie acquise, amusie congénitale, désordre auditif, trouble d'apprentissage, musique

Abstract

This thesis sought to evidence the idea that the musical system is autonomous from other cognitive functions. To this aim, we studied individuals, with or without known brain lesion, who manifested a musical agnosia, that is, a specific impairment in musical recognition. In the first study, we have verified if the rupture of an aneurysm located on the middle cerebral artery (MCA) results in disorders of music recognition. To this aim, 20 patients having sustained brain surgery for the clipping of a unilateral left (LBS), right (RBS) or bilateral (BBS) aneurysm(s) of the MCA and 20 neurologically-intact control subjects (NC) were evaluated with a series of tests assessing most of the abilities involved in music recognition. In general, the study shows that a ruptured aneurysm on the MCA that is repaired by brain surgery is very likely to produce deficits in the auditory processing of music. The results show that the LBS group was more impaired than the NC group in all three tasks involving musical long-term memory. The study also uncovered two new cases of apperceptive agnosia for music. These two patients (N.R. and R.C.) were diagnosed as such because both exhibit a clear deficit in each of the three music memory tasks and both are impaired in all discrimination tests involving musical perception. The lesions overlap in the right superior temporal lobe and in the right insula. Altogether, the results are also consistent with the view that apperceptive agnosia results from damage to right-hemispheric structures while associative agnosia results from damage to the left hemisphere. In the second study, we recruited individuals with no known lesion, who had a music-specific disorder, which we refer to as congenital amusia. This condition has been described for more than a century but has received little empirical attention. In this study, a group of 11 adults, responding to stringent criteria of musical disabilities, were examined in a series of tests originally designed to assess the presence and the specificity of musical disorders in brain-damaged patients. The results show that congenital amusia is related to severe deficiencies in processing pitch variations. The deficit extends to impairments in music memory and recognition as well as in singing and the ability to tap in time to music. Interestingly, the disorder appears specific to the musical domain. Congenital amusic individuals process and recognise speech, including speech prosody, common environmental sounds and human voices, as well as control subjects. Thus, the present study convincingly demonstrates the existence of congenital amusia as a new class of learning disabilities that affect musical abilities. In the third study, results obtained from psychophysical tests with one particular amusic case indicated the presence of a severe deficit in detecting pitch changes. Thus, this third study suggests that music-processing difficulties of the congenital amusics may result from problems in fine-grained discrimination of pitch, much in the same way as many language-processing difficulties arise from deficiencies in auditory temporal resolution.

Keywords: music agnosia, congenital amusia, amusia, auditory disorder, learning disabilities, music

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INTRODUCTION GÉNÉRALE

La musique est omniprésente dans toutes les cultures. On n'a qu'à penser à sa présence dans les différentes activités auxquelles on s'adonne ou aux différents endroits que l'on visite dans une journée pour réaliser son ubiquité dans notre société. La musique y occupe non seulement une place importante, mais elle le fait également dans le quotidien de beaucoup de gens sur la planète. D'ailleurs, l'industrie de la musique rapporte des milliards de dollars à travers le monde, signe d'un indéniable engouement planétaire pour celle-ci. Bien que la publicité et les médias favorisent la divulgation et la promotion de cet art, la musique date de bien avant l'arrivée des nouvelles technologies. En effet, la musique serait apparue il y a 40 000 à 80 000 ans, tel que le suggère la découverte récente d'une flûte d'os ayant appartenu à un homme de Néanderthal (Turk, Dirjec & Kavur, 1996). Il est donc difficile de nier la présence de la musique à travers les époques.

Le caractère universel et ancien de la musique soulève la question des fondements biologiques. En effet, il semble que la musique ne soit pas un artéfact culturel, mais que l'humain serait biologiquement prédisposé à l'apprécier. Plusieurs recherches en psychologie et en neuropsychologie de la musique apportent des appuis scientifiques à cette hypothèse. Un de ces appuis est la démonstration que les bébés possèdent déjà à six mois des capacités de perception mélodique et rythmique de la musique (pour une revue, Trehub, 2001). Ces habiletés découleraient davantage d'une prédisposition du cerveau à la perception musicale que d'un apprentissage car une période d'exposition de quelques mois n'est pas assez longue pour permettre le développement de nouvelles fonctions cérébrales.

Une autre façon d'appuyer l'hypothèse biologique de la musique est d'apporter des éléments de preuve scientifique quant à l'autonomie du système musical par rapport aux autres fonctions cognitives et à la présence de substrats neuroanatomiques dédiés au traitement musical. En effet, on s'attend à ce qu'une fonction biologique soit indépendante des autres fonctions cognitives, ce qui assure que la musique n'est pas le parasite ou le sous-produit d'une autre fonction cérébrale. De plus, une organisation neuronale biologiquement déterminée devrait avoir une localisation fixe d'un cerveau humain à un autre. Si la spécialisation du cerveau pour la musique résultait de la simple utilisation d'un espace neuronal libre suite à une stimulation précoce, cela signifierait que l'organisation cérébrale pour la musique répondrait aux pressions culturelles plutôt qu'aux forces biologiques. Dans un tel cas, on s'attendrait à une variabilité dans la distribution et la localisation des réseaux musicaux car la musique utiliserait aléatoirement n'importe quel espace libre dans le cerveau ou dans le cortex auditif. Ainsi, une organisation cérébrale prédéterminée devrait montrer une constance au niveau de sa localisation.

Les travaux de recherche exposés dans la présente thèse tentent de dégager des arguments additionnels en faveur de l'autonomie du système musical et de la présence de substrats neuroanatomiques voués au traitement musical. Pour démontrer l'autonomie du système musical, nous avons étudié des personnes manifestant des problèmes musicaux mais sans atteinte aux autres fonctions cognitives. Plus particulièrement, nous nous sommes intéressées aux personnes démontrant un trouble spécifique de reconnaissance musicale (agnosie musicale) car la reconnaissance d'un air musical est une activité effectuée par la majorité des gens (profanes ou érudits) et qui ne nécessite aucune éducation musicale particulière. De plus, comme la reconnaissance musicale requiert la capacité à traiter plusieurs composantes (p.ex. perception mélodique), elle permet d'étudier un ensemble d'habiletés musicales. Nous avons d'abord étudié les problèmes de reconnaissance musicale chez des personnes ayant une lésion cérébrale. Pour trouver des patients cérébrolésés manifestant un tel trouble, nous nous sommes basées sur les trois personnes atteintes d'agnosie musicale étudiées dans notre laboratoire qui avaient toutes subi une opération pour ligaturer un anévrisme sur l'artère cérébrale moyenne (Peretz, 1996; Peretz et al., 1994, 1997). Nous avons donc sélectionné et évalué des patients caractérisés par cette même histoire neurologique. Nous avons également voulu jeter un regard nouveau sur l'agnosie musicale et apporter davantage de poids aux fondements biologiques de la musique en étudiant des personnes manifestant un trouble spécifique de reconnaissance musicale d'origine congénitale.

Cette thèse a également pour but, par l'analyse du cerveau de patients manifestant des déficits musicaux, de préciser les réseaux neuronaux en jeu dans la reconnaissance musicale et de déterminer si leur localisation est constante. Finalement, l'analyse systématique des problèmes musicaux de ces personnes a fourni l'occasion de comprendre davantage le fonctionnement du cerveau et, plus spécifiquement, celui du cortex auditif.

Étapes identifiées dans la reconnaissance de la musique occidentale

Éléments constitutifs de la musique occidentale

Tel que mentionné précédemment, la musique est un art répandu dans toutes les sociétés humaines. Par contre, l'organisation musicale des sons diffère à travers les époques et les sociétés. Dans notre système musical occidental, système que l'on qualifie de tonal, les pièces musicales comportent une structure harmonique et une structure monodique. La structure harmonique consiste en une succession d'accords (plusieurs notes jouées simultanément). Cependant, dans la musique occidentale, la monodie occupe une place particulière car elle suffit, à elle seule, pour permettre une reconnaissance musicale (Peretz, Babaï, Lussier, Hébert & Gagnon, 1995). Elle sera donc traitée en profondeur dans le paragraphe suivant.

La monodie est une suite de notes reliées les unes aux autres et formant un tout cohérent. Ces notes peuvent varier selon plusieurs paramètres sonores (hauteur, durée, timbre, intensité). Seuls les paramètres de hauteur et de durée sont abordés ici car ils sont considérés comme les plus informatifs sur la structure de la monodie occidentale (Terhardt, 1978). La variation de la durée permet l'établissement d'une dimension temporelle qui est composée du rythme (la durée relative des notes) et de la métrique (l'alternance de temps forts et de temps faibles). Le traitement de la hauteur permet l'établissement d'une dimension mélodique qui est composée de trois paramètres primordiaux : l'intervalle, la tonalité et le contour mélodique. L'intervalle correspond à la différence de hauteur entre deux sons consécutifs, la tonalité renvoie aux règles gouvernant la combinaison des différents sons et le contour est défini comme la trajectoire des fréquences fondamentales (F0)[1] à travers le temps. La perception d'une pièce musicale requiert donc une analyse simultanée de plusieurs paramètres sonores et constitue ainsi une tâche complexe pour le cerveau.

Modèle de reconnaissance musicale

Pour tenter d'illustrer et de comprendre la façon dont le cerveau procède pour reconnaître une mélodie, Peretz (1993) a élaboré un modèle de reconnaissance musicale issu des études avec des sujets normaux et cérébrolésés. Ce modèle décrit les différentes étapes et habiletés musicales nécessaires à la reconnaissance musicale. Suivant ce schéma, lors de l'écoute musicale, l'auditeur opère une analyse structurale des variations séquentielles des hauteurs (appelée organisation mélodique) et, parallèlement, effectue une analyse de ses particularités temporelles (appelée organisation temporelle). L'organisation mélodique comprend l'extraction du contour, des intervalles et de la tonalité, et l'organisation temporelle le rythme et la métrique. L'extrait musical ne serait reconnu que lorsqu'il y a un appariement adéquat entre les représentations fournies par les deux voies d'analyse, principalement la voie mélodique, et la représentation emmagasinée en mémoire au niveau du répertoire. La voie temporelle aurait moins d'impact sur la reconnaissance musicale (Hébert et Peretz, 1997).

Troubles spécifiques de la reconnaissance musicale : Évidences de l'autonomie du système musical et de l'existence de structures dédiées au traitement musical

Bien que pour la plupart des gens la reconnaissance d'un air musical se fasse naturellement et sans aucun effort conscient, il en est autrement pour un faible nombre d'individus. En effet, certaines personnes éprouvent des troubles musicaux les conduisant à une incapacité à reconnaître un air musical connu, c'est-à-dire à une agnosie musicale. Cette situation peut se produire suite une atteinte cérébrale à un endroit particulier du cerveau (Griffiths et al., 1997; Peretz, 1996; Peretz et al., 1994, 1997; Piccirilli et al., 2000). Il s'agit alors d'une agnosie musicale acquise, mais nous

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[1] La fréquence fondamentale (F0) correspond à la fréquence la plus basse d'un son et donne la sensation de hauteur du son. Elle constitue donc le paramètre physique d'un son (mesurée en Hertz),

utilisons généralement le terme plus large d'amusie acquise car la plupart des patients ont des atteintes musicales ne se limitant pas à la reconnaissance (par exemple, troubles de perception et de production). Par ailleurs, des troubles de reconnaissance musicale peuvent également s'observer chez des personnes, sans aucune atteinte cérébale connue, n'ayant pas réussi à développer normalement certaines compétences musicales de base (Geshwind, 1984; Grant Allen, 1878). Le terme "amusie congénitale" est utilisé dans ces cas-ci à cause du caractère inné du déficit musical. Ce qui est d'autant plus fascinant, c'est que la perte ou l'absence de développement de ces fonctions musicales surviendrait de façon sélective, c'est-à-dire sans que d'autres fonctions cognitives soient perturbées .

Étude de patients manifestant une amusie acquise

Les désordres de la reconnaissance musicale sont relativement communs suite à une atteinte cérébrale. Ils sont également connus depuis longtemps. Le premier cas à être rapporté dans la littérature est celui d'un patient de Bonvicini en 1905 (cité dans Peretz, 2000). Ce patient ne pouvait plus reconnaître des chansons connues même si le traitement de l'information musicale était fonctionnel. En effet, il pouvait détecter une fausse note (note hors tonalité) insérée dans un extrait musical, mais ne pouvait pas reconnaître l'extrait entendu. Par contre, l'agnosie musicale pure est un phénomène rare car les atteintes musicales surviennent souvent en présence d'une perturbation des autres événements auditifs, c'est-à-dire le langage et les sons environnementaux (pour une revision voir Dalla Bella et Peretz, 2000; Peretz, 1993). On parle alors d'agnosie auditive.

Pour prouver l'autonomie du système musical, on doit tenter d'obtenir ce que les neuropsychologues appellent une "double dissociation". Dans le cas présent, cela signifie la découverte de patients avec une perturbation du langage ou des sons environnementaux, mais sans atteinte aux fonctions musicales, et celle de la situation inverse, c'est-à-dire des patients avec une atteinte musicale, mais une préservation du langage et des sons environnementaux. Plusieurs cas d'agnosie du langage (aphasie réceptive) sans amusie et un cas d'agnosie pour les sons environnementaux sans

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tandis que la hauteur désigne l'attribut sensoriel d'un son (mesurée en ton).

amusie ont été rapportés dans la littérature (pour une revision voir Dalla Bella et Peretz, 2000). La dissociation inverse, c'est-à-dire une amusie sans aphasie, est cependant rare. Récemment, quelques cas d'agnosie musicale sans aphasie et sans trouble de reconnaissance des sons environnementaux ont été observés. D'abord, Peretz et ses collaborateurs (1994, 1996, 1997) ont étudié trois patients (C.N., G.L. et I.R.) manifestant une telle dissociation. Les équipes de Griffiths (1994) et de Piccirilli (2000) ont également chacune rapporté un cas similaire d'agnosie musicale. La reconnaissance des événements musicaux semble donc indépendante des autres fonctions cognitives, même du langage.

Suivant le modèle de reconnaissance musicale détaillé plus haut, l'agnosie musicale peut avoir deux origines. D'abord, elle peut découler d'un problème à traiter les variations de hauteur dans une mélodie. Une telle perturbation de la perception mélodique empêcherait l'information musicale de se rendre et de s'apparier aux représentations musicales en mémoire afin de vérifier si l'extrait musical correspond à une mélodie connue. Cependant, les représentations en mémoire à long terme seraient intactes, mais elles ne seraient plus accessibles par la voie mélodique. Un problème de reconnaissance découlant d'une atteinte perceptive fait partie de la catégorie des agnosies apperceptives. L'autre forme d'agnosie musicale résulte d'une atteinte des représentations musicales en mémoire, sans que les habiletés perceptives soient perturbées. Cette forme de désordre est connue sous le nom d'agnosie associative.

Ainsi, selon ce modèle, quatre des cinq patients agnosiques musicaux signalés plus haut souffriraient de la forme apperceptive de l'agnosie. D'abord, H.V., étudié par Griffiths et al (1997), démontre un trouble de perception des hauteurs en plus d'une agnosie musicale. En effet, il ne peut discriminer des sons de hauteur différente dans une séquence rapide, mais il est capable de chanter de mémoire, signe d'une préservation des représentations en mémoire. Le patient étudié par l'équipe de Piccirilli (Piccirilli et al., 2000) ne peut discriminer des mélodies par les variations de hauteur et est incapable de chanter une mélodie. Deux des patients décrits par Peretz, G.L. et I.R. (Peretz et al., 1994, 1997), manifestent également une atteinte importante de la perception des variations de hauteur dans une mélodie accompagnant leur agnosie (Peretz et al., 1994, 1997). En effet, G.L. ne peut plus reconnaître des airs familiers et discriminer des mélodies différant au niveau des variations de hauteur. Il peut, par contre, chanter, mais avec quelques erreurs au niveau de la hauteur. De façon un peu similaire, I.R. ne peut plus reconnaître des airs familiers ou discriminer des mélodies et est incapable de chanter. Par contre, une troisième patiente de Peretz (1996), C.N., a évolué vers une agnosie musicale associative, malgré une agnosie apperceptive initiale. En effet, elle a récupéré la plupart de ses habiletés perceptives musicales, mais continue d'être incapable de chanter de mémoire, de nommer une chanson familière, de juger de la familiarité d'un extrait musical ou de reconnaître des airs musicaux nouveaux ou familiers.

L'analyse du type de lésion et de l'histoire neurologique de ces patients agnosiques donne des indications sur les régions cérébrales intervenant dans la perception et la reconnaissance musicales. Les trois patients étudiés par Peretz ont tous la même histoire neurologique. En effet, ils ont subi plusieurs chirurgies dans le but de ligaturer des anévrismes situés sur l'artère cérébrale moyenne de chaque hémisphère. Les régions cérébrales irriguées par cette artère semblent donc importantes dans la perception et la reconnaissance musicales. Chez le patient de l'équipe de Piccirilli, le gyrus temporal supérieur a été lésé par un hématome causé par une malformation artéroveineuse. Par contre, la lésion se situait à l'hémisphère gauche uniquement, ce qui s'explique par la latéralisation à l'hémisphère droit des fonctions du langage chez ce patient gaucher. Finalement, le patient H.V. a souffert d'une atteinte dans les territoires de l'artère cérébrale moyenne et postérieure de l'hémisphère droit seulement. À la lumière de ces études de cas, une lésion à l'hémisphère droit (ou non dominant) semble suffir à causer une agnosie apperceptive (comme chez H.V. et le patient de l'équipe de Piccirilli), mais une atteinte bilatérale serait nécessaire pour provoquer une agnosie associative, c'est-à-dire une perturbation du système de représentation en mémoire (comme chez C.N.).

L'importance de l'hémisphère droit, plus particulièrement du gyrus temporal supérieur droit, pour la perception des variations de hauteur dans une mélodie, a d'ailleurs été démontrée par plusieurs études de groupes avec patients cérébrolésés (Liégeois-Chauvel et al., 1998; Peretz, 1990; Samson et Zatorre, 1988) et par l'utilisation de la tomographie par émissions de positons (TEP) chez des sujets normaux (Zatorre et al., 1994). Plusieurs recherches ont également démontré une contribution des aires frontales droites dans le traitement mélodique (Shapiro et al., 1981; Zatorre et Samson, 1991; Zatorre et al., 1994), possiblement parce que ces régions permettraient de maintenir l'information auditive active en mémoire de travail (Marin et Perry, 1999; Perry, 1990).

Les réseaux neuronaux des composantes mnésiques en jeu dans la reconnaissance musicale sont plus élusifs. L'apprentissage et la rétention à long terme de nouvelles mélodies semblent être reliés davantage à l'hémisphère droit (Plenger et al., 1996; Samson et Zatorre, 1991, 1992), mais les deux hémisphères pourraient aussi intervenir (Peretz, 1990; Zatorre, 1985). Cependant, une étude avec la technique TEP a montré que la reconnaissance d'airs musicaux familiers dépendrait plus de l'hémisphère gauche (Platel et al., 1997). Devant ces résultats contradictoires, d'autres études sont nécessaires afin de préciser la latéralisation de la mémoire musicale.

Les études auprès de patients avec une amusie acquise sont donc informatives à plusieurs niveaux. D'abord, elles permettent de mieux comprendre la façon dont le cerveau procède pour reconnaître un air musical. La spécificité des problèmes musicaux observés chez ces patients suggère également que le système musical est indépendant des autres fonctions cognitives. De plus, ces recherches donnent des indications sur les régions cérébrales impliquées dans la perception et la mémoire musicales, bien que ces informations demeurent incomplètes, particulièrement concernant la mémoire. Ces études sont donc un appui à l'existence de circuits neuronaux voués au traitement musical. Cependant, il est possible que l'exposition régulière à la musique et ce, dès les premières années de la vie, ait favorisé la création de connexions neuronales dans des régions cérébrales non biologiquement spécialisées pour la musique. Ainsi, certaines structures, qui n'étaient pourtant pas dédiées au traitement musical, deviendraient spécialisées dans l'analyse et l'interprétation des sons musicaux. Lorsque lésées, ces structures ne seraient plus en mesure de traiter correctement la musique. L'hypothèse de la stimulation musicale à la base de la spécialisation de régions cérébrales pour la musique n'est pas incompatible avec l'amusie acquise, mais elle rend plus difficilement compte de l'amusie congénitale. En effet, la découverte de personnes n'ayant pas réussi à acquérir sélectivement les habiletés musicales de base, et ce, malgré une exposition normale à la musique et l'absence de lésions cérébrales, indiquerait que la stimulation musicale fréquente et précoce ne serait pas le seul facteur expliquant la spécialisation de certaines aires cérébrales pour la musique. Cette observation serait donc un appui supplémentaire à l'existence de réseaux biologiquement voués au traitement musical.

Études de personnes manifestant une amusie congénitale

Les problèmes sévères d'acquisition du langage, lesquels ne découlent pas d'une surdité, d'un retard mental ou d'un manque de stimulation par l'environnement, sont largement connus et étudiés dans la littérature (p.ex. Gopnik et Crago, 1991). Par contre, peu de chercheurs se sont intéressés aux personnes, sans lésion cérébrale connue, ne possédant pas les éléments essentiels au développement normal des habiletés musicales élémentaires. Cette condition a été appelé de façon variée « tune-deafness », « tone-deafness » ou dysmusie dans la littérature. Cependant, nous préférons utiliser le terme moins restrictif d'amusie congénitale car il peut y avoir autant de formes d'amusies développementales que d'amusies résultant de dommages cérébraux acquis à l'âge adulte. De plus, dans la littérature anglophone, le préfixe « dys » ne signifie pas nécessairement un trouble développemental, mais peut être interprété comme un trouble moins sévère que lorsqu'on utilise le préfixe « a ».

Même si nous n'avons pu retracer que deux études de cas dans la littérature, ce phénomène serait connu depuis plus d'un siècle. En effet, le premier cas d'amusie congénitale fut répertorié par Grant Allen en 1878. Dans cet article, on décrit le cas d'un homme de 30 ans, avec une solide éducation et sans lésion cérébrale connue, rapportant un grave problème musical. L'homme était, entre autres, incapable de chanter et de distinguer des sons de hauteur différente, et de reconnaître des mélodies de chansons connues. De plus, il éprouvait une indifférence face à la musique. Le manque d'exposition musicale ne pouvait expliquer ce trouble car cet homme avait reçu une formation musicale dans son enfance. Un siècle plus tard, Geshwind rapportait un cas similaire (Geshwind, 1984). L'homme venait d'une famille affichant de piètres habiletés musicales, en dépit de fréquentes expositions aux enregistrements musicaux à la maison. Lorsqu'il était enfant, cet homme a tenté de suivre des cours de piano, mais le professeur a réalisé très tôt que son élève ne pouvait ni chanter ou discriminer deux notes de hauteurs différentes, et ni garder la pulsation rythmique.

Bien qu'informatives, ces deux études demeurent anecdotiques car elles ne sont que descriptives et ne sont pas appuyées par des évaluations systématiques. À ces études de cas s'ajoutent quelques recherches épidémiologiques. En 1948, Fry publie une étude menée auprès de 1200 sujets. Selon ses résultats, 5 % de la population anglaise serait «tone deaf» (terme équivalent à l'amusie congénitale) lorsque évaluée dans des tests de détection de fausses notes et de jugement de similarité entre deux notes ou deux phrases musicales. L'auteur conclut que les troubles de mémoire musicale à court et à long terme aussi bien que les problèmes de discrimination des hauteurs sont les facteurs les plus déterminants dans l'amusie congénitale. Cependant, le manque de précisions méthodologiques (les résultats ne sont pas rapportés) empêche de juger de la validité de cette recherche. Plusieurs années plus tard, Kalmus et Fry (1980) complètent une étude, cette fois chez 604 adultes, où ils estiment à 4,2% la fréquence d'adultes anglais dits «tone deaf». Ces chercheurs considèrent "tone deaf" les sujets qui commettent trois erreurs ou plus à une tâche de détection de fausses notes dans une mélodie. Cependant, cette estimation est problématique. Premièrement, la mesure manque de sensibilité puisque plus de 90% des individus n'ont commis aucune erreur. Deuxièmement, le recours à une seule mesure de l'habileté musicale entraîne des faiblesses à la validité et la fiabilité. Finalement, une telle définition psychométrique de l'amusie congénitale n'est pas convaincante car la seule considération d'une extrémité de la distribution normale lors d'un unique test ne fournit pas une preuve convaincante que l'amusie congénitale est une pathologie réelle, et non une anomalie statistique.

En somme, ces études sur l'amusie congénitale fournissent des informations intéressantes sur la nature de ce trouble, bien qu'elles n'offrent pas une base empirique solide. On retrouve tout de même une constance dans les cas publiés. En effet, des problèmes de discrimination des hauteurs, de perception mélodique et de mémoire musicale sont souvent rapportés par les auteurs. Les fonctions cognitives non musicales semblent d'ailleurs épargnées chez les sujets amusiques, mais ces fonctions n'ont pas été évaluées formellement.

Grâce au modèle de reconnaissance exposé plus haut, on peut émettre des hypothèses quant au(x) niveau(x) du traitement musical possiblement atteint(s) chez les amusiques congénitaux. Tout comme l'amusie acquise, le sujet avec une amusie congénitale peut avoir une atteinte au niveau de la perception ou de la mémoire musicales. Étant donné le caractère inné du déficit, on pourrait s'attendre à une absence de répertoire musical chez les sujets avec un problème sévère de perception des variations de hauteur dans une mélodie car ce trouble perceptif n'aurait pas permis l'encodage et l'emmagasinage de l'information musicale (agnosie apperceptive). Si, au contraire, aucun déficit perceptif notable n'est signalé chez le sujet amusique congénital, le trouble se situerait au niveau de la mémoire et on serait donc en présence d'une agnosie associative. Les problèmes d'encodage, d'emmagasinage ou de consolidation en mémoire pourraient également entraver la création d'un répertoire musical chez les personnes avec une agnosie associative congénitale. Cependant, contrairement aux cas d'amusie acquise, ce problème ne peut pas faire suite à une atteinte (perte) du répertoire car aucun accident cérébral n'est survenu. Les cas d'amusie congénitale étudiés jusqu'à maintenant orientent davantage vers la forme apperceptive de l'agnosie musicale.

Troubles de perception et de reconnaissance musicales : hypothèse d'une atteinte au traitement de bas niveau pour les hauteurs

Tel que mentionné précédemment, la plupart des amusiques étudiés manifestent des difficultés de discrimination des variations de hauteur dans une mélodie accompagnant leur agnosie musicale. Comme la musique n'est pas le seul événement auditif où l'on retrouve des variations de hauteur, ce trouble de discrimination pourrait également s'observer dans un contexte auditif non musical. Si tel est le cas, le problème de discrimination des hauteurs ne se situerait pas à un niveau musical, mais à un plus bas niveau dans le traitement auditif, ce qui expliquerait pourquoi les événements auditifs non musicaux seraient aussi affectés par ce trouble discriminatif.

Similairement, une hypothèse d'atteinte au traitement de bas niveau a été avancée pour expliquer les troubles de compréhension du langage. En effet, plusieurs études suggèrent que les problèmes d'acquisition du langage chez les enfants dysphasiques[2] dériveraient de difficultés à traiter les changements temporels rapides des sons (p. ex. Tallal et Piercy, 1973). Des recherches effectuées chez des patients aphasiques suite à une lésion au lobe temporal gauche ont également montré un seuil de détection de changement temporel plus élevé chez ces patients que chez les sujets normaux (Swisher et Hirsh, 1972). L'analyse temporelle fine serait donc importante dans le décodage du langage car la perception des consonnes est reliée à l'habileté à traiter des différences temporelles de l'ordre du dixième de seconde (pour une revision, voir Zatorre et al., 2002).

Même si les variations temporelles semblent déterminantes, d'autres composantes, telles que la hauteur, sont également analysées par le système auditif pour décoder le langage. On pourrait donc s'attendre à ce qu'un trouble de discrimination des hauteurs perturbe également la compréhension du langage, et non seulement la musique. Alors, comment peut-on expliquer la spécificité du trouble au domaine musical si celui-ci origine d'un problème de bas niveau pour la discrimination des hauteurs? Une hypothèse est que le traitement fin des hauteurs soit essentiel à la perception musicale, mais non au décodage des autres événements auditifs comme le langage. Cette hypothèse est appuyée par une étude de Shannon et collaborateurs (1995) qui a démontré, à l'aide d'un matériel où seules les variations temporelles étaient présentes (l'information spectrale étant enlevée), que les changements temporels suffisent pour décoder le langage.

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[2] La dysphasie est un trouble d'acquisition du langage oral réceptif ou expressif ne découlant pas d'un problème d'acuité auditive, d'un retard mental ou d'un manque de stimulation dans l'environnement.

L'hypothèse d'une atteinte au traitement de bas niveau des hauteurs chez les amusiques est compatible avec les données neuroanatomiques. En effet, les mêmes régions cérébrales, c'est-à-dire le gyrus temporal supérieur droit, semblent actives au niveau du traitement des hauteurs dans une mélodie et dans un contexte non-musical. Zatorre (1988) a montré que les patients avec une excision du cortex auditif droit, mais non de gauche, éprouvaient des difficultés à percevoir la hauteur des sons dont la fondamentale est absente. De plus, Johnsrude et collaborateurs (2000) ont démontré que les patients avec une excision du lobe temporal droit, qui empiète sur le gyrus de Heschl, ont un seuil plus élevé que les contrôles ou les patients avec une ablation du lobe temporal gauche pour détecter la direction du changement de hauteur dans une séquence sonore. D'ailleurs, Zatorre et collaborateurs (2002) ont dernièrement fourni plusieurs éléments de preuve de la spécialisation de l'hémisphère droit pour la discrimination fine des variations de hauteur. Par contre, aucune étude avec des patients cérébrolésés n'a observé des troubles de discrimination des hauteurs pour les tâches simples (p. ex. dire si les sons entendus sont pareils ou différents). Même les patients C.N., G.L. et I.R., qui pourtant ont subi des lésions bilatérales au cortex auditif et éprouvent d'importants problèmes de discrimination des variations de hauteur dans une mélodie (du moins pour les patients G.L. et I.R.) ne démontrent aucun problème dans la discrimination de deux sons par la hauteur (Peretz et al., 1994; Peretz et al., 2001). Cette capacité semble donc très résistante à une lésion cérébrale corticale, possiblement parce que ce traitement peut être effectué à plusieurs niveaux. Par exemple, des structures sous-corticales peuvent effectuer, mais de façon moins efficace, le traitement des hauteurs de son.

Bien qu'il soit peu probable qu'une atteinte au traitement auditif de bas niveau soit à la base de l'amusie acquise, il pourrait en être autrement pour un trouble congénital. En effet, les structures nécessaires au traitement des hauteurs pourraient ne pas s'être développées et organisées correctement dans le cerveau des personnes atteintes d'une amusie congénitale. D'ailleurs, les sujets amusiques congénitaux étudiés par Grant Allen (1878) et Geshwind (1984) éprouvaient des difficultés à dire si deux sons entendus étaient identiques ou différents. Ainsi, une atteinte à la discrimination des hauteurs pourrait nuire à la perception et la reconnaissance mélodique chez ces personnes, sans affecter les autres fonctions cognitives.

Hypothèses de recherche

Afin d'appuyer notre postulat voulant que la musique soit indépendante des autres fonctions musicales et que certains circuits neuronaux soient voués au traitement musical, et plus spécifiquement à la reconnaissance musicale, trois hypothèses sont émises dans la présente thèse.

Premièrement, nous nous attendons à ce que les patients ayant subi une ligature d'anévrisme sur l'artère cérébrale moyenne droite ou dans les deux hémisphères aient de la difficulté à percevoir des variations de hauteur dans une mélodie et à mémoriser (reconnaître et identifier) des mélodies musicales.

Nous prévoyons également découvrir des personnes, sans lésion cérébrale connue, n'ayant pas réussi à développer les habiletés musicales nécessaires à la reconnaissance musicale. Ces personnes (les amusiques congénitaux) devraient avoir une faible performance dans les tâches comportementales évaluant la perception des variations de hauteur dans une mélodie et la mémoire musicale (reconnaissance et identification de mélodies musicales). Comme conséquence à ce désordre perceptif, les amusiques congénitaux devraient également manifester une faible capacité de chanter. En ce qui concerne les compétences temporelles (perception du rythme et de la métrique, frapper des mains au rythme de la musique), nous ne formulons pas de prédiction particulière car les données disponibles sont peu concluantes. Finalement, nous prédisons que l'administration d'une tâche psychoacoustique de détection de changement de hauteur dans une séquence sonore révèle un trouble auditif de bas niveau chez les amusiques congénitaux.

Finalement, nous posons l'hypothèse que les troubles musicaux observés chez les amusiques (acquis et congénitaux) seront limités au domaine musical. Ainsi, nous nous attendons à ce que les résultats aux batteries d'évaluation des fonctions intellectuelles et mnésiques générales se situent dans la normalité. De plus, l'évaluation du langage et des sons environnementaux devrait démontrer une préservation de ces fonctions. Ces tests d'identification et de reconnaissance de sons non musicaux ont d'ailleurs été construits le plus similairement possible aux tests musicaux, afin de minimiser la contribution d'autres facteurs.

Pour vérifier ces hypothèses, trois études utilisent un ensemble d'examens neuropsychologiques, psychophysiques et neuroradiologiques. Dans la première étude, des tests sont administrés à un groupe de 20 patients ayant subi une opération visant à réparer un anévrisme sur l'artère cérébrale moyenne à l'hémisphère droit ou gauche ou aux deux hémisphères. Leurs résultats sont comparés à ceux d'un groupe de 20 sujets contrôles appariés. La deuxième étude comprend un groupe de 11 personnes, sans lésion cérébrale connue, s'avérant très déficientes dans une batterie musicale de dépistage évaluant des habiletés perceptives et mnésiques musicales. Une de ces participantes a également fait l'objet d'une étude de cas dans la troisième étude. Les résultats obtenus par les sujets amusiques congénitaux sont comparés à ceux d'un groupe contrôle de sujets normaux appariés et non appariés.

PREMIER ARTICLE : PATTERNS OF MUSIC AGNOSIA ASSOCIATED WITH MIDDLE CEREBRAL ARTERY INFARCTS

Patterns of music agnosia associated with middle cerebral artery infarcts

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Abstract

The objective of the study is to evaluate if the rupture of an aneurysm located on the middle cerebral artery (MCA) results in disorders of music recognition. To this aim, 20 patients having sustained brain surgery for the clipping of a unilateral left (LBS), right (RBS) or bilateral (BBS) aneurysm(s) of the MCA and 20 neurologically-intact control subjects (NC) were evaluated with a series of tests assessing most of the abilities involved in music recognition. In general, the study shows that a ruptured aneurysm on the MCA that is repaired by brain surgery is very likely to produce deficits in the auditory processing of music. The incidence of such deficit was not only very high but also selective. The results show that the LBS group was more impaired than the NC group in all three tasks involving musical long-term memory. The study also uncovered two new cases of apperceptive agnosia for music. These two patients (N.R. and R.C.) were diagnosed as such because both exhibit a clear deficit in each of the three music memory tasks and both are impaired in all discrimination tests involving musical perception. Interestingly, the lesions overlap in the right superior temporal lobe and in the right insula, making the two new cases very similar to an earlier case report (H.V.) reported by Griffiths and his collaborators (1997). Altogether, the results are also consistent with the view that apperceptive agnosia results from damage to right-hemispheric structures while associative agnosia results from damage to the left hemisphere.

Key words: auditory agnosia, amusia, auditory organization, music, hemispheric differences

List of abbreviations : BBS = bilateral brain surgery; LBS = left brain surgery; MCA = middle cerebral artery; NC = normal control; RBS = right brain surgery.

Recognition of familiar music is immediate and easy for every human being. Despite its apparent effortlessness, music recognition is a complex procedure that implies multiple processing components. Damage to one or many of these components produces music agnosia. Such a neurologically-based deficit is characterized by the inability to recognize music in the absence of sensory, intellectual, verbal and mnesic impairments (Peretz, 1996).

As Peretz (1993) has argued elsewhere, music agnosias may either have a perceptual melodic basis or a memory basis. Music recognition may be conceptualized as a two-stage process as illustrated in Figure 1. According to this model, music agnosia may be due to a failure to properly encode melodic information, defined by sequential variations of pitch. Such a perceptual melodic impairment would prevent the familiar musical passage from making contact with its stored representation. The long-term memory representations may, however, be spared by the brain damage although the traces are no longer accessible by auditory input. Such a recognition deficit due to a perceptual defect falls into the class of apperceptive agnosias. The other form of music agnosia results from an isolated loss of memories for music. That is, the breakdown can spare most perceptual abilities but interfere with the recognition process by damaging the network of the long-term memory representations of music. This form of disorder is known as associative agnosia.

[Insert Figure 1 about here]

According to this model, the patient H.V. studied by Griffiths and collaborators (1997) would suffer from an apperceptive form of music agnosia. Actually, H.V. has been shown to suffer from a perceptual defect and to be able to sing from memory. Conversely, Peretz (1996) has described a case of associative agnosia for music. This case, C.N., who had recovered most perceptual skills, is still unable to sing from memory, to name a familiar tune, to judge its familiarity, or to memorize familiar and novel music. Since H.V. suffered from an unilateral lesion in the right hemisphere and C.N. suffered from bilateral damages, one can tentatively propose that apperceptive agnosia is related to a right-sided lesion and associative agnosia is associated to bilateral infarcts.

The two patients, H.V. and C.N., differ not only in the nature of their music agnosic disorder but also in neurological history. H.V. suffered from a unilateral posterior infarction involving the posterior superior temporal gyrus and the inferior parietal and anterolateral occipital lobes in the right hemisphere. C.N. had more anterior lesions in both superior temporal gyri resulting from successive surgeries for the repair of an aneurysm located on the middle cerebral artery (MCA). Interestingly, C.N.'s neurological history is identical to that of two fruther cases of music agnosia, G.L. and I.R., who were independently discovered and whose selection was symptom-based (Peretz et al., 1994; Peretz et al., 1997). All three sustained successive brain surgeries for the clipping of aneurysms located in mirror position on each MCA. This particular brain condition associated to surgical intervention might be conductive to music agnosia. One goal of the present study was to test these neural correlates of music agnosia.

The other major objective of the present study was to assess the idea that apperceptive agnosia is more likely to arise from a right-sided infarct of the superior temporal gyrus because it would compromise the melodic route. As seen in Figure 1, there are two main processing routes that are assumed to lead to memory representations: The melodic and the temporal routes. However, the melodic route is conceived as having primacy for accessing stored music representations. In effect, it is relatively well-established that the essential processing components of the melodic route lie in the right superior temporal gyrus (Peretz, 1990; Liégeois-Chauvel et al., 1998; Zatorre et al., 1994) with possible connections with the right frontal areas (Zatorre & Samson, 1992; Zatorre et al., 1994). Moreover, this right-sided melodic route can be interrupted without disturbing the temporal route (Peretz, 1990; Peretz & Kolinsky, 1993; Liégeois-Chauvel et al, 1998). Finally, this isolable melodic route is conceived as primary because neurologically-intact subjects have been shown to use more effectively melodic features than temporal patterns to recognize familiar musical selections (White, 1960; Hébert & Peretz, 1997). Therefore, there is ground to consider apperceptive agnosia as resulting from a right-sided interruption of the melodic route.

The neural correlates of the memory component of the music recognition system are more elusive. Learning and long-term retention of novel melodies seem to rely more on the integrity of the right than the left hemisphere (Samson & Zatorre, 1991, 1992; Plenger et al., 1996; but see Zatorre, 1985, and Peretz, 1990, for bilateral involvement). However, recognition of highly familiar music has been shown to depend more on the left hemisphere (Platel et al., 1997). To avoid the confusion created by variable levels of prior familiarity with the musical material, both highly familiar and totally unfamiliar musical excerpts will be tested here for memory recognition. Yet, no clear prediction as to the side(s) of the infarct(s) that would lead to selective impairments of memory recognition for music (i.e. associative agnosia) will be formulated at this stage.

In the present study, in order to test the neural underpinning of music agnosia, the musical agnosic pattern associated with brain surgery for the clipping of an aneurysm located on the left, the right or both MCAs was investigated in twenty patients and their neurologically-intact matched controls. The participants were presented with the same series of musical tests as the ones used in earlier studies with music agnosics (Peretz et al., 1994, 1997) and with patients with unilateral temporal excisions for the relief of epilepsy (Liégeois-Chauvel et al., 1998). These tests have been designed to test the major processing components that are known to be involved in music recognition. These include several discrimination tasks in which pitch contour, pitch interval and scale steps are assessed on the melodic dimension, and rhythm and regularity on the temporal dimension. Several music recognition tasks are included as well to assess the memory component of the music recognition system. These tests cover memory recognition of novel musical excerpts as well as recognition and identification of well-known musical selections. The results of the patients obtained across these multiple tests should provide us with a good picture of the relation between the surgery on the MCA and the ensuing musical condition.

Method

Subjects

Twenty patients having sustained brain surgery for the clipping of a ruptured aneurysm located on the temporal region of the right (n = 10), the left (n = 7) or both (n = 3) MCAs participated to the present study. Informed consent was obtained from all of them. CT scans were carried out from 1 day to 63 months post surgery (mean: 15 months) with 10 mm axial section. Not all patients showed evidence of brain infract on CT scan examination (MRI scans could not be obtained due to the use of metallic clips). However, they all sustained the same brain surgery which was performed by the same neurosurgeon (the last author --M.B.). The patients were tested, on average, 30 months (range: 6 months to 7 years) post-operatively. The majority of patients (15) were evaluated more than one year post-operatively. A summary of the patients' characteristics is presented in Table 1 along with the Wechsler intelligence scale revised (1981) scores and the Wechsler Memory scale (1974) scores. The right (RBS) and left (LBS) brain-surgery patient groups were not found to differ in I.Q. ($F(2,17) = 2.12$, n.s.) or in M.Q. ($F<1$). Two LBS patients had some aphasic problems but their speech comprehension was preserved.

[Insert Table 1 about here]

There were twenty neurologically-intact controls (NC) who were selected to match the brain-damaged patients in age, sex, handedness, education and musical background. In Table 1, the sex distribution, average age and years of education are summarized for each group. A history of alcohol abuse, psychiatric disorder or other neurological illness was grounds for exclusion. Only people raised in the French culture of Quebec were selected in order to have a homogeneous group with respect to musical knowledge. Most participants were right-handed to the exception of two LBS patients (and their respective matched normal controls) one of whom was left-handed and the other ambidextrous. None of the subjects was currently or has recently been involved in music. Only 10 percent of the subjects in each group could be considered as having had some musical experience, given that they practiced an instrument during childhood.

Material and procedures

Eight behavioral tests involving monodies that obeyed the rules of the Western tonal system were employed. Six of them use the same pool of 30 *novel* monodies and are used in what we refer to as the musical battery. Two further tests, the familiarity decision test and the identification test, employ monodies that are expected to be familiar to anyone from the Quebec Francophone culture.

Musical battery: The musical battery, which is fully described in Liégeois-Chauvel, Peretz, Babaï, Laguitton and Chauvel (1998), is composed of six tests, three of which deal with pitch variation discrimination, two with temporal variation discrimination and one with memory.

In the Pitch organization conditions, three types of manipulation were applied to the same tone in 15 sequences. One manipulation consisted in creating a scale-violated alternate melody by modifying the pitch to bring it out of scale (within the same semi-tone distance across stimuli), in keeping with the original contour. This change is particularly salient because the changed pitch sounds out of tune (see melody B in Fig. 2). The second manipulation consisted in creating a contour-violated alternate melody by modifying the critical pitch so that it changed the pitch direction of the surrounding intervals while maintaining the original key (see melody C in Fig. 2). The third manipulation consisted in creating a contour-preserved or interval-violated alternate melody of the contour-violated and scale-violated melodies by modifying the same critical pitch to the same extent (in terms of semi-tone distance), while maintaining the original contour and scale (see melody D in Fig. 2). Average pitch interval changes were made equivalent across the three conditions.

[Insert Figure 2 about here]

Three sets of stimuli, each consisting of 2 practice trials and 30 experimental trials, were constructed with these melodies. Each trial consisted of a warning signal and a target melody followed by a comparison melody after a 2 s. silent interval. Duration of the inter-trial interval was 5 s. long. A first set, which was prepared for the scale-violated condition, was constructed so that 15 trials were made of identical melodies and 15 trials of different scale-violated melodies. The second and third set, which were designed for the contour-violated and the interval-violated condition, respectively, were similar to the scale-violated condition set in that they kept the same target melodies; the only modification was that each comparison melody was replaced by its contour-violated alternate or by its preserved-contour alternate. Melody pairs were presented in each set in a random order. These three conditions will be referred to as the scale, contour and interval conditions, respectively. Subjects were required to perform a "same-different" classification task. They had to judge, on each trial, whether the target and the comparison sequence were the same or not.

The temporal organization tasks involved two tests, one rhythmic and one metric test. For the rhythmic test, the stimuli were the same as those used in the pitch organization tests. To create different comparison patterns, a change in the duration values of two adjacent tones was applied to keep the meter and the total number of sounds identical. The serial positions of these changes varied across patterns (see melody E in Fig. 2). Thus, the only cue available for discrimination was the rhythmic pattern. A set of 2 practice and 30 experimental trials was constructed with the temporal patterns. The task also required a "same-different" classification. For the metric test, two-phrase sequences instead of the one-phrase sequences used in the previous tests were recorded in a random order with a 5 s. inter-trial interval. Half of these sequences were written in a duple meter and half were written in a triple meter. Subjects were informed that they would be hearing waltzes and marches which they had to discriminate along this dimension (see melody F in Fig. 2). They were encouraged to tap along with what they perceived to be the underlying beat of each sequence. There were four practice trials preceding 30 experimental trials.

The last test of the musical battery was a memory recognition test. From the initial set of 30 single-phrase melodies, 15 were selected for the recognition part of this study. Each had been presented at least five times in the same format. In addition to these "old" melodies, a set of 15 recognition foils was prepared. The "new" melodies were constructed along the same principles, but differed from the "old" ones in their exact temporal and pitch pattern. The 30 sequences were then recorded in a random order with a 5 s. silent interval in between. The subjects were requested to respond "yes" if they recognized a melody as having been presented earlier during the session and to respond "no" if otherwise. This last test came as an incidental memory test since the subjects were not informed in advance that their memorization of the material would be tested later.

Familiarity decision test: The melodic part of the beginnings of 40 folk songs selected from a list of pieces well-known to Quebec Francophones (Peretz et al., 1995) were mixed, in a random order, with 40 melodies coming from the same repertoire of folk songs but which were unfamiliar because they are no longer sung or played. The familiar melodies had a mean rating of 4.5 (following our norms, 1 means unfamiliar and 5 highly familiar). The duration of melodies, which was on average 8.5 s. long, was equivalent in familiar and unfamiliar excerpts. There was a 5 s. silent interval between melodies. The subjects had to judge, on each trial, if the melody was familiar or unfamiliar. Prior to the task, two practice trials were presented and feedback on the response was only provided for the two practice trials.

The identification test: Fifty-two melodic beginnings of folk songs were chosen from the same pool of familiar musical excerpts (Peretz et al., 1995). They were associated to a mean familiarity rating of 4.6 and lasted 9 s., on average. The melodies were separated by a 5 s. silent interval. Each presented melody was associated with a choice of 4 written titles, one of which was the correct title. The foils were of the same genre (for example, all titles would be Christmas songs). The subjects were first invited to give the title of the heard melody; in case of failure, they were presented with the 4 written choices from which they had to choose. No feedback on the accuracy of the choice was provided.

All stimuli were generated on an IBM-AT compatible microcomputer controlling a Yamaha TX-81Z synthesizer. The voice was the approximation of a piano sound. The analog output was recorded on a digital DAT SONY recorder which was also used to play melodies to the subjects. The subjects were run individually in two sessions of about two hours each with as many pauses in between conditions as requested. They listened to the pre-recorded tapes via a speaker placed on a table in front of them. The intensity level was adjusted to a confortable level for each subject. Subjects were presented, successively, with the pitch organization conditions (scale-violated, contour-violated and interval-violated), the temporal organization tasks (rhythmic task, metric task), the memory recognition test of unfamiliar melodies, the recognition test of familiar melodies and the music-title identification test. The musical battery was always presented in the same session.

Results and comments

Individual scores were transformed into hits and false alarms except for the identification test where it was not appropriate, and examined by way of analyses of variance (ANOVAs). The scores obtained on different tests were grouped in the same analysis when the tests were assessing abilities in comparable experimental conditions. For example, both the memory recognition test and the familiarity decision test involve a binary decision that requires explicit memory for melodies. Accordingly, the test scores were examined together in the same ANOVA. Non-parametric tests were performed when the results were not homogeneous. Because there were few subjects in the BBS group, the results for this group were not included in the statistical analyses but they are presented in Figures and Tables. All individual scores were examined (fully presented in Table 2) and classified with respect to the lowest performance obtained by the NC which was considered as the cut-off point below which the scores can be regarded as indicating a genuine deficit. Note, however, that this criterion is conservative since some scores above the cut-off points could reflect deficient systems that were excellent premorbidly. The results will be presented below according to the processing component that is assessed by the test(s), starting with the diagnostic tests for music agnosia followed by the more perceptual tests and ending with lesion localization.

Recognition tests

The old-new recognition tests: The responses were considered as hits when the subjects responded "old" to studied melodies and "familiar" to well-known melodies, and as false alarms when responding "old" to nonstudied or unfamiliar melodies. The mean percentages of hits and false alarms obtained by each group in the memory recognition test and the familiarity decision test are presented for each group in Figure 3. Since the two tests mainly differ in terms of pre-experimental familiarity with the melodies, they will be treated in a single ANOVA. Hits and false alarms were submitted to separate ANOVAs with the test material (familiar and unfamiliar melodies) taken as the within-subjects factor and the three groups (NC, LBS, RBS) as the between-subjects factor. On hits, the analysis yielded an effect of Test material ($F(1,34)=8.41$, $P < .01$) but no effect of Group ($F<1$) nor interaction between the two factors ($F<1$). Subjects performed generally better in the familiarity decision test that in the memory recognition one. False alarm rates exhibit a slightly different pattern, as supported by the presence of an interaction between Test material and Group ($F(2,34)=3.26$, $P < .05$). The LBS group was found to produce more false alarms than the NC group ($P < .001$) and the RBS group ($P < .06$) in the memory recognition test (by way of Tukey a posteriori tests). In the familiarity decision test, LBS patients also made more false alarms than the NC group ($P < .02$) but not significantly more so that the RBS patients. Thus, all subjects performed well on these melody recognition tests when hit rates are examined. However, patients with LBS tend to make more false recognition than the other groups. This impairment is, however, not as serious as the deficit exhibited by BBS patients (see Figure 3 and Table 2 for the individual scores) who performed poorly on the memory recognition test.

[Insert Figure 3 about here]

The identification test: The mean percentages of correct responses for each group obtained globally by adding the correct naming responses to the correct title choices are presented, along with the naming responses, in Figure 4. The two scores were analyzed separately with Group as the between-subjects factor. On both the global scores and the correct naming scores, a Group effect was obtained, with $F(2,34)=7.37$ and 4.20, both $P < .05$, respectively. Tukey comparisons reveal that this group effect was due to the lower performance of the LBS group ($P < .02$ compared to the NC group). Note that, on this identification test, the LBS patients are as impaired as the BBS patients (see also Table 2 for individual data). Thus, a left-brain surgery seems to interfere with the ability to reliably recognize familiar melodies.

[Insert Figure 4 about here]

The results obtained across the three recognition tests --the memory recognition test of studied novel melodies, the familiarity decision test and the identification test-- allow the classification of patients as exhibiting agnosic symptoms or not. Patients exhibiting a deficit in any of the recognition tests is qualified as music agnosic. As expected from the previous analyses, there are more agnosic patients after LBS (3 out of 7) and BBS (2 out of 3) than after RBS (2 out of 10; see Table 2). However, if we use more stringent criteria, as is currently applied to single case studies, and require evidence of a deficit in each of the music recognition tests, since all of them aim at assessing the same processing component (the memory component in Figure 1), then only two patients can be qualified as showing clear evidence of music agnosia. These two patients are RBS16 and BBS19 (see Table 2), hence suggesting a contribution of the right-hemispheric structures. The origin of the agnosic problem is expected to differ for LBS (and BBS) and RBS patients, with the latter pertaining more to the apperceptive type of agnosia and the former to the associative type. The scores obtained in the discrimination tests, to be examined next, will allow the classification of the patients according to each type of agnosia.

[Insert Table 2 about here]

Perceptual tests

In the discrimination tests, a "different" response given to a " different " trial was considered as a hit whereas a "different" response given to a "same" trial was considered as false alarm. Since the false alarm rates were found to be very similar in all patients groups, we used the number of hits minus the number of false alarms as a unique discrimination score in both statistical analyses and in Figures and Tables to simplify data description.

In the <u>pitch organization tests,</u> the hits minus false alarms scores were submitted to an ANOVA with the three conditions (scale, contour, interval) as the within-subjects factor and the three groups (NC, LBS, RBS) as the between-subjects factor. As can be seen in Figure 5, the RBS group appears impaired in all conditions while the LBS group seems to experience less difficulties with the scale condition. However, this different pattern lacks robustness since no significant interaction between Group and Condition was obtained ($F < 1$,n.s.). The patients were impaired across conditions, irrespective of the melodic condition considered and the side of the surgery ($F(2,34)=3.57$, $P < .04$, for the Group effect). Nonparametric statistical analyses of the data yielded essentially to the same results

[Insert Figure 5 about here]

Inspection of individual data (in Table 2) reveals a series of interesting facts. First, out of the nine patients exhibiting a deficit in the contour condition, six were clearly, and three mildly, impaired in the interval test. This systematic association between the deficits observed in contour and interval conditions is consistent with the anchorage role conferred to the contour for encoding intervals (Peretz, 1990). Secondly, three patients (RBS16, BBS19 and BBS20) demonstrated a deficit in all three pitch tests. These three patients are expected to have agnosic disorders since there is converging evidence that their melodic route does not function properly, hence interfering with the most essential access to music memory (see the model in Figure 1). The latter prediction is consistent with the data. As can be seen in Table 2, all three patients show signs of recognition failures.

For the temporal organization tests, the mean percentages of hits minus false alarm scores are displayed in Figure 6. A correct response to a "waltz" was considered here as a hit and a "waltz" response given to a march was considered as false alarm. Performance on the two tasks was analyzed separately because task parameters were different, the meter task requiring an on-line judgment for each sequence and the rhythm task requiring a "same-different" classification for two such sequences. No significant group effect was obtained in neither task (with $F(2,34)=2.69$ and $F<1$, for the rhythmic and metric test, respectively).

[Insert Figure 6 about here]

Examination of individual scores are more informative than group data, as was the case for the pitch organization conditions. As can be seen in Table 2, performance is highly variable in the metric task and is associated with a very low cut-off score. Consequently, results in this particular test are difficult to interpret. This is not the case for the rhythmic test on which the large majority of patients perform within the normal range. Only two out of the 20 patients show a deficit in rhythm discrimination. The latter result suggests that either rhythmic deficits are less frequent than melodic ones or that the brain surgery under study is less detrimental to rhythm processing than other types of brain accident, such as brain excision in the temporal lobe for the relief of epilepsy which will be examined next.

Incidence of musical disorders

As pointed out previously, brain surgery for the clipping of an aneurysm located on the MCA was thought to be instrumental in producing music perception and music recognition disorders. In order to assess the validity of such a claim, we compared the frequency of occurrence of a deficit in the present study with a previous study of ours where the same musical tests were administered to another population. The latter sample was made of 62 patients having sustained unilateral excision of brain tissue in the temporal lobe for the relief of epilepsy. The percentages of patients exhibiting a deficit (defined as a score below the lowest score of NC subjects) in each test that was used in both studies are presented in Table 3.

[Insert Table 3 about here]

Although the present study includes fewer patients than the previous one, the comparison is very instructive. In effect, the incidence of pathological performance, defined as a score falling outside the normal distribution, is much higher in the present sample than in the epileptic population (Liégeois-Chauvel et al., 1998). This may, however, not be related to the type of brain surgery. The functional brain organization of the two populations may well differ pre-operatively. The premorbid brain of the individuals tested in the present study was, in all likelihood, similar to the one of the normal controls. This is probably not the case of the epileptic brain which has often been ill-functioning since early childhood, and hence, may have known some functional reorganization. Therefore, we cannot conclude that surgical repair of a ruptured aneurysm on the MCA is a perfect road to music agnosia. Any vascular infarct of the same territories may well lead to a similar high rate of musical deficits.

It is, however, remarkable to note that bilateral intervention appears more disturbing than unilateral intervention. It does not seem to be a trivial mass effect, whereby the more damaged tissue the more likely a deficit will be observed. Bilateral intervention seems to spare temporal organization processes while severely compromising both melodic organization processes and memory.

Lesion localization

Out of the 20 patients' post-operative brain CT scans, only 15 could be examined for infarction localization. Two scans were not available and the extent of infarction could not be visualized in three scans because they were taken in the acute ischemic phase. Moreover, among the 15 scans, five contained artifacts in the regions of interest due to the presence of metallic clips on the MCA, preventing us from properly examining the temporal lobe structures. Two neuroradiologists (the authors I.R. and C.B.) reviewed the cases according to the following structures: The anterior, middle and posterior portion of the superior temporal gyrus, the middle temporal gyrus, the temporal pole, the inferior parietal lobe and the frontal operculum. A summary is presented in Table 4.

[Insert Table 4 about here]

As indicated in Table 4 and as expected, most patients show infarcts involving structures of the superior temporal gyrus, except cases LBS3 and RBS9. These two cases do not undermine the idea that the critical areas for music processing lie in the superior temporal gyrus because the surgical act involves compression of those areas and subsequent lesions often do not appear on CT scans, particularly clinical CT scans like the ones obtained here which are of low resolution. Of more interest are the CT scans of the two music agnosic patients identified in the present study (RBS16 and RBS19) which do show some overlap in their lesion localization. Both have visible lesions in the right primary auditory cortex (heschl gyrus), the right temporal pole and the right insula. The images of the scans corresponding to these structures are presented in Figure 7. The right temporal pole may not be critically associated with music agnosia since the only unimpaired patient (RBS8), whose performance lies in the high normal range (see Table 2) and for whom we had a readable CT scan, shows a visible lesion in that region (see Table 4). Moreover, excision of that particular region has been shown to have little impact on music discrimination and memory except for the metric test (Liégeois-Chauvel et al., 1998). The right primary auditory area (Heschl gyrus) must also play a role, although its damage does not seem to be mandatory since the right Heschl gyrus was spared in I.R., C.N. and G.L. Interestingly, the right insula appears as a serious possibility since it is damaged in all documented cases of music agnosia (in H.V., Griffiths et al., 1997; in C.N. and in G.L., Peretz et al., 1994; and in I.R., Patel et al., 1998, for more detailed reports and images of the respective scans).

[Insert Figure 7 about here]

General discussion

The results confirm the high incidence of disorders of music perception and memory that result from the surgical repair of aneurysm(s) located on the MCAs. The results also emphasize the usefulness of systematically investigating patients having sustained such brain accident so as to shed light on the neural correlates of the seemingly rare condition of music agnosia. Such an approach has led to the discovery of two new cases of music agnosia. It also led to evidence that the left side of the brain is better equipped for hosting long-term memory representations of music and that the right side is essential in mediating access to these stored representations.

Two clear-cut cases of music agnosia have been identified. Milder forms of music agnosia were found in five other cases, amounting the chances to find patients with music recognition disorders to 35% (7 out of 20) in an unselected sample of patients having undergone brain surgery for the clipping of an aneurysm on the MCA. When present, the disorder consists in failures to properly encode the musical material and in poor performance on at least one of the three memory tests that were used in the study. Since the memory deficits were systematically associated with deficits in the discrimination tests, the pattern is most consistent with the observation of disorders pertaining to apperceptive agnosias. Associative agnosia, reflecting preserved perceptual abilities in the absence of recognition skills, must be rare since we did not observe such cases here.

Of the two clear-cut cases of apperceptive agnosia documented here, one (RBS16) is a 51-year-old right-handed woman, to which we will refer as N.R., who works as a hairdresser. The other (BBS19), to which we will refer as R.C., is a 44-year-old man who works as an audio-visual technician in college. Both are musically uneducated although the second has recently (and ironically) started to learn to play the accordion. These two cases are considered clear instances of the agnosic syndrome because they consistently and systematically performed below the normal range in all three tests that required music memory consultation. Both performed poorly in the recognition of recently learned melodies, in the discrimination of familiar from unfamiliar melodies, and in identifying highly familiar melodies by naming or title recognition. All tests require normal access to an intact memory network for music; such access was obviously deficient in these two patients. Both N.R. and R.C. performed very poorly in all tests that require discrimination of musical sequences along the melodic and temporal dimension. Thus, both N.R. and R.C. must experience difficulties to properly encode musical information. This encoding deficiency may account for their poor recognition skills for music. This perceptual impairment is not general, though. The music agnosic disorder appears more marked for music than for other domains. For instance, both patients were able to distinguish between lyrics taken from familiar songs and familiar idiomatic expressions (this test is referred to as "familiarity decision" in Table 5). They were able to provide the titles of songs for which they can no longer recognize the isolated melody when presented with the corresponding spoken lyrics. R.C. performed much better than N.R. in this task (see Table 5); it should be noted, however, that the task is particularly laborious even for neurologically-intact subjects and that N.R.'s performance falls in the low normal range. However, R.C.'s auditory disorder is not music-specific either. R.C. is slightly impaired in the recognition of non-verbal patterns, such as when recognizing speakers' voices and when invited to recognize particular categories of environmental sounds (Faïta et al., 1996).

[Insert Table 5 about here]

The discovery of these two music agnosic patients increases our studied pool to five cases, all having the same neurological etiology. This constitutes a relatively high hit rate, given the paucity of similar cases of music agnosia in the literature (Dalla Bella & Peretz, 1999; Griffiths et al., 1999). This observation supports in turn our hypothesis that brain damage incurred by the clipping of a ruptured aneurysm on the MCA is likely to lead to music agnosia and hence may serve as a good resource for studying this particular form of auditory agnosia. In this respect, it is worth mentioning that the deficit is not transient since both patients (N.R. and R.C.) have been tested at least 3 years post-operatively. Note, however, that the music agnosic problem does not appear as severe as the one observed in our prior bilateral cases (C.N., G.L. and I.R.) but is similar to that experienced by the unilateral case H.V. (Griffiths et al., 1997). Similar to H.V., both N.R. and R.C. show damage to the right side in the brain images, an evidence that a right-sided lesion appears sufficient to produce the music agnosic disorder. Images of the brain of N.R. (bottom images) and of R.C. (top) are presented in Figure 7. Unilaterality of the lesion is perhaps not warranted in the case of R.C. since he sustained brain surgery on the left side as well, for clipping a mirror aneurysm before it ruptured. Nevertheless, it is worth emphasizing that all three cases (H.V., N.R. and R.C.) of apperceptive agnosia for music have damage to similar areas in the right side of the brain. All three cases show evidence of brain lesions in the right posterior region of the superior temporal gyrus and in the right insula. These regions are in all likelihood critically involved in music processing for the purpose of recognition. It remains to be determined if the insula association is accidental, and thus silent with respect to music recognition, or rather instrumental.

As seen in these two cases of apperceptive music agnosia, a right-sided lesion appears to disrupt recognition of music because of a perceptual defect and not because of a memory defect per se. As mentioned above, the music recognition disorder is probably due to a failure to properly encode musical information. By this account, the perceptual impairment would prevent the presented musical passage from leaving new memory traces or, when familiar, from making contact with stored memory representation. Such perceptual defect, when occurring along the melodic organization pathway, is typically associated with a right-hemisphere infarct (Peretz, 1990; Liégeois-Chauvel et al., 1998; see Figure 1). This lateralization effect was also observed in the present study. The large majority of patients (six out of seven) who were found to suffer from a deficit in discriminating melodies along the pitch dimension had sustained brain surgery on the right side of the brain. This right-sided surgery apparently interferes with melodic contour formation, as suggested here and demonstrated in several prior studies (Peretz, 1990; Liégeois-Chauvel et al., 1998).

Another important finding in the present study concerns the observation of a left-hemisphere contribution to music recognition. A left-sided lesion was found to disrupt performance in all three tasks requiring recognition of melodies. Patients having undergone a surgery on the left MCA generally display depressed performance in the recognition of recently studied melodies, in deciding whether a melody is familiar or not, and in recognizing the title of heard familiar melodies. This memory problem is a graded one in that it mainly emerges as a group effect. An obvious account of this left-sided bias to music memory is to relate it to the well-established superiority of the left-hemisphere in verbal behavior. By this account, left-brain surgery would depress memory performance because of a diminished propensity to use verbal mediation as a mnemonic. This verbalization account is always difficult to dismiss even if the task requirements did not make any specific verbal demands. For instance, the memory recognition task of novel melodies and the familiarity decision test do not require any verbal mediation; only the identification test does. However, the music memory scores were found to be highly correlated with the logical story memory test scores ($\underline{r} = .73$) of the Wechsler memory scale and not with the nonverbal visual reproduction score ($\underline{r} = .04$) of the same scale. Thus, the left-brain surgery patients appear to suffer from a verbal memory deficit as well. This verbal memory deficit might be the product of a mere association with the musical memory deficit since the brain lesion can interfere with two adjacent but nevertheless separate functions. By this accidental association account, the left-hemispheric structures would be the depository of the music representation system depicted in Figure 1 (and referred to as the "repertoire" in previous studies; e.g. Peretz, 1996). Alternatively, if verbalization did contribute to the music memorization tests, then the memory impairment exhibited by the left-brain damaged patients would simply reflect the intervention of a verbal strategy. Future studies exploiting brain imagery techniques with normal brains should help to tease apart these two interpretations.

Whatever the exact interpretation of the left-side effect on music memory may be, the observed contribution of the left-hemispheric structures to the recognition of music coupled with the confirmation that right-hemispheric structures are instrumental in allowing perceptual access to these memories fit with classical views of agnosias. It is a recurrent proposal, even in the auditory domain, that apperceptive agnosia and associative agnosia are associated to damage to the right and left hemisphere, respectively. In the late sixties, Faglioni et al. (1969; later confirmed by Vignolo, 1982) proposed such a division of functions between hemispheres for auditory agnosias, that is, for the recognition of nonverbal and nonmusical sounds such as animal cries. These neurologists noticed that patients who were impaired in tasks requiring discrimination of the acoustic pattern of sounds had a right brain lesion. In contrast, a deficit in the identification of familiar environmental sounds (by pointing to visual objects as possible sources) was observed in left brain-damaged patients. These results have recently been confirmed by Schnider and al. (1994). A similar idea has been proposed for the recognition of voices (Van Lancker et al., 1988) and, in vision, for the recognition of objects (Warrington, 1985). The present study extends this view to the musical domain.

To conclude, damage to the territory irrigated by the middle cerebral is likely to lead to deficits in music perception and recognition. These deficits are probably also related to the surgical gesture that compresses the superior temporal gyrus and the structures hidden inside the sylvian fissure (which also hosts the primary cortex). Unfortunately, images of the lesions were poor due to the presence of metallic clips. The use of titanium clips will permit further studies with MRI technology. Nevertheless, the outcome of the present study shows that the scope of the disorders arising after MCA infarcts is wider and richer than traditionally construed. Hence, it is likely that these patients will continue to offer a unique opportunity to study music-related deficits but also to investigate brain organization that is conductive to auditory disorders in general.

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Table 1. Characteristics of subjects.

Group	Sex		Age	Education	I.Q. (s.e.)	M.Q. (s.e.)
	M	F	(years)	(years)		
LBS	1	6	49	10	97(1,54)	105 (2,11)
RBS	1	9	47	13	108 (1,41)	111 (0,92)
BBS	1	2	51	12	99 (1,64)	102 (2,12)
NC	3	17	48	13	-	-

Note : s.e. = standard error

Table 2

Individual hits minus false alarms rates in each test and percentage of correct responses in the identification test for each patient. Cut-off points, mean percentages and corresponding standard deviations, for normal controls are given for each test.

	Pitch organization			Temporal organization		Memory Rec.	Familiarity Dec.	Identification
	Scale	Contour	Interval	Rhythmic	Metric			Global
LBS1	67	20*	47	47	73	40	43*	65*
LBS2	93	47	67	80	40	80	98	100
LBS3	47	40*	7*	73	60	7*	88	81
LBS4	73	80	73	33*	7*	27*	70	67*
LBS5	87	87	73	67	87	60	75	81
LBS6	73	53	53	47	67	33	83	79
LBS7	80	80	47	67	53	40	70	96
RBS8	100	80	87	80	87	87	93	100
RBS9	27*	40*	33	60	67	20*	78	81
RBS10	60	80	53	100	40	60	83	96
RBS11	47	27*	20*	40	73	47	80	92
RBS12	53	0*	13*	87	33	67	85	87
RBS13	93	87	87	93	87	80	100	100
RBS14	47	73	60	67	27	67	85	100
RBS15	93	93	93	100	87	87	78	100
RBS16**	0*	-7*	-7*	0*	33	27*	63*	75*
RBS17	67	67	53	80	73	60	90	98
BBS18	53	20*	33	53	53	33	75	98
BBS19**	13*	20*	13*	40	13*	27*	55*	69*
BBS20	33*	40*	20*	73	53	-7*	75	71*
cut-off point	47	47	33	40	20	33	65	79
mean	81	74	71	80	58	72	91	96
s.d.	15	14	19	16	20	17	7	6

Note : * = scores under the cut-off point; ** = agnosic patients; s.d. = standard deviation.

Table 3

Percentages (and number) of patients with temporal surgery for the repair of an aneurysm (present study) and for the excision of epileptic tissue (Liégeois-Chauvel et al., 1998) in each task used in both studies.

		Scale	Contour	Interval	Rhythm	Meter	Memory rec.
Aneurysm							
	Left (7)	0	29	14	14	14	29
	Right (10)	20	40	30	10	0	20
	Bilateral (3)	67	100	67	0	33	67
	Total (20)	**20**	**45**	**30**	**10**	**10**	**30**
Epilepsy							
	Left (27)	0	11	11	0	11	0
	Right (35)	11	6	9	11	31	14
	Total (62)	**6**	**8**	**10**	**7**	**23**	**8**

Table 4

Summary of CT scan low density areas.

	Heschl	STGa	STGm	STGp	MTG	PT	IPL	FO	Insula	Frontal
LBS1 (lefthanded)	+	+	+							
LBS2	–	–	–	–	–	–				
LBS3										+
LBS4	+	+	+	+	+	+	+	+	+	
LBS6				+		+	+	+	+	
LBS7 (ambidextrous)	–	–	–	–	–	–				+
RBS8						x				
RBS9									x	
RBS10	–	–	–	–	–	–				x
RBS11		x								
RBS14	x	x	x	x	x					x
RBS16**	x	x	x	x	x	x	x		x	x
RBS17		x								x
BBS18	x	x	x	x	x	x +	x		x	
BBS19**	x					x			x	

Note : "**" = agnosic patients; "x" = right lesion; "+" = left lesion; "_" = not assessed due to artefacts; STG = superior temporal gyrus (a = anterior; m = middle; p = posterior); MTG = middle temporal gyrus; PT = planum temporal ; IPL = inferior parietal lobule; FO = frontal operculum.

Table 5. Performance of RBS16 / N.R. and BBS 19 /R.C. on neuropsychological non-musical tests

Tests	N.R.	R.C.
Audiometry	---	mild bilateral loss for frequencies > 3 kHz
I.Q.	87	105
Verbal	88	108
Performance	89	98
M.Q.	97	103
Lyrics recognition		
Naming	13/22	18/23
Familiarity decision	20/20	18/20
Voice recognition	---	22/33
Environmental sounds recognition	---	33/45

---: not assessed

Figure captions

Figure 1. Peretz's (1993) model of music agnosias.

Figure 2. Example of an initial melody (A), its scale-violated (B), its contour-violated (C), its interval-violated (D) and its rhythmic (E) transformation. F represents the entire two-phrase sequence (of which the second phrase corresponds to A) used in the metric task. * indicates the critical note.

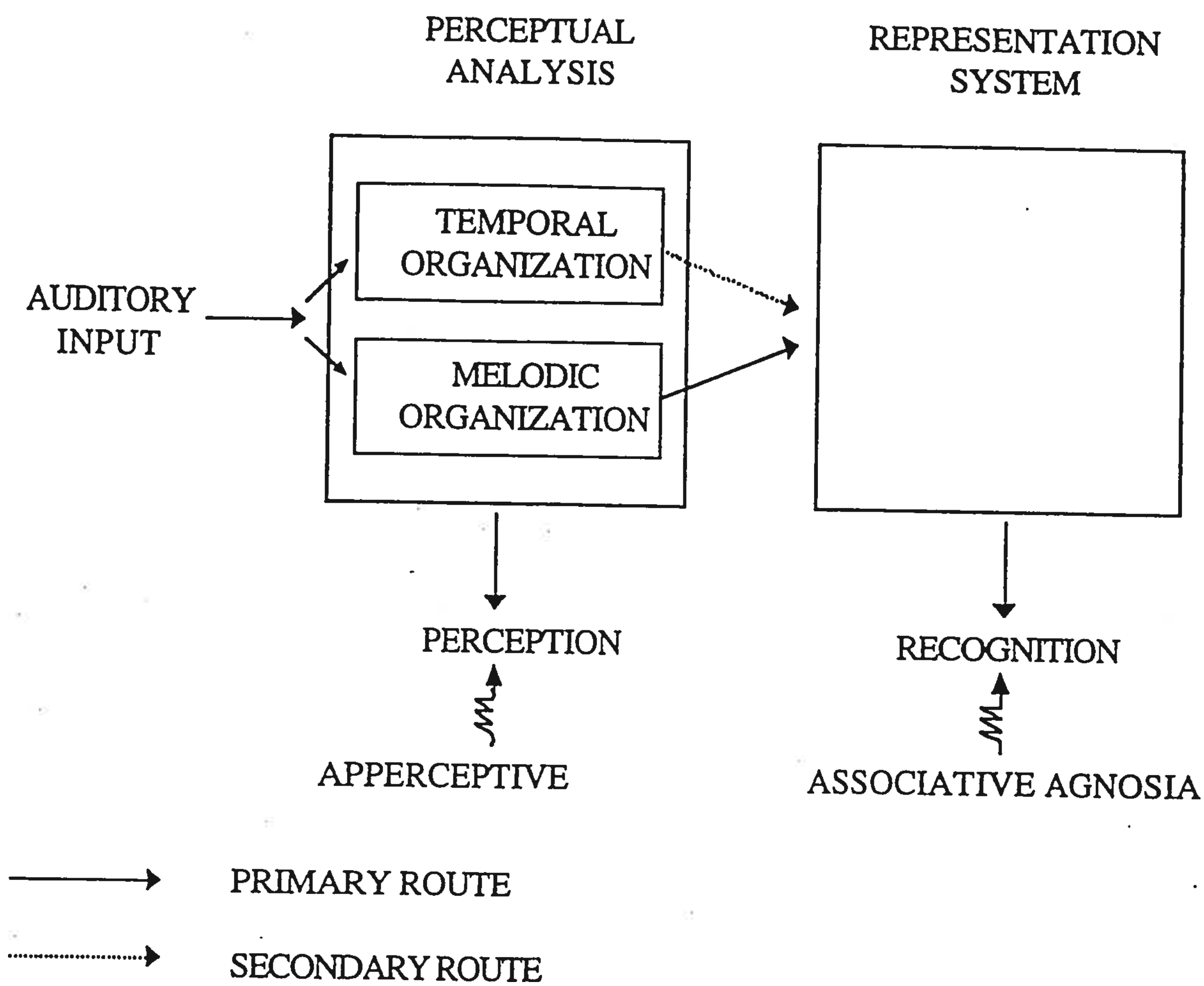
Figure 3. Mean percentages of hits and false alarms obtained, in each group, for the two old-new recognition tests. Error bar represents standard error.

Figure 4. Mean percentages of correct responses obtained, in each group, for the identification test. The global score includes both correct naming and correct title selection. Error bar represents standard error.

Figure 5 Mean percentages of hits minus false alarms obtained, in each group, for the three pitch organization tests. Error bar represents standard error.

Figure 6. Mean percentages of hits minus false alarms obtained, in each group, for the two temporal organization tests. Error bar represents standard error.

Figure 7 Axial CT scan images of the brain of two music agnosic patients, BBS19 or R.C. (top images, without contrast injection) and RBS16 or N.R. (bottom images, with contrast injection): Middle cerebral artery infarct involving the insular cortex (☢) and the temporal lobe (→) including Heschl's gyrus. Right is on the left.


PERCEPTUAL
ANALYSIS
REPRESENTATION
SYSTEM
TEMPORAL
ORGANIZATION
AUDITORY
INPUT
MELODIC
ORGANIZATION
PERCEPTION
APPERCEPTIVE
RECOGNITION
ASSOCIATIVE AGNOSIA
PRIMARY ROUTE
SECONDARY ROUTE

A

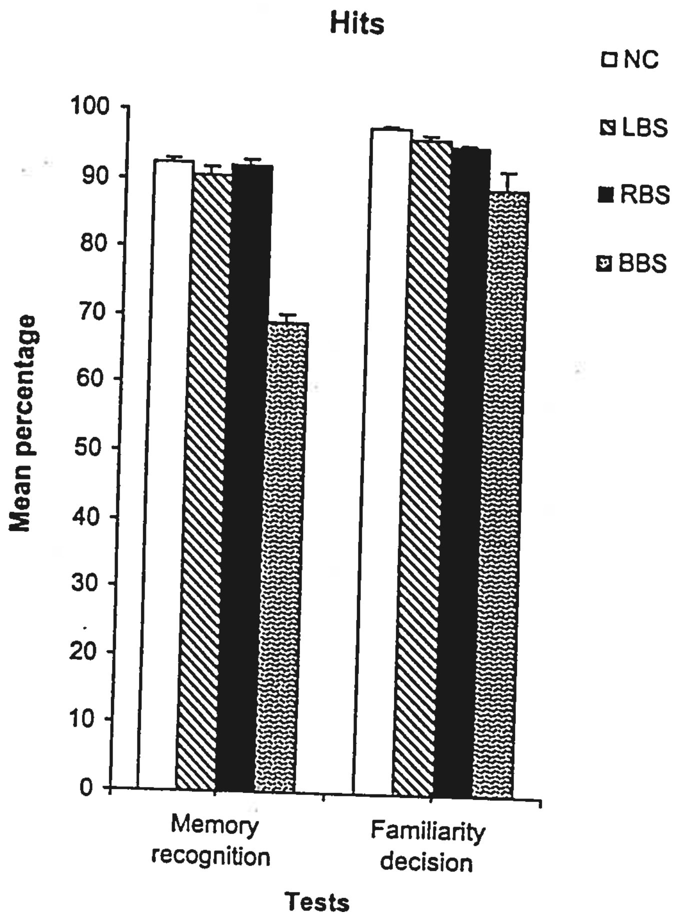
B

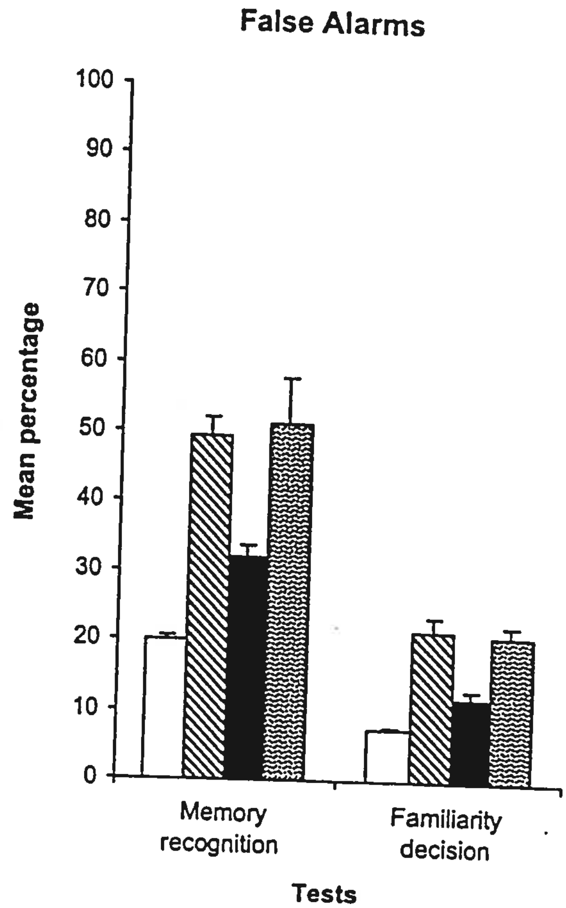
C

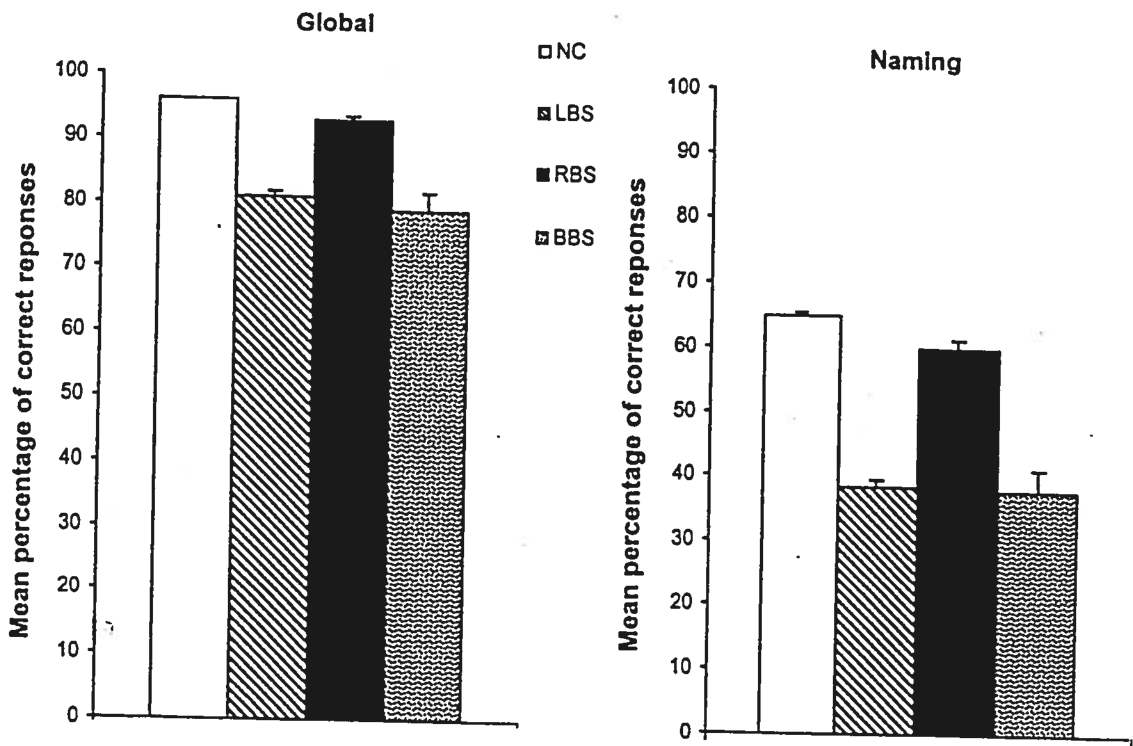
D

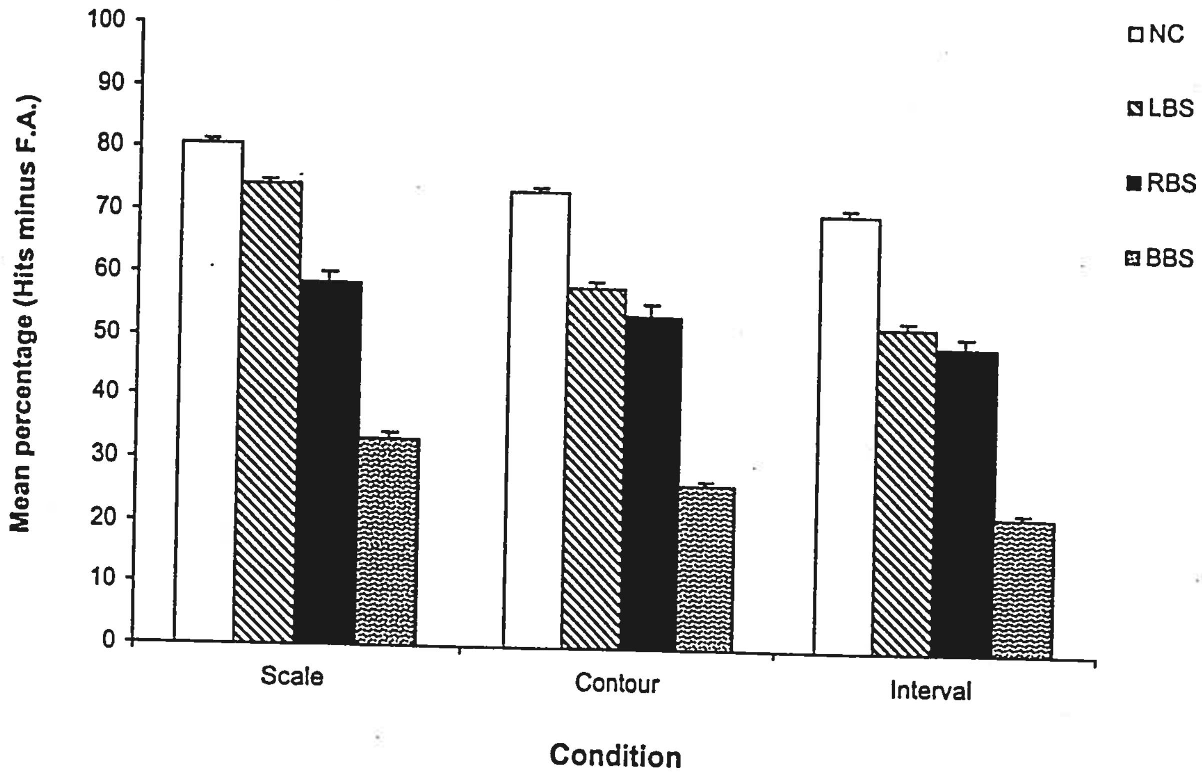
E

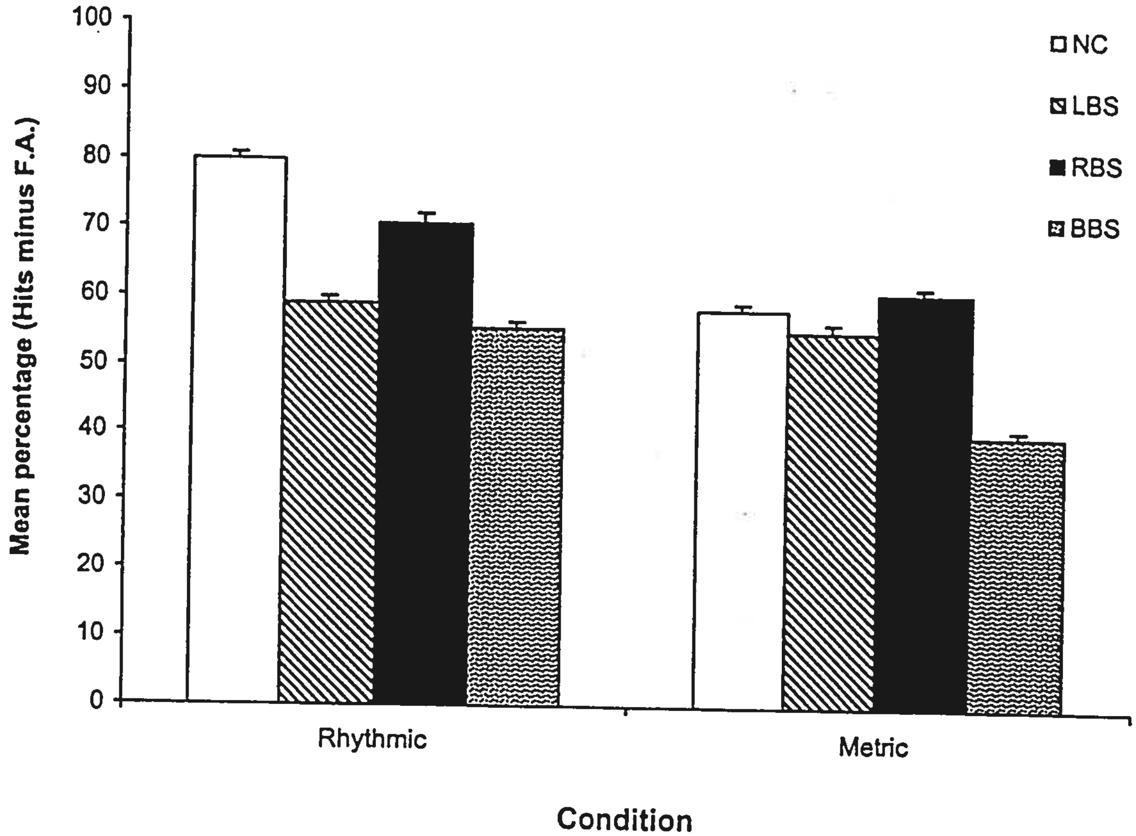
F

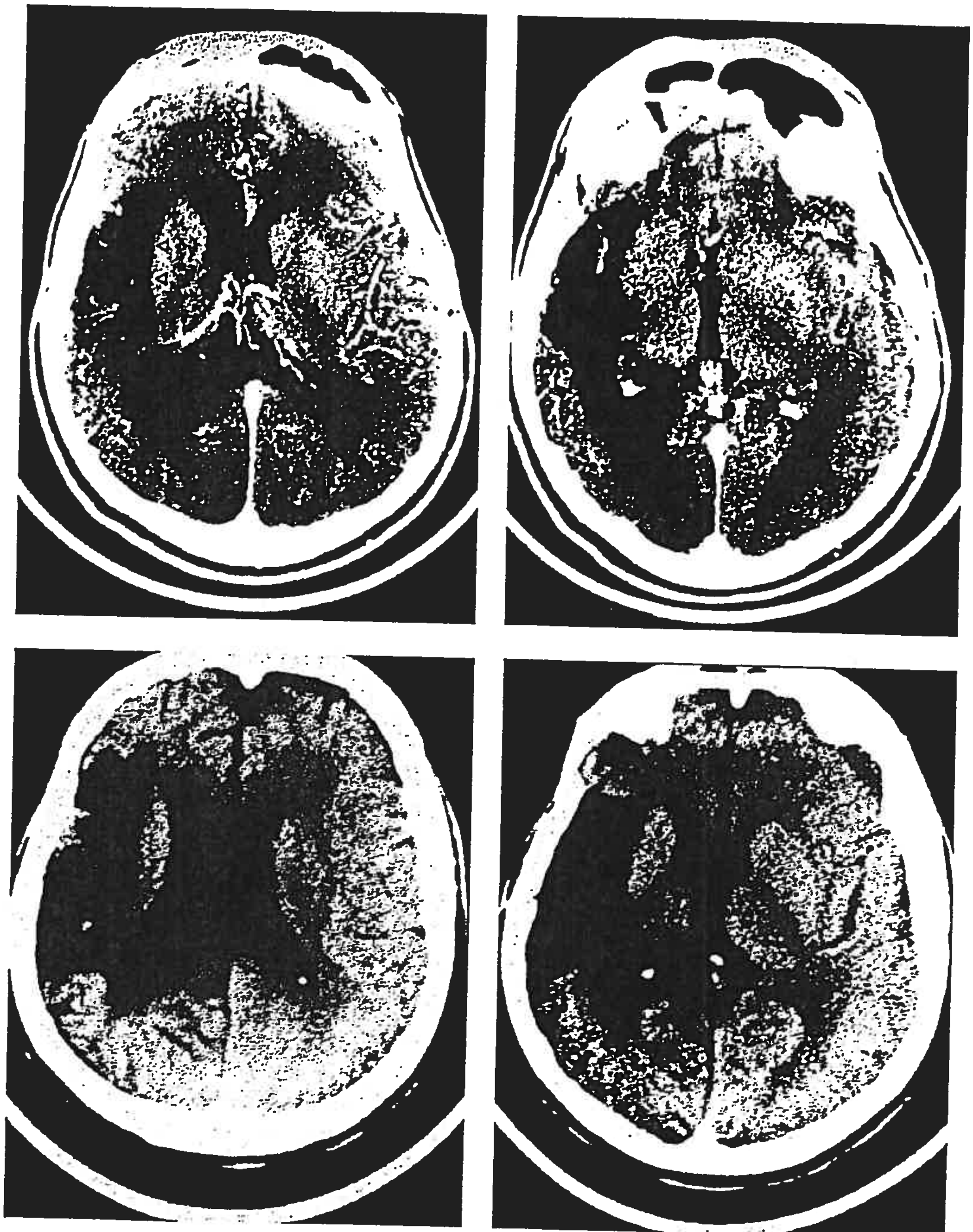

Hits
NC
LBS
RBS
BBS
Mean percentage
100
90
80
70
60
50
40
30
20
10
0
Memory recognition
Familiarity decision
Tests


False Alarms
Mean percentage
100
90
80
70
60
50
40
30
20
10
0
Memory recognition
Familiarity decision
Tests


Global
NC
LBS
RBS
BBS
Mean percentage of correct reponses
100
90
80
70
60
50
40
30
20
10
0
Naming
Mean percentage of correct reponses
100
90
80
70
60
50
40
30
20
10
0


100
90
80
70
60
50
40
30
20
10
0
Mean percentage (Hits minus F.A.)
NC
LBS
RBS
BBS
Scale
Contour
Interval
Condition


100
90
80
70
60
50
40
30
20
10
0
Mean percentage (Hits minus F.A.)
NC
LBS
RBS
BBS
Rhythmic
Metric
Condition



DEUXIEME ARTICLE: CONGENITAL AMUSIA :
A GROUP STUDY OF ADULTS AFFLICTED WITH A MUSIC-SPECIFIC DISORDER

Congenital Amusia :
A Group Study of Adults Afflicted With a Music-Specific Disorder

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running head: congenital amusia

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Abstract

The condition of congenital amusia, commonly known as tone-deafness, has been described for more than a century but has received little empirical attention. In the present study, a research effort has been made to document in detail the behavioural manifestations of congenital amusia. A group of 11 adults, responding to stringent criteria of musical disabilities, were examined in a series of tests originally designed to assess the presence and the specificity of musical disorders in brain-damaged patients. The results show that congenital amusia is related to severe deficiencies in processing pitch variations. The deficit extends to impairments in music memory and recognition as well as in singing and the ability to tap in time to music. Interestingly, the disorder appears specific to the musical domain. Congenital amusic individuals process and recognise speech, including speech prosody, common environmental sounds and human voices as well as control subjects. Thus, the present study convincingly demonstrates the existence of congenital amusia as a new class of learning disabilities that affect musical abilities.

Key words: Congenital amusia, tone deafness, auditory disorder, learning disabilities, music.

Language and music have many similarities. Notably, language and music are universal and specific to humans. Despite the complex abilities involved in both domains, linguistic and musical competence develop in the child spontaneously, without conscious effort or formal instruction. However, a few individuals suffer from severe language acquisition impairments, which are not consequent to any hearing deficiency, mental retardation or lack of environmental stimulation (e.g. Benton, 1964; Gopnik & Crago, 1991). Such a specific language impairment affects between 3 and 6% of the population (e.g. Wright et al., 1997). Considering the similarities between music and language, we can expect that a similar proportion of individuals from the general population experience music specific impairments. Affected individuals would be born without the essential wiring elements for the development of a normally functioning system for music. This condition is variously termed tune-deafness, tone-deafness, dysmelodia or dysmusia in the literature. However, we prefer to refer to this learning disability with the less restrictive label of "congenital amusia", because there may be as many different forms of developmental amusias as there are varieties of acquired amusias that result from accidental brain damage in adulthood.

Congenital amusia is a condition that has been known for more than a century, since the pioneering study published by Grant-Allen in 1878. Grant-Allen reports the case of a 30-year-old man with a solid education and without neurological lesion, who suffers from a severe musical handicap. The man was unable to discriminate the pitch of two successive tones, failed to recognise familiar melodies, and could not carry a tune. He exhibited an overall indifference towards music. Yet, the musical defect could not be explained by a lack of exposure to music since the man had received musical lessons during childhood. A century later, in 1984, Geshwind published a similar case. The case was a man who came from a musically impaired family, despite their frequent exposure to recorded music at home. As a child, this man attempted piano lessons, but his teacher soon realised that he could not sing, nor discriminate between two pitches, and could not keep time. Interestingly, this same subject could fluently speak three foreign languages.

Though indicative, these two studies are anecdotal since they are descriptive and not supported by systematic evaluations. Two large-scale studies were run to quantify the musical disorder. In 1948, Fry evaluated a 1200 subject sample on tests requiring to compare two notes or two musical phrases in order to detect a change in pitch. From these results, Fry estimated that 5% of the British population were amusic. This author further argued that musical memory problems as well as a difficulty in pitch discrimination might be the major determinants of congenital amusia. However, these claims were not supported by data analyses. More recently, Kalmus and Fry (1980) ran another large-scale study with 604 unselected adults who were required to detect anomalous pitches inserted in melodies. From these results, 4.2% of the British adult population were estimated to be amusic. However, this estimate is problematic. First, the measure lacks sensitivity since more than 90% of the participants were performing at ceiling. Secondly, a single measure of musical ability may have both poor validity and poor reliability. Above all, such a psychometric definition of congenital amusia is unconvincing because the sole consideration of one tail of the normal distribution on a single test does not provide convincing evidence that congenital amusia is a real affliction and not a statistical anomaly.

In summary, previous studies of congenital amusia provide valuable information regarding the nature of congenital amusia, while they do not offer a solid empirical basis. Thus, the major objective of the present study is to document the probable existence of congenital amusia by the systematic evaluation of individuals who, despite normal exposure to music and a high level of general education, failed to develop basic musical skills. These basic musical abilities rest on core mechanisms that are assumed to be shared by all members of a given society, musicians and nonmusicians alike, and that allow humans to appreciate and respond to the music of their culture (Peretz, 2001). At the very least, these skills should encompass the ability to discriminate and recognise the music of the environment and, above all, to respond to it emotionally. Moreover, within limits, all humans should be able to carry a tune and to synchronise with the musical behaviour of the others, by tapping along with the music and by dancing, for example. These are the basic musical abilities that will be assessed experimentally in the present study.

Based on the published findings, we expect amusic participants to show a particular deficit in discriminating musical pitch variations and in recognizing familiar melodies. As a consequence of this receptive disorders, we also expect amusic individuals to have poor singing abilities. In contrast, we have no particular predictions regarding their competence in monitoring rhythmic structure in music. Therefore, amusic subjects will be tested in various tests tapping mostly pitch-related abilities but also rhythmic ones for comparison.

In order to assess the domain specificity of the auditory disorder exhibited by congenital amusic subjects, their ability to recognise and memorise music was compared to their ability to recognise and memorise other familiar auditory tokens, such as spoken lyrics, speakers' voices, and animal cries, under identical testing conditions.

Case description

The most challenging part of the present study was to set the criteria that would allow classification of individuals as congenitally amusic, and to find appropriate means to discover them. In what follows, we will describe a) the procedures and the inclusion criteria used to identify amusic cases; b) a summary of the behavioural assessment and self description of the amusic participants.

Recruitment of amusic subjects

Various procedures were used, all requiring self-declaration of a handicap for music. The most effective mean consists of making announcements in the media (radio, newspapers, university local papers and vocal recording machines). However, self-declaration does not suffice. Nonmusicians are prone to complain about their musical deficiencies, in general. To exclude these false alarms as well as borderline cases, we used a detailed questionnaire and focussed our attention to individuals whose self-description was as close as possible to the case reports of Grant-Allen (1878) and Geshwind (1984). Out of more than 100 interviews, we selected 37 individuals and tested them in the laboratory on a musical screening battery, to be described below. Out of these 37 potential subjects, 22 exhibited a pattern of performance that unambiguously indicated the presence of a receptive musical disorder. However, only 11 of them were either willing to participate to further evaluations or were eligible because their past history fitted with the following criteria:

- a high level of education, preferably university level, to exclude general learning disabilities or retardation
- music lessons during childhood, to ensure exposure to music in a timely fashion.
- a history of musical failures that goes back as far as one can remember, to increase the likelihood that the disorder is inborn
- no previous neurological or psychiatric history to eliminate an obvious neuro-affective cause

Assessment and description of the amusic group

In order to verify the presence of a deficit in music perception and memory, all self-declared amusic subjects who corresponded to our set of stringent criteria were tested over a battery of tests designed to diagnose musical deficits in brain-damaged patients of variable age and education level. This battery contains six subtests, in which the material is conventional and kept as constant as possible across conditions. Each subtest measures the use of a musical characteristic that is known to contribute to music perception and memory. These are scale, interval and contour information on the melodic organisation dimension, and rhythm and meter on the temporal organisation dimension. The sixth subtest of the battery probes memory recognition abilities. The melodic discrimination subtests as well as the rhythmic test, all require a "same- different" classification task. The metric test requires a waltz-march classification of each musical excerpt. Finally, the memory recognition subtest appears as an incidental memory test at the end of the evaluation, by asking subjects to judge whether or not they had heard the musical selections in the previous subtests. All stimuli are computer-generated and delivered with a piano sound (see Liégeois-Chauvel et al., 1998, for a full description).

The musical battery has been used extensively in our laboratory and, above all, has been shown to be effective in identifying adult nonmusicians with deficits in either the melodic or rhythmic dimension (Ayotte et al., 2000; Liégeois-Chauvel et al., 1998; Peretz, 1990; Peretz et al., 1994; Peretz et al., 1997). The conservative cut-off score of three standard deviations below the mean obtained by control subjects is used to indicate the presence of a deficit. Control subjects are 61 nonmusicians (Age range: 14-74 yrs. Range in years of education: 7-20 yrs), who have no known music impairment and who were individually tested in our laboratory as neurologically intact controls for brain-damaged patients.

(Insert Table 1 about here)

The performance of the eleven amusic subjects on the battery is expressed in percentages of correct responses and is presented for each individual in Table 1. When the score is below the cut-off score for a particular subtest, it is marked by an asterisk. As can be seen, all amusic subjects perform three standard deviations under the mean of the 61 controls in at least two out of the six subtests. More importantly, all amusic participants fail in at least two of the three subtests involving discrimination of pitch modifications (see scale, contour and interval subtest). Performance on the rhythm subtest is more variable, with about half the subjects showing a deficit. None of the amusic participants scores below the cut-off score for the « metric » task, probably because this task was relatively difficult[1] for a few control subjects as well. Finally, eight amusic participants also suffer from a severe difficulty in memory as indicated by their impaired performance on the incidental memory recognition test.

Thus, the presence of a musical pitch discrimination deficit is clearly supported by the results. However, it is worth mentioning that no single subtest of the battery can be used to discriminate amusics from controls since, in each subtest, a few amusic subjects managed to perform in the low but normal range.

The amusic group is composed of 9 women and 2 men, are French speaking to the exception of one English-speaking participant. The mean age is 57 years and the mean level of education is 17 years. The higher proportion of women and of older people is probably not related to the condition of congenital amusia but rather reflects the general characteristics of educated volunteers. One drawback from this sample is that many elderly individuals suffer from hearing problems. To assess the presence and importance of a hearing impairment, each subject underwent standard audiometric testing with a recently calibrated Beltone 9D apparatus. A summary of the outcome is presented in Table 1 along with the characteristics of each subject. When there is an impairment, the loss is expressed in dB HL for the left and right ear, respectively. As is often the case with ageing, most hearing losses are confined to the high frequency range. However, two amusic participants (A2 and A11) have a more important loss. The loss was not congenital but acquired as a result of regular exposure to loud noise during adulthood in one case (A2) and due to successive ear infections in childhood for another (A11).

However, the hearing impairment and the maturity of the amusic participants have no apparent influence on their general intellectual and memory functioning, as attested by their homogeneously high scores on standardised batteries (see Table 1). They score above average in the WAIS-III scale (Wechsler intelligence scale III) and the Wechsler Memory scale III. Moreover, amusic participants do not report any other learning disability than for music.

According to their self-report, all amusic participants mentioned their inability to discern wrong notes in a musical passage and to sing in tune. These reports rely on peer accounts because the amusic participants are unable to perceive their own impairments. A majority (7) reported a difficulty in recognising musical melodies without lyrics and in dancing (8 people). Most (7) do not appreciate music and two subjects even find music unpleasant and try to avoid it. While all amusic participants affirmed having these difficulties as far back as they could remember, many subjects realised the scope of their problem during music classes at school. Six amusic subjects also mentioned that one of their parents (most often the mother) and certain siblings had musical problems as well. However, we found members in each family who were not affected, thus discarding a familial negative attitude toward music as an explanatory factor.

General method

Matched control subjects:

A group of 20 persons matched to each amusic subject in gender, age, education and musical background served as matched control participants. Sixteen control subjects were raised in the francophone culture of Quebec while 4 controls were raised in the English culture of North America. Amusic and control participants were naive with regard to behavioural testing and were seen at the same pace. Although controls were not actively involved in music, none reported problems in the musical domain and indeed none was exhibiting a deficit in the screening musical battery (see Table 1). A history of alcohol abuse, psychiatric disorder or other neurological illness was ground for exclusion. All controls were right-handed and suffered from a hearing problem in the same proportion as the amusic participants. One control subject suffered from a hearing loss that spanned the whole frequency range as did A2 (see Table 1) and four controls had a hearing loss in the high frequency range, as do amusic participants. All subject's informed consent was obtained to participate in this project, which was approved by the ethical committee of the Institut universitaire de Gériatrie de Montreal.

Material and procedures

Most tests that are used to study the amusic subjects and their matched controls have been previously designed for and validated with brain-damaged nonmusicians who suffered from musical impairments as a result of brain damage. Therefore, a detailed description of the tests can be found in prior published papers. The references will be provided below in the corresponding section; accordingly, only the methodological aspects that are relevant to the understanding of the testing situation will be specified.

Stimuli are pre-recorded and delivered from a DAT Sony recorder to the subject via two loudspeakers that are set to a volume that is confortable to the subject. The subject is individually tested in our laboratory and provides his/her judgements on answer sheets. Each subject was tested in at least three sessions, each lasting about two hours, with as many pauses as requested. The order of tests was identical for all subjects who started with the musical screening battery described previously. They were then tested with the three types of memory recognition tests (presented in part 2) followed by the musical pitch perception tests (presented in part 1). The production tasks (presented in part 3) were administered at the end.

In the following sections, the results will be presented along with the corresponding test material and procedure. Test results will be grouped in three different parts, each devoted to the behavioural assessment of a particular question regarding the functioning of amusic subjects.

Part 1. Musical pitch perception

1.1. Anomalous pitch detection task

All amusic cases reported in the literature experience a marked deficit in discerning pitch differences. This suggestion finds support in the present study since all 11 amusic participants failed in at least two out of the three screening tests that probed their ability to discriminate melodies on the pitch dimension. The results were obtained with unfamiliar melodies in a "same-different" classification task whereas the proportion of congenital amusics in the British population was estimated with an anomalous pitch detection task and familiar melodies (Kalmus and Fry, 1980). Thus, it was deemed appropriate to test the present sample of amusic individuals with a similar test. Moreover, a variant of the anomalous pitch detection task has been recently used in a twin study and shown to tap an ability that is genetically determined (Drayna et al., 2001). In order to facilitate comparisons across studies and to further test the musical pitch defect characterising congenital amusia, we examined here the ability of our amusic group to detect a pitch anomaly in both familiar and unfamiliar melodies.

Subjects were presented with two sets of melodies. The first set comprises only familiar melodies and the second set, only unfamiliar ones. The familiar melodies were different for the French- and English-speaking participants. The "French" version was constructed with 36 melodies (ranging in length from 6 to 15 notes) that are well-known to Quebec French-speakers (Peretz et al., 1995); the test has been used in a prior study on acquired amusia (Peretz & Gagnon, 1999). The "English" version consisted of 30 familiar melodies (ranging in length from 7 to 14 notes) selected from a list of musical pieces that are well known to North-America English-speakers (Steinke et al., 2001). The unfamiliar set corresponded to the 30 comparison melodies used in the "scale" subtest of the musical battery. In each set, half the melodies were modified by shifting the pitch of one note by one semitone higher or lower so that the note fell out of key while it preserved the original contour. The position of the modified note varied across melodies, avoiding the first and last note positions. Each melody was presented once. After each presentation, subjects were asked to judge whether the melody contained a "wrong note" or not.

The responses were considered as hits when the subjects responded "yes" to a melody containing an anomalous pitch and as false alarms when responding "yes" to an intact melody. The results are presented in Figure 1. As can be seen, amusic subjects are performing close to chance and well below their matched controls. There is no overlap between the amusic and control distribution. The mean percentages of hits minus false alarms were submitted to an ANOVA with Familiarity (familiar versus unfamiliar melodies) taken as the within-subjects factor and Groups (amusics versus controls) as the between-subjects factor. As expected, a highly significant Group effect is obtained, with ($F(1,28)=162.89$, $P < .001$, due to the clear deficit exhibited by amusics compared to controls. A main effect of Familiarity was also observed ($F(1,28)=35.34$, $P < .001$), reflecting the higher performance observed for the familiar over the unfamiliar melodies. There was no interaction with the Group factor ($F<1$).

[Insert Figure 1 about here]

The results obtained with this anomalous pitch detection test are key for several reasons. From a practical perspective, this test clearly distinguishes amusic subjects from normal individuals, and hence may serve as a diagnostic tool in the future. The results are theoretically important because they converge with prior findings in identifying a deficiency in musical pitch perception in congenital amusia. Such a pitch defect is the most likely origin of the musical disorder, as it will be argued further below and in the general discussion.

1.2. Sensitivity to dissonance

Another striking experience of musical pitch perception occurs when several tones sound together resulting in a harmonious blending effect. This pleasant experience is mainly determined by the ratio between the constituent frequencies. When the ratio is simple, such as between two tones lying an octave apart, the combination is considered consonant and pleasant for most listeners, including infants (e.g. Zentner & Kagan, 1996). When the ratio is complex, such as between two tones that lie a semitone apart, the resulting combination is perceived as dissonant and unpleasant by the vast majority of listeners, from an early age (see Schellenberg & Trehub, 1996, for a review). Thus, sensitivity to dissonance is a fundamental experience of music that is tightly related to the ability to perceive fine-grained pitch differences. If the latter ability is impaired, as is apparently the case in congenital amusia, then one can predict that amusic individuals will be indifferent to the presence of dissonance. The test of this particular prediction was the goal of the present investigation.

In order to assess sensitivity to dissonance, we used pleasantness judgements for a set of 24 musical excerpts that were presented in two versions (see Peretz et al., 2001, for a full description). In the original version, the excerpts that comprise a melody and an accompaniment, are highly consonant and are taken from pre-existing classical music (e.g., the first measures of Albinoni's adagio). The dissonant version was created by shifting the pitch of all tones of the leading voice by one semitone either upward or downward. Each excerpt was presented in their consonant version and in their new dissonant version, amounting to 48 trials. The task of the subject was to judge the pleasantness of each excerpt on a 10-point scale, with 1 referring to very unpleasant and 10, to very pleasant. Since half the excerpts evoke a sense of happiness (they were all played in the major mode with a median tempo of 138) and the other half, a sense of sadness (they were played in the minor mode at a median tempo of 53), these were presented in their consonant version in a separate control task. The task was to judge on a similar 10-point scale, if each excerpt was sad (corresponding to a rating of 1) or happy (corresponding to 10). In this happy-sad distinction task, the pitch intervals (defining the mode) was not the only cue that the subject could use to recognise the emotion, the tempo was also available. To the extent that amusic subjects can derive the tempo of music, they should be able to attribute the correct emotional label to the 24 original excerpts.

The mean pleasantness ratings given by the amusic and control subjects to the consonant and dissonant version of the same excerpts are presented in Figure 2 (left panel). As shown, control subjects judge the consonant versions as generally pleasant whereas they judge the dissonant versions as unpleasant. This distinction does not emerge in the ratings of the amusic subjects[2] who tend to judge all excerpts as weakly pleasant. The ratings were submitted to an ANOVA considering Dissonance (consonant versus dissonant version) as the within-subjects factor and Group as the between-subjects factor. The analysis gives rise to a highly significant interaction between Dissonance and Group, with $F(1,24)=52.37$, $P < .001$). The interaction supports the observation that control subjects find the consonant versions much more pleasant than the dissonant versions of the same excerpts ($t_{15}= 12.456$; $P < .001$) whereas the amusic group shows a less marked preference ($t_9= 2.123$; $P < .05$). Thus, amusic subjects appear less sensitive to dissonance than normal controls.

(Insert Figure 2 about here)

However, amusic subjects are able to recognise the affective tone of music to some extent. They are able to distinguish happy from sad music reliably (see Figure 2, right panel; $t_9=9.692$; $<.001$), although their judgements are less extreme than those emitted by their controls. This difference is supported by a significant interaction between Structure (major/fast excerpts versus minor/slow excerpts) and Group, with ($F(1,23)=5.78$, $P < .025$).

Altogether the results obtained with emotional judgements are consistent with the results obtained in non-emotional tasks. Amusic subjects are unable to perceive and interpret musical pitch differences normally. Yet, this is not the result of poor auditory attention or of a deficient affective system in general, since they are able to infer the happy or sad tone of music in a rather consistent manner.

1.3. Discrimination of pitch variations (intonation) in speech

Fine-grained discrimination of pitch is probably more relevant to music processing than to any other domain. Speech intonation contours, for example, use variations in pitch that are larger than half an octave, to convey relevant information. In contrast, melodies mostly use small pitch intervals (of the order of a $1/12^{th}$ or $1/6^{th}$ of an octave, which correspond to the semitone and whole tone, respectively). Therefore, a degraded pitch perception system may compromise music perception but leave speech prosody unaffected.

To assess this hypothesis regarding the domain-specificity of the pitch defect experienced by amusic subjects, we exploited once again experimental tests that have been previously used with brain-damaged amusic patients (Patel et al., 1998). These tests are constructed by computer-editing two basic sets of sentences so that they only differ from each other by local pitch changes. In the first set, the change affects the last word by marking a rise in pitch so as to indicate a question (e.g. He speaks French?) or a falling pitch (e.g., He speaks French.) to indicate a statement. The pitch change is in the order of 6 to 7 semitones (range : 3-11) for questions in French and English, respectively. In the statements, the pitch fall is of 2 to 3 semitones. In the second set of sentences, the pitch difference (of an average magnitude of 8 semitones) affects an internal word of the sentence to mark emphatic stress like in « SING now please ! » (Capitals indicate the stressed word) and « Sing NOW please ! ». Both sets were naturally spoken by a native female speaker and exist in French and English; each set comprises 15 or 16 pairs of sentences, depending on language.

The sentences were first presented in isolation and subjects judged whether the sentence indicates a statement or a question for the first set of sentences differing by their final pitch rise or they indicated which word bore the stress for the second set of sentences. These two tasks were relatively easy to complete for both amusics and controls who obtain 94 and 98 % correct, respectively, in the final pitch change condition, and 87 and 90%, respectively, in the internal pitch change condition.

The same sets of sentences have also been presented by pairs in a "same-different" classification task. For example, when subjects were presented with the pair " He speaks French? " followed by "He speaks French. " they were expected to respond "different". The only cue available for the discrimination was the presence or absence of a pitch change on the final word in 32 trials (30 in English) and in the location of the internal pitch change in 32 additional trials (30 in English). An equal proportion of trials contained no change. No feedback was provided to the subject. Again, amusic subjects performed slightly lower than the control subjects (see left panel in Figure 3) but not significantly so ($F(1,28)= 1.41$). Thus, amusic subjects do not seem to be impaired in processing speech intonation, even when these prosodic variations are limited to pitch changes as studied here.
(Insert Figure 3 about here)

When all linguistic information is removed from the sentences by a process of computer analysis and synthesis (see Patel et al., 1998, for details about the edition of these non-speech analogs), a different picture emerges. With the non-speech derivations of the sentences, amusic subjects show evidence of a deficit compared to normal controls, as can be seen in the right panel of Figure 3. Yet, the only difference between the speech results (left panel) and the nonspeech analogs (right panel) lies in the acoustic waveform of the stimuli, not the size of the pitch differences. This difference significantly affected amusics' performance, as confirmed by an overall ANOVA taking Material (speech versus nonspeech), and Type of pitch change (final versus internal pitch change) as the within-subjects factors and Groups as the between-subjects factor. The interaction between all three factor reached significance ($F(1,28)=5.04$, $P < .033$). This was due to the lower performance of the amusic subjects in the nonspeech condition relative to normal controls ($F(1,28)=12.88$; $P < .001$). Non-parametric tests (by way of Mann-Whitney U) yield similar results.

One can conclude from this set of tests that congenital amusia does not compromise interpretation and discrimination of speech intonation. However, this spared area of performance is contingent upon the presence of linguistic information. When linguistic cues are removed from the signal, amusic subjects exhibit depressed performance. The latter finding suggests that amusics' pitch defect is not limited to music but can extend to other auditory patterns varying on the pitch dimension, provided that amusics cannot use speech cues to support discrimination.

Part 2. Specificity of the musical disorder

The tests using intonation patterns in speech suggest that amusic subjects may have difficulties in the processing of auditory patterns other than musical ones. Yet, following the literature as well as amusics' self reports, the disorder seems limited to the musical domain. In order to delineate the auditory domains in which amusic subjects seem to be at a disadvantage compared to normals, they were tested here with auditory meaningful stimuli pertaining to speech, human voices and environmental sounds. Care was taken to make cross-domain comparisons in identical experimental conditions so as to minimise the contribution of other factors to the tasks. In doing so, we heavily relied on prior work that succeeded to demonstrate the domain-specificity of the musical impairments observed in brain-damaged patients (see, in particular, Ayotte et al., 2000; Peretz et al., 1994; Peretz, 1996; Peretz et al., 1997).

2.1. Naming and recognition of tunes, lyrics, voices and environmental sounds

The identification of a familiar auditory pattern by name is one of the most demanding tasks, particularly for nonverbal material. Yet, it is one of the rare tasks that allows the rapid assessment of the integrity of a processing system from the analysis of the acoustic input up to the name retrieval via appropriate contacts in memory. This is why identification tasks are regularly used in neuropsychological settings. In addition, this explains why we administered this test first so as to assess the domain-specificity of the difficulties experienced by congenital amusics.

Subjects were presented with auditory stimuli coming from four different domains, but blocked by domain. The musical block consisted of 52 melodies (without lyrics) coming from familiar folk songs (Peretz et al., 1995, Steinke et al., 2001) and presented one at the time. After each presentation, the subjects were requested to name the tune. In case of failure, they were presented with four written titles among which to choose; the foils were of the same genre (for example, all titles would be Christmas songs). The lyrics block comprised 25 spoken lyrics coming from the same pool of well-known songs. However, care was taken to select lyrics in which content words could not cue the title (For example, "prête-moi ta plume pour écrire un mot" for the song name "Au clair de la lune" or "for nobody else gave me a thrill" of the song name "Had to be you"). In case of failure, they were presented with 4 written titles semantically related to the lyrics excerpt. The voice block involved 33 speech excerpts pronounced by famous public figures for Quebec residents. The stimuli were edited so as to remove any context word that could cue the speaker's professional activities. Three choices of names of the same sex and age were presented in case of a naming failure. Finally, 45 environmental sounds, including animal cries, transportation noises, human noises and indoor noises, were presented in a block. In case of a naming failure, subject could choose between four pictures of the same category (for example, all pictures would be transportation means).

(Insert Figure 4 about here)

The mean percentages of correct naming scores and global scores, that adds correct naming to correct name choices, are presented in Figure 4, for each domain. The naming and global scores are highly similar. By both scoring procedures, amusic subjects are disproportionately impaired in music identification relative to the other domains and to control performance. To the exception of one outlier, all amusics were able to retrieve the name of the song corresponding to the spoken lyrics. This finding is important because it shows that amusics have learned the songs although the musical part is problematic for them.

The outcome of the ANOVAs performed separately on the naming and global scores confirms these observations. Each ANOVA was computed with Material (tunes, lyrics, voices and environmental sounds) as within-subjects factor and Group (amusics and controls) as the between-subjects factor[1]. An interaction was obtained between Material and Group ($F(3,69)= 17.26$ and12.23, $P < .001$, on global and naming scores, respectively). The interaction was due to the fact that the amusic group only performed below the control group in the music identification test ($t_{29}=8.526$ and 6.609, $P < .001$, for the naming and global scores, respectively). No difference was observed between groups for the other materials (with $t_{29}=0.551$ and 1.494 for lyrics, N.S., $t_{24}= 0.019$ and 0.982 for human voices, N.S. and $t_{23}= 0.203$ and 0.073 for environmental sounds, N.S., for the naming and global scores, respectively). The non-parametric statistical analyses (Mann-Whitney U) yielded the same results.

The results confirm the self-report of amusic subjects in showing a selective problem in recognising the melodic part of songs. Otherwise, the amusic subjects appear to recognise lyrics, speaker's voice and environmental sounds as easily as everybody else. The disorder appears music-specific.

2.2. Separate recognition of tunes and lyrics from songs

An important result in the previous identification tests was to find a clear dissociation in all but one amusic subjects between the recognition of lyrics, which was intact, and the recognition of tunes, which was impaired, although both components were learned together in songs. To assess this particular dissociation further in a less demanding task, we used here a binary familiarity decision. Requiring a feeling of knowing without identification may reveal residual recognition abilities. To this aim, a familiarity decision task was devised for tunes and lyrics, separately. Half the stimuli were coming from familiar songs and half were unfamiliar. The unfamiliar melodies were taken from the same repertoire of folk songs but are unknown to the subjects because they are no longer sung (see Peretz, Gaudreau & Bonnel, 1998, for details about stimuli selection). The task is simply to indicate whether or not each song part sounds familiar.

(Insert Figure 5 about here)

The responses were considered as hits when the subjects responded « familiar » to familiar excerpts and as false alarms when responding « familiar » to unfamiliar excerpts. The mean percentages of hits minus false alarms obtained by each group in the two familiarity decision tests are presented in Figure 5 (the negative values indicate a greater number of false alarms than of hits). The data were submitted to an ANOVA with Material (melodies versus lyrics) as the within-subjects factors and Groups as the between-subjects factor[2]. An interaction between Group and Test was obtained ($F(1,24)=11.05$, $P < .003$). The amusic group was again only impaired in the melody condition relatively to the control group ($t_{24}=4.603$; $P < .001$). Amusics did not perform significantly below normals in the lyric condition (N.S.), despite the presence of a very poor score obtained by the same outlier as previously. The same results were obtained by way of Mann-Whitney U tests.

2.3. Memory recognition for tunes, lyrics and environmental sounds

One important question regarding the amusics' failure to recognise familiar melodies is to what extent they are able to relearn these melodies in the laboratory. The results obtained in the screening battery indicate an overall deficit in the memorisation of novel music (see Table 1). However, in this memory recognition test, memorisation of melodies was incidental. Incidental encoding of music may not reflect the optimal performance of amusic subjects since they were not fully engaged in memorising the material. Thus, it remains possible that amusics are able to memorise melodies if they are explicitly told to do so. We also need to assess whether memorisation of familiar music is possible. Although the amusic group has difficulties in recognising music as familiar, they might remember it nonetheless. For example, when presented with the music of "La vie en rose", they might recognise it as music which they had heard in the laboratory, without recognising it as familiar. Evaluation of these possibilities was the goal of the following experiment.

To test memory, we used the same recognition memory test with three different materials as previously validated with brain-damaged patients (Peretz, 1996; Peretz et al., 1997). In the presentation phase, 20 auditory targets are presented one after the other. The subject's task is to memorise each of them. After a short pause, the test phase occurs during which the 20 targets are randomly mixed with 20 lures; the subjects' task is to tell for each stimulus whether or not they have heard it in the study set. No feedback is provided to the subject. In one form of the test, all stimuli consist of familiar tunes. Subjects performed the same task for two non-musical materials, one including lyrics and one involving environmental sounds, similar to the ones used in the previous tests of identification. The three memory recognition tests were performed in different testing sessions.

(Insert Figure 6 about here)

The responses were considered as hits when the subjects responded "yes" to studied stimuli, and as false alarms when responding "yes" to nonstudied stimuli. The mean percentages of hits minus false alarms obtained by each group in the three memory recognition tests are presented in Figure 6. These scores were submitted to an ANOVA with Material (melodies, lyrics, environmental sounds) taken as the within-subjects factor and Group as the between-subjects factor. As shown in Figure 6, the amusic group scored significantly below the control group on the tune recognition test (t_{29}=5.994; $P < .001$) but not so on lyrics (t_{29}=0.502; N.S.) and environmental sounds (t_{29}=1.541; N.S.). The selectivity of the impairment to the musical domain was supported by the presence of an interaction between Material and Group ($F(2,58)=30.86$, $P < .001$).

To conclude, congenital amusics perform poorly in all tasks requiring recognition and memory of melodies. The musical deficit is apparent in all tasks, from the most difficult situation that requires naming a well-known tune to the least demanding task requiring a familiarity decision. Even re-learning is unsuccessful, when provided with a single study episode (i.e. in the last condition referred to as the memory recognition condition). The performance of the amusic subjects is low but variable; yet, it is remarkably consistent across the three recognition tasks involving familiar melodies.

Pearson correlations (with n=10) are .90 between the scores obtained in identification (global score) and familiarity, .77 between familiarity and memory recognition, and finally, .79 between identification and memory recognition (all $P < .05$). In contrast, under identical testing conditions, the same amusic subjects show no particular difficulty to identify familiar songs on the basis of their spoken lyrics, or to recognise and memorise nonmusical auditory events such as common environmental sounds and speakers' voices. Clearly, the amusic participants do not suffer from general difficulties in memory or attention in the auditory domain. They suffer from a highly selective memory deficit that seems limited to music.

Part 3. Musical production tasks

Our sample of amusic participants were selected because they report and show evidence of severe perceptual impairments for music. Yet, congenital amusics are notoriously famous for singing out of tune. They are usually detected on this basis. In the case of our sample, the production deficit can be expected to arise as a consequence of their poor perceptual and memory system. To assess this rather obvious prediction in a controlled manner, each amusic subject (as well as their controls) was encouraged to sing three songs in the microphone. To assess their potential to synchronise with music (for dancing, for example), they were also assessed in their ability to tap the beat along three different music while being videotaped.

The ability to carry the tune of « Au clair de la lune", "Frère Jacques", and "Vive le vent", which are highly familiar children songs in French-speaking cultures, was assessed in two conditions. In the first condition, the lyrics were provided in print and the participant had to sing the corresponding tune from memory. Since amusic subjects show poor memory for those tunes, as documented previously, their ability to repeat these same tunes, after the experimenter, was checked in a second condition. The singing production of only seven amusic participants could be recorded (A3 and A6 could not participate and A9 and A11 refused to sing in the repetition condition). Controls were tested in equal number in the same conditions. All amusic and control renditions were recorded on audiocassette and then mixed randomly. There were two cassettes: one containing singing from memory and the other, sung repetition.

The ability to synchronise with music was assessed with the first 30 measures of pre-recorded music. The recordings were selected in three different genres; one piece was classical (Ravel's Bolero; duration: 1min 22s), another was disco (« Stayin'alive » from the BeeGees; duration: 1min 28s) and the third was folk (« reel des soucoupes volantes » from the Bottine Souriante; duration: 1min 11s). Subject were required to tap in time with each piece of music, and to do so as regularly as possible with their dominant hand (so as to avoid complicated or syncopated rhythms). All amusic and control tapping hands were filmed and were then copied in a random order on another cassette.

The three tapes (two audio- and one video- tape) were judged by six judges (four musicians and two nonmusicians) who were blind to the classification of the participants. The judges evaluated each performance on a 10-point scale, where 1 meant « very poor» and 10 meant « very good». They also had to decide if each recording was produced by an amusic or not. The ratings were averaged for the three songs produced by the same subject in the same condition. The agreement between judges was high, since correlations between all pairs of judges were significant in each condition (each r being > .76; $P < .01$). The results are presented in Figure 7. They were assessed by nonparametric Mann-Whitney U tests that confirm the presence of a clear distinction between amusic and control performance in each production condition (U= 82, 63, 80 for singing from memory, repetition and tapping, respectively). Across conditions, amusic renditions were judged to be rather poor compared to controls. Moreover, the judges were generally accurate in their classification of each production as coming from an amusic or nonamusic person. Out of the 74 amusic productions, 42 were correctly attributed to an amusic performer by at least four judges, whereas none of the 87 control productions were judged as such (the difference is significant with $\chi^2 = 63.89$, $P < .001$). However, the classification is not perfect since one amusic individual (A9) managed to produce acceptable performance in each condition, by both scoring procedures.

(Insert Figure 7 about here)

In order to evaluate which musical aspect of the vocal production was most affected in amusic singing, we asked two further musicians to blindly judge the accuracy of the rendition in terms of the pitch variations and the temporal variations, separately. The judges provided their ratings on different 10-point scales (with 1 meaning very poor and 10, very good). The ratings were averaged for the three songs in each singing condition and assessed for consistency by Pearson correlations. The ratings were highly correlated, with r= .94 and .92 for the pitch and time dimension, respectively ($P < .01$). Amusic singing were judged to be more impaired on the pitch dimension (with a mean rating of 3.9 across conditions) than on the temporal dimension (mean rating = 6.4; t_8=6.726, $P < .001$). However, a similar trend also emerged for the control performance, with a better rendition of rhythmic than melodic aspects (with 8.9 and 8.2 mean ratings, respectively, t_9=2.289, $P < .05$).

As expected, amusic singing and tapping performance is impaired as compared to normal performance. The difficulties were judged to mostly affect the accuracy of pitch variations. Yet, the problem was not limited to pitch dimension since the rhythmic aspect of amusic singing was not highly rated and, above all, their tapping performance was generally not well synchronised with the music. Finally, and more surprisingly, one amusic subject was judged to perform normally in these tasks. This spared performance may be genuine, pointing to a nontrivial dissociation between perception and performance. Alternatively, this exceptional performance may simply reflect a lack of sensitivity of the crude measures of performance considered in the present study.

General discussion

This study suggests that congenital amusia is not a myth but a genuine learning disability for music. The systematic evaluation of eleven adults, who reported to be severely handicapped in the musical domain despite their efforts to learn it, largely confirms the presence of a under-developed system for processing music. Amusic individuals are severely impaired in music discrimination and recognition tasks. These impairments cannot be explained by hearing losses, since they all have, or grew up with, a normal audiometry. The musical disorder cannot be explained by a lack of exposure since all amusic participants had music lessons during childhood and were raised in families in which a few siblings are musically normal. Finally, the musical deficit cannot be ascribed to some general cognitive slowing since all amusic participants have reached a high level of education. The musical disorder appears as an accidental disturbance in an otherwise fully normal cognitive and affective system.

One remarkable characteristic of the amusic condition is the selectivity of the disorder. The deficit appears highly specific to the musical domain. Amusic subjects retain the ability to process non-musical material as well as their matched controls. In the present study, amusics were shown to interpret properly intonation in speech, to identify well-known figures from their voice alone, to identify and recognise common environmental sounds, such as animal cries and ringing sounds. To the exception of a single amusic subject, they all identify and recognise familiar songs when hearing the first lyrics. This high level of achievement in the auditory domain stands in sharp contrast with the rather poor level of functioning displayed by amusic individuals in recognising and memorising musical patterns. The disorder appears music-specific.

Having established congenital amusia as a real pathological condition, we should now turn to plausible accounts of the observed deficiency. Presently, we can only offer functional explanations, derived from the present behavioural studies. Although we construe congenital amusia as resulting from a slight disruption in the wiring of the auditory cortex, we are presently unable to support this claim because the search of these neural anomalies will require sophisticated brain imaging studies. In this endeavour, specifying the functional origin of congenital amusia is essential because it may narrow down the possible neural loci to a sizeable set of circuitries that can be further inspected for the presence of an anomaly.

On the basis of the present behavioural results and in line with the literature, we propose that one likely origin for congenital amusia is related to a deficiency in musical pitch recognition. Indeed, all amusic participants score below the normal range in the discrimination of musical stimuli that differ on the pitch dimension while a majority of them succeed to discriminate the same stimuli when these differ in temporal structure. This difficulty in detecting pitch-related changes extends to dissonance for which amusics show little sensitivity. The pitch-related defect also extends to the detection of an anomalous pitch inserted in an otherwise conventional melody. This task is particularly sensitive to the presence of amusia since there was no overlap between the normal variations and the amusic scores. The test is diagnostic in the sense that it provides a behavioural marker of congenital amusia. Interestingly, the detection of an anomalous pitch in conventional melodies is a test that is very similar to the one originally used by Kalmus and Fry (1980) to discover congenital amusics in the general British population. More importantly, the test has been shown to tap an ability that is genetically determined in the general population (Drayna et al., 2001). Therefore, it is tempting to propose that heritability of pitch recognition abilities can also be demonstrated by its deficiency, and that its manifestation is congenital amusia.

It is important to note that the pitch defect of amusic individuals does not seem to compromise music exclusively. The impairment extends to the discrimination of intonation patterns, when all linguistic cues are removed. This observation suggests that the pitch deficiency experienced by amusic individuals is not music-specific but is music-relevant. In effect, as mentioned previously, fine-grained discrimination of pitch is probably more relevant to music than to any other domain, including speech intonation. Music is probably the only domain in which fine-grained pitch discrimination is required for its appreciation. Accordingly, a degraded pitch perception system may compromise music perception but leave other domains, such as speech intonation in which meaningful pitch variations are coarse, relatively unaffected. Yet, the same pitch tracking mechanism may subserve both domains. The validity of this prediction is currently under closer examination in our laboratory by studying single amusic cases in detail. The current evidence is largely consistent with the notion that pitch is essential to music. More specifically, a drastic pitch perception defect has been documented with psychophysical methods in one amusic participant (A1; Peretz et al., submitted). Similarly, a deficit in monitoring pitch changes but not timing changes in speech derivations has been further studied and isolated in another case (A6; Hyde et al., 2001).

However, the fine-grained pitch discrimination disorder is not the only impaired musical ability in congenital amusia. As mentioned above, amusics have memory problems for music, some are even impaired in discriminating melodies by their rhythm, and most have difficulties in keeping time to a musical beat. All these tasks require an accurate representation of musical time, not solely musical pitch. There are several possible explanations for the presence of this myriad of musical deficits. The explanation that we presently favour, but for which we do not have yet empirical support, is that the ensemble of musical deficits are cascade effects of a faulty pitch processing system. That is, fine-grained pitch perception might be an essential component around which the musical system develops in a normal brain.

Taken together, the neuropsychological evaluation of self-declared amusic adults has provided a framework for the diagnosis of congenital amusia and has served to delineate the nature and specificity of the disorder. Although there has been limited explorations into developmental musical disorders in the past, the research enterprise constitutes a rich study avenue. At the very least, continued effort in understanding the causes of congenital amusia should shed light on the question as to whether or not music processing corresponds to a genuine specialisation of the brain. From an educational perspective, knowledge of every aspect of congenital amusia should enrich current view of other forms of learning disabilities such as dysphasia and dyslexia. The broader the context in which learning disabilities are viewed, the more likely we are to understand their causes.

Footnotes

[1]We are currently revising the metric subtest of the battery so that all individuals (without musical impairments) achieve at least 75% correct.

[2] One amusic subject (A6) did not perform this test.

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Table 1. Subjects' characteristics and individual scores for the amusic group

	A1	A2	A3	A4	A5	A6	A7	A8	A9	A10	A11	controls (s.d.)	
												matched	unselected
Sex	F	M	M	F	F	F	F	F	F	F	F	16F 4M	42F 19M
Language	Fr	Fr	Fr	Fr	Fr	E	Fr	Fr	Fr	Fr	Fr	16Fr 4A	57Fr 4A
Age	41	62	57	51	71	69	58	59	53	49	57	60.2 (12.8)	45.3(17.6)
Education	19	16	19	15	17	19	15	16	19	19	17	16.5 (2.2)	13.8 (3.7)
Handedness	A	R	R	R	R	R	R	R	R	R	R	R	59R 1L 1A
I.Q.	111	116	107	100	104	112	117	108	128	110	120	-	-
M.Q.	113	135	112	114	127	130	134	114	137	114	130	-	-
Audiometry (1)													
low frequency (250-500 Hz)	n	15-35	n	n	n	n	n	n	n	n	n		-
middle frequency (1000-2000 Hz)	n	35-50	n	n	n	n	n	n	n	n	15-55		-
high frequency (4000-8000 Hz)	n	50-70	n	n	n	n	60-70	n	70-80	n	25-65		-
Musical battery													
scale	76,7	60*	50*	50*	56,7*	46,7*	63,3*	53,3*	56,7*	53,3*	46,7*	90 (7,8)	91,7 (6,8)
contour	50*	43,3*	50*	70	53,3*	46,7*	80	56,7*	66,7*	53,3*	66,7*	91,5 (6,4)	90,2 (7,0)
interval	56,7*	56,7*	50*	50*	50*	53,3*	60*	53,3*	73,3	53,3*	73,3	88,7 (7,2)	89,3 (7,9)
rhythm	53,3*	73,3	50*	53,3*	53,3*	76,7	76,7	63,3*	96,7	63,3*	93,3	91,7 (8,2)	91,5 (6,8)
metric	63,3	66,7	56,7	53,3	60	76,7	60	70	73,3	73,3	70	83,5 (10,3)	81,6 (9,9)
memory	66,7*	53,3*	50*	46,7*	40*	53,3*	73,3	50*	73,3	66,7*	80	92,8 (6,3)	89,5 (7,2)

Note: Fr=French; E=English; (1) a loss is expressed in dB HL for the left and right ear; n=normal; s.d.=standard deviation;
*=scores under three standard-deviations from the mean of the unselected control group.

Figure Caption

Figure 1. Mean percentage of hits minus false alarms obtained for the amusic and control group in the anomalous pitch detection task. Each dot represents an individual score.

Figure 2. Mean ratings obtained in pleasantness judgments of dissonance (left panel) and in happy-sad control judgments (right panel).

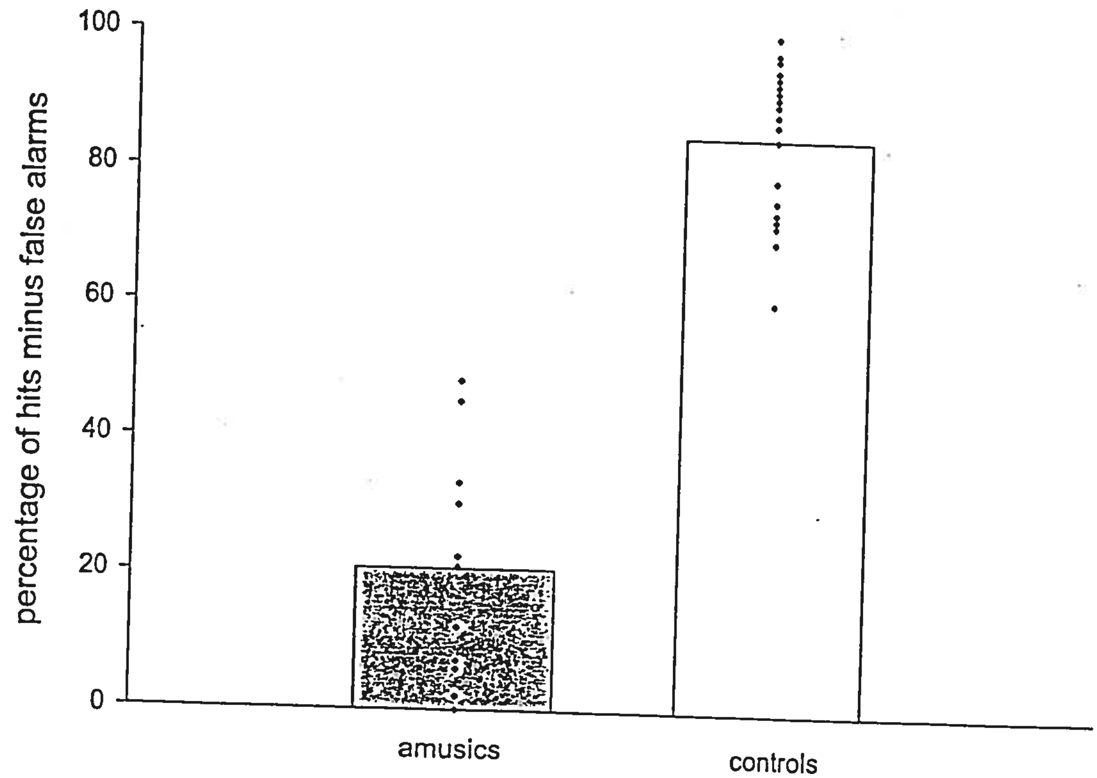
Figure 3. Mean percentage of hits minus false alarms and standard errors obtained, in each group, for spoken sentences (left panel) and non-speech derivations (right panel) as a function of the location of the pitch discrimination change.

Figure 4. Mean percentage of correct responses and standard errors obtained for each group in the identification of melodies, lyrics, speakers' voices and environmental sounds. Naming and recognition scores are represented separately.

Figure 5. Mean percentages of hits minus false alarms obtained, for each group, in familiarity decisions for melodies and spoken lyrics of songs. Dots represent individual results.

Figure 6. Mean percentages of hits minus false alarms obtained, for each group, in the memory recognition test as a function of the material presented. Dots represent individual results.

Figure 7. Average ratings and standard errors obtained by each group in singing from memory, sung repetition and in keeping time with the music. Dots represent individual results.



percentage of hits minus false alarms
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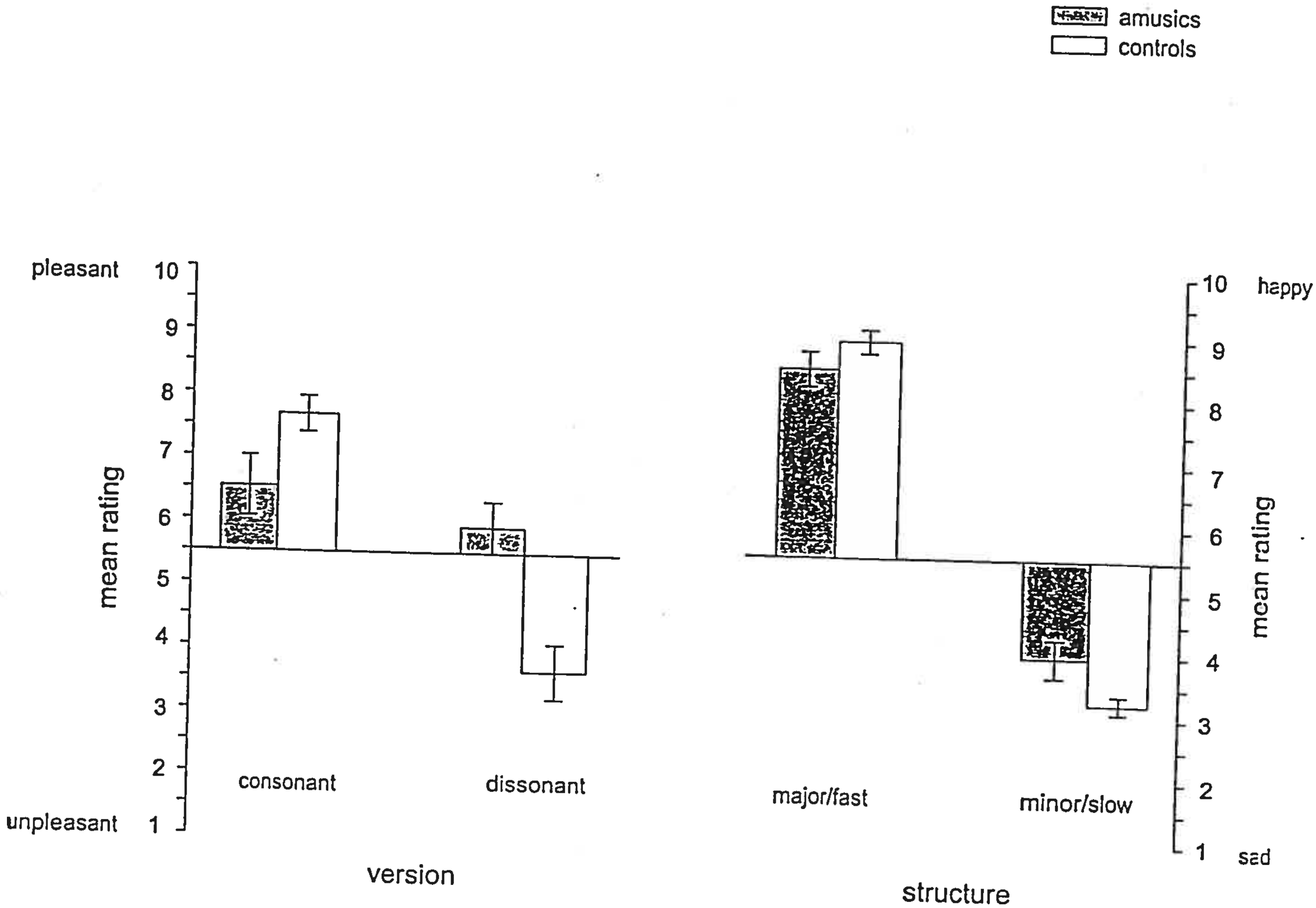
amusics
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pleasant
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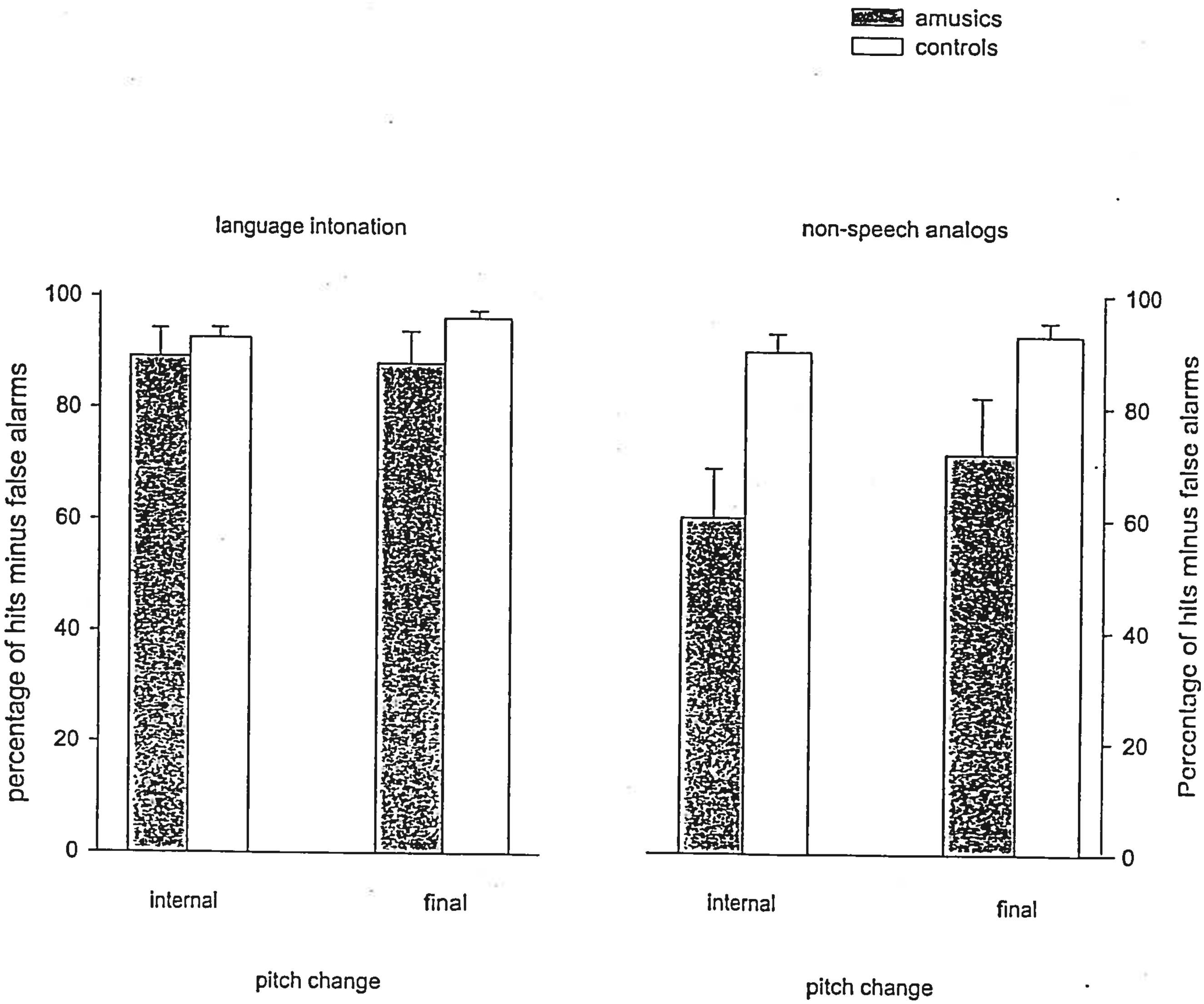

amusics
controls
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percentage of hits minus false alarms
Percentage of hits minus false alarms
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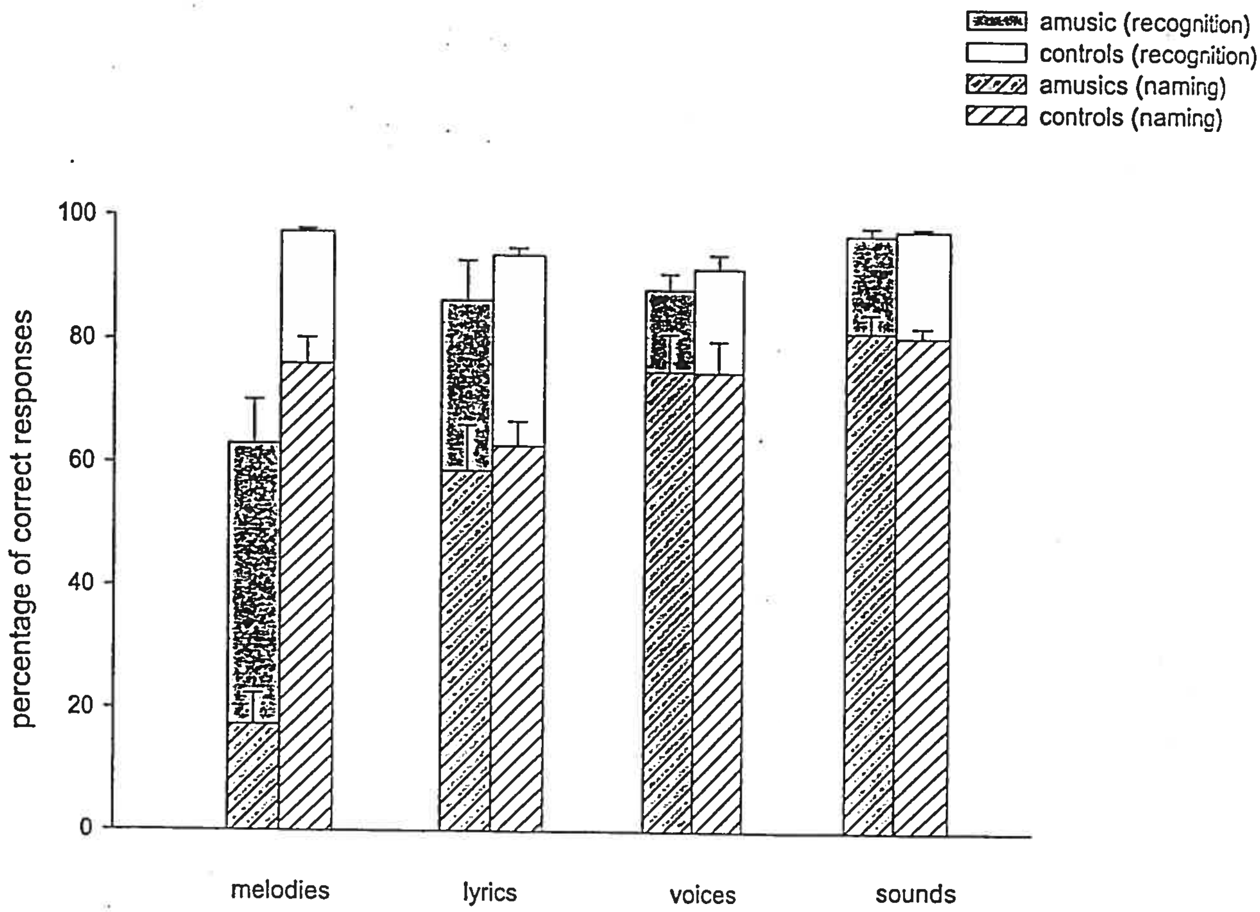
amusic (recognition)
controls (recognition)
amusics (naming)
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percentage of correct responses
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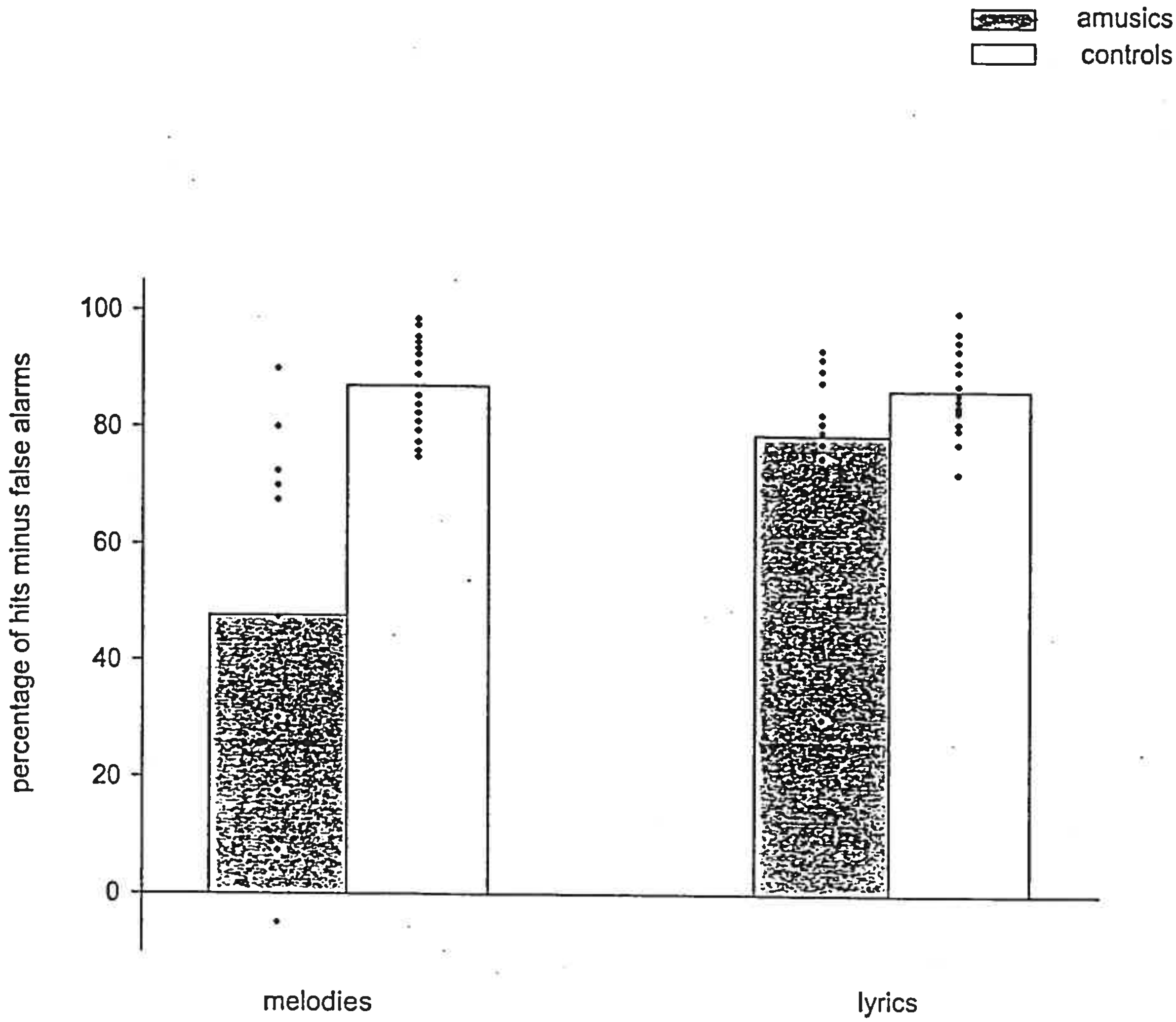
melodies
lyrics
voices
sounds


amusics
controls
percentage of hits minus false alarms
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melodies
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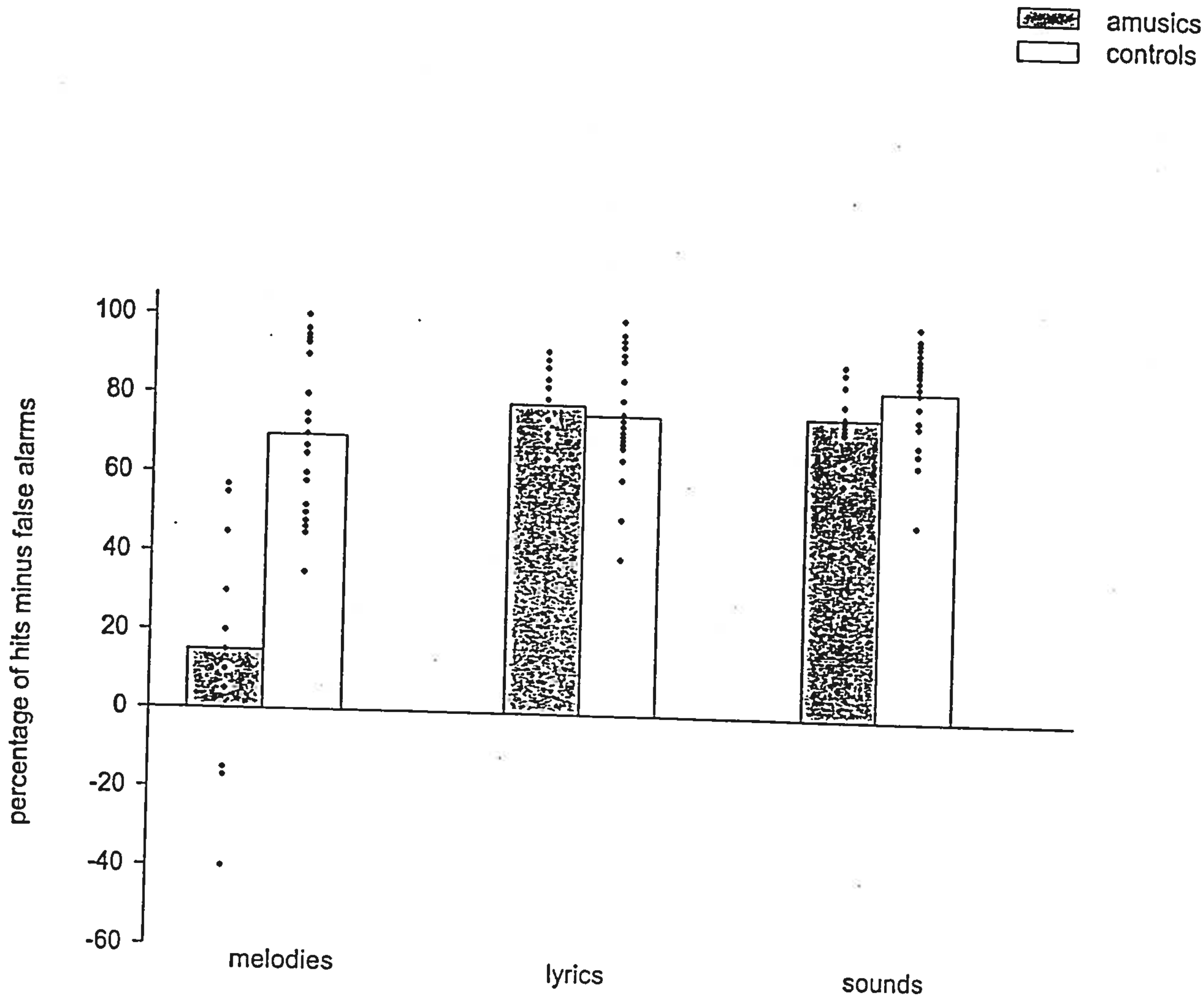
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controls
percentage of hits minus false alarms
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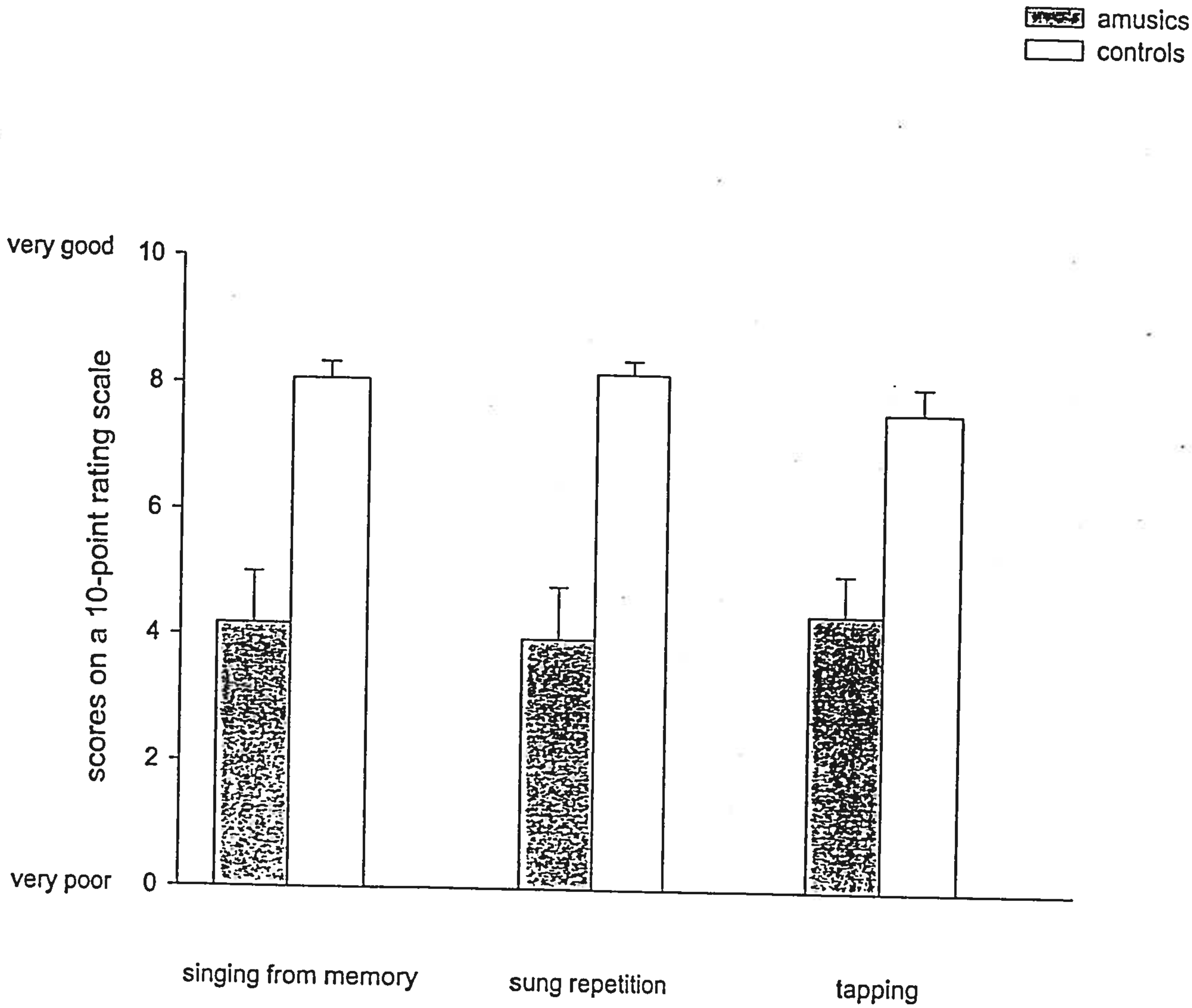
amusics
controls
very good
very poor
scores on a 10-point rating scale
10
8
6
4
2
0
singing from memory
sung repetition
tapping

TROISIEME ARTICLE : CONGENITAL AMUSIA: A DISORDER OF FINE-GRAINED PITCH DISCRIMINATION

Congenital Amusia:
A Disorder of Fine-grained Pitch Discrimination

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Running head: no pitch, no music

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Summary

We report the first documented case of congenital amusia. This disorder refers to a musical disability that cannot be explained by prior brain lesion, hearing loss, cognitive deficits, socio-affective disturbance, or lack of environmental stimulation. This musical impairment is diagnosed in a middle-aged woman –hereafter, Monica – who lacks most basic musical abilities, including melodic discrimination and recognition, despite normal audiometry and above average intellectual, memory and language skills. The results of psychophysical tests show that Monica has severe difficulties in detecting pitch changes. The data suggest that music processing difficulties may result from problems in fine-grained discrimination of pitch, much in the same way as many language processing difficulties arise from deficiencies in auditory temporal resolution.

Introduction

Music, like language, is a universal and specific trait to humans. Similarly, music appreciation, like language comprehension, appears to be the product of a dedicated brain organization (Peretz and Morais, 1993). Accordingly, as language, normal acquisition of musical competence is expected to recruit and fine-tune distinct neural networks in the human brain. In the advent of a slight but congenital neural deviation, selective deficits of learning may occur. Indeed, some individuals suffer from language-specific disorders and a large research effort has been undertaken to understand the origin and varieties of these disorders. Some researchers claim that language impairments arise from failures specific to language or cognitive processing (Studdert-Kennedy and Mody, 1995). Others hold that language deficiencies result from a more elemental problem that makes individuals unable to hear fine acoustic temporal changes (Tallal and Piercy, 1973). Nothing comparable has been initiated in the musical domain.

The possibility that certain persons are born with a specific musical deficit has been envisaged for more than a century (Grant-Allen, 1878; Geshwind, 1984). However, the evidence rests on anecdotal case descriptions. In this paper, we present the study of a female volunteer --Monica-- who manifests a music-specific disorder, sometimes called tone deafness but for which we prefer the term "congenital amusia". The term reflects better the likelihood that there are multiple forms of music developmental disorders, as there are various patterns of acquired amusia resulting from brain accident (Marin and Perry, 1999).

Case history

Monica declared herself as "musically impaired" in response to a newspaper advertisement. She was selected as the most clear-cut case among 37 volunteers with similar problems who were tested in our laboratory. Monica is a French-speaking woman in her early forties, currently completing a Master's degree in Mental Health after having practiced as a nurse for many years. She has no psychiatric or neurological history, or any hearing loss. A thorough and detailed audiological examination failed to reveal any anomaly in the ear. The audiometry, tympanometry, and oto-emissions yielded normal results.

A quantitative volumetric examination of Monica's brain with magnetic resonance imaging was carried out after transformation by linear scaling and rotation into the standardized stereotaxic space of Talairach. Inspection of the scan did not reveal any remarkable anatomical abnormalities. There was no indication of subtle changes or atrophy within the region of the auditory cortex. The volume of both the left (4028 mm^3) and right (1298 mm^3) Heschl's gyrus lies within two standard deviations of those (2363 ±963 and 1626 ±534 mm^3) of 40 normal unselected subjects (Penhune, Zatorre, MacDonald and Evans, 1996; see Figure 1). Similarly, the volumes of secondary auditory cortices found within the planum temporale in the two hemispheres (3410 and 4916 mm^3 in left and right, respectively) were within one standard deviation (4199 ±1366 and 4105 ±1562 mm^3) of those of a normal population (Westbury, Zatorre and Evans, 1999). These findings do not exclude the possibility of abnormalities at a microstructural level, but they do indicate that there is no obvious cortical atrophy or pathology.

Monica scores above average in a standard intelligence test: Her WAIS-R Intellectual Quotient is 111. She has an excellent memory for both verbal and nonverbal information: Her Wechsler Memory Quotient is 113. Her working memory is also normal, as measured by the digit span test (with a forward span of 7 and backward span of 4). This span score corresponds to the 53rd and 81st percentile in the intelligence and memory scale, respectively. Nonetheless, Monica suffers from a lifelong inability to recognize or perceive music, sing or dance. This musical handicap has persisted despite the fact that Monica was given the opportunity to develop at least some musical competence during childhood and adolescence. She was involved in a church choir as a child and later, in a high-school band. On both occasions, Monica did these musical activities under social pressure. Monica now reports that she does not like to listen to music because it sounds to her like noise and induces stress. She admits, however, that she came to realize her shortcoming more acutely after her recent marriage to a college music teacher. It is possible that her musical disability might have a genetic origin because other family members (her mother and brother but not her father and sister) are reported to be similarly impaired, although they have not been formally tested.

Tests and results

To confirm Monica's disorder and to determine whether the deficit is specific to music, we evaluated her with numerous tests that were initially designed for the assessment of brain-damaged patients with probable acquired amusia. Monica was then tested for the presence of a low-level pitch perception deficiency. This seems quite likely since fine-grained discrimination of pitch is probably more relevant to music than to any other domain, including speech intonation. Speech intonation contours, for example, use variations in pitch that are larger than half an octave, to convey relevant information (Patel, Peretz, Tramo and Labrecque, 1998). In contrast, melodies mostly use small pitch intervals, of the order of a $1/12^{th}$ or $1/6^{th}$ of an octave (Vos and Troost, 1989). Therefore, a degraded pitch perception system may compromise music perception but leave speech prosody unaffected.

Is Monica amusic ?

Monica was evaluated with a battery of tests that measure the use of musical characteristics that are known to contribute to the perception and memory of unfamiliar but conventional music. In the battery, the same pool of novel monodies are arranged in multiple tests, of which two deal with pitch variation discrimination and two with temporal variation discrimination. The tests have been used extensively and shown to be effective in identifying adult nonmusicians with deficits in basic musical abilities (Ayotte, Peretz, Rousseau, Bard and Bojanowski, 2000; Liégeois-Chauvel, Peretz, Babaï, Laguitton, and Chauvel, 1998). Monica's performance on most tests of the battery lies at chance level, well below the traditional cut-off score of three standard deviations below the mean obtained by control subjects.

Monica's performance relative to the normal distribution is illustrated in Figure 2 with the data obtained in two essential subtests which explore the processing of sequential pitch variations, the focus of the present study. In these two subtests, two types of manipulation were applied to the same tone in the short monodies. One manipulation consisted in creating a *contour*-violated alternate melody by modifying the pitch of one note so that it changed the pitch direction of the surrounding intervals while maintaining the original key. The other manipulation consisted in creating a contour-preserved but *interval*-violated alternate melody by modifying the same critical pitch to the same extent (in terms of semitone distance), while maintaining the original contour and key. These melodies were used in the contour and interval subtests, respectively. Each subtest consists of 15 pairs made of identical melodies and 15 pairs of different melodies. Only the nature of the pitch change inserted in the melodies distinguish the contour and interval subtest. Subjects are required to judge, for each pair, whether the target and the comparison sequence are the same or different. As can be seen in Figure 2, Monica's scores indicate her inability to discriminate between melodies on the basis of both their global contour and local interval pattern.

Monica's musical deficit extends to the discrimination of rhythm. In the rhythmic test, the same basic set of monodies as those used in the contour and interval subtests serve for discrimination. Only the nature of the change inserted in the comparison melody differs by involving a rhythmic modification. The manipulation consists in changing the duration values of two adjacent tones so as to change rhythmic grouping by temporal proximity while keeping the meter and the total number of sounds identical. This can be done either by changing two quarter notes for a dotted quarter and an eighth note, or by interchanging the order of two successive but different duration values (e.g., a half note followed by a quarter note becomes a quarter note followed by a half note). Thus, the only cue available for discrimination is the rhythmic pattern. The task also requires a "same-different" classification of 30 melody pairs.

On this rhythm discrimination subtest, Monica scores at chance level (with 16 correct responses out of 30 while the controls' mean is 27.4 with a s.d. of 2). However, she is able to derive the metrical pattern of the musical sequences to some extent, since she scores in the low but normal range when classifying the same melodies as a march or a waltz (with 19 correct responses out of 30; controls' performance range: 18 to 30). The monodies were the two-phrase versions of the one-phrase sequences used in the other tests. The 30 melodies differed in meter since half of these were written in a duple meter and half were written in a triple meter. This difference in meter induces the perception of a periodic alternation between strong and weak beats every two or three beats (as in a waltz : ONE, two, three, ONE, two, three,...) depending on its meter. No accent (i.e., intensity cue) is present in the music itself.

Monica's difficulties with rhythm were, however, not further examined. Her deficiency in processing melodic variations was studied in more detail. The first step consisted in documenting the perceptual basis of her problem in the discrimination of sequential pitch variations. To this aim, Monica was tested with a pitch anomaly detection task. She was presented with the same melodies as used in the musical battery but this time she was asked to detect the presence of a "wrong note" (that is, a note that was deliberately played out of key in half the melodies) by giving a "yes" or "no" response. Monica's performance was at chance (with 17/ 30 correct responses), again far below that of unselected controls (mean= 25.1; s.d.=2.5). Thus, Monica seems to suffer from a perceptual defect and not from a general deficiency in working memory for musical material.

Monica's perceptual disorder may account for her poor recognition of highly familiar music. Presented with a written forced choice among four possible titles, she is able to recognize only 22 out of 52 melodies that are familiar to most people in Quebec

(Peretz, Babaï, Lussier, Hébert and Gagnon, 1995). As expected, the result is much inferior to the scores obtained by two control groups. The first group consisted of four women of Monica's age and education level but without music-related symptoms, referred to as " matched controls " (mean: 51; χ^2= 36, p<.001), and the second group consisted of 29 unselected control adults (mean: 50; χ^2=32, p<.001). Thus, Monica failed to learn the songs and melodies that everyone else can easily identify. This recognition failure cannot be explained by inattentiveness or a poor auditory system in general, since Monica is able to identify without hesitation 30 of 33 voices of well-known speakers, as do matched controls (mean: 30.3; s.d.: 3.1). The speakers can only be identified from voice characteristics due to the fact that the speech excerpts were selected so as to contain casual speech without any contextual word cues.

Is the disorder specific to the musical domain ?

Monica's deficit seems limited to music, as suggested by her intact ability to recognize voices. In order to further assess the domain specificity of Monica's impairment in comparable testing conditions, we asked her to learn 20 melodies taken from familiar songs, presented auditorily, one at a time. In a subsequent recognition test phase, Monica was asked to distinguish the studied targets from 20 unstudied (but equally familiar) melodies with which they were mixed. For comparison, Monica was also asked to learn and recognize 20 spoken lyrics (taken from the same familiar songs) and 20 environmental sounds (e.g. a barking dog). The three tests were performed in different sessions (as in Peretz, 1996). Monica performed at chance (with 52 % of correct responses) for melodies and much worse than controls (with 84 and 82 %, for matched and unmatched controls, respectively, χ^2=22 and 19, p < .001). In contrast, her scores are comparable to those of controls for the nonmusical materials, whether song lyrics (85 % correct; controls' mean: 86; s.d.: 8) or nonverbal environmental sounds (88 %; controls' mean: 90; s.d.: 6). These results illustrate the domain-specificity of Monica's musical impairment.

Is the musical impairment due to poor pitch discrimination ?

Monica's impaired monitoring of pitch errors in melodies led us to assess the presence of a low-level deficiency in pitch perception. To assess the presence or absence of a pitch defect, we trained Monica to respond to a pitch change inserted in an five-tone sequence. Monica was asked to respond " yes " whenever she detected a pitch change on the fourth tone in an otherwise constant pitch tone sequence and to respond with a " no " when unable to detect a pitch change. Feedback was provided after each trial. The results are reported in Figure 3 (left panel).

As can be seen, Monica can detect a pitch change of 11 semitones, if and only if the pitch change is rising, not when it is falling. The pattern is similar whether pure tones or piano tones are used and whether tone duration is longer (700 ms) or shorter (350 ms). With pure 700 ms tones, the only tones to be judged "intelligible" by Monica, she can barely detect a rising pitch change as large as two semitones. She is at chance for one-semitone changes. Her performance is slightly better when presented with pure-tone pairs (see right panel in Figure 3). Yet, she is still at chance for descending pitches of two semitones.

The difference between the tone-sequence and the tone pair condition cannot be explained by a working memory problem. Pitch changes are identical in the tone sequence and the tone pair; the only difference lies in the repetition of a constant pitch in the sequence context. This tone repetition would be expected to facilitate pitch change detection by providing more reference tones and twice as many cues in terms of pitch intervals and pitch directions compared to a tone pair. This facilitatory effect of the sequence context was not observed with Monica. The fact that Monica detects somewhat better rising than falling pitch changes across contexts also argues against a working memory problem. There is no reason to expect working memory to be tuned to specific pitch directions. In contrast, this peculiar and systematic pitch defect points to the presence of an anomalous pitch perceptual system.

We construe that this striking pitch processing deficit may well account for the music-specific disorder observed in Monica. In effect, Monica is unable to perceive pitch variations that lie at a semitone distance, which corresponds to 1/12th of an octave and is the smallest pitch interval that is used in Western scales. Normal discrimination abilities in the large majority of adult and infant subjects are more fine-grained, in the order of 1/4th tone, in both ascending and descending directions (Olsho, Schoon, Sakai, Turpin and Sperduto, 1982). In contrast, for Monica, most musical pitch variations lie below threshold.

Is speech intonation spared ?

Since Monica suffers from a pitch discrimination deficit that is remarkably severe, she might suffer from an impairment in the discrimination of speech intonation patterns on the basis of pitch cues alone. To assess this possibility, we exploited once again experimental tests that have been previously used with brain-damaged amusic patients (Patel et al., 1998). These tests are constructed by computer-editing two basic sets of sentences that only differ from each other by local pitch changes. In the first set, the change affects the last word by marking a rise in pitch so as to indicate a question (e.g. He speaks French?) or a falling pitch (e.g., He speaks French.) to indicate a statement. The pitch rise is in the order of 3 to 11 semitones for questions, while the pitch fall is of 2 to 3 semitones for statements. In a second set of sentences, the pitch difference (of a magnitude of 8 semitones) affects an internal word of the sentence to mark emphatic stress like in " SING now please ! " and " Sing NOW please! " (Capitals indicate the stressed word).

The two sets of sentences were presented in isolation. Subjects judged whether the sentence indicates a statement or a question for the first set of sentences differing by their final pitch change; or they indicated which word bore the stress for the second set of sentences. These two tasks were relatively easy to complete for both Monica and controls who obtain 100 and 98 % correct, respectively, in the final pitch change condition, and 77 and 87% (range: 67-100), respectively, in the internal pitch change condition. Thus, Monica's disorder in monitoring pitch variations does not appear to encompass speech intonation.

Discussion

From the data presented, we conclude that congenital amusia, or tone-deafness, is not a myth (Kazez, 1985), but a genuine and specific learning disability for music. Affected individuals, who are otherwise unimpaired, have extreme difficulties appreciating, perceiving and memorizing music. The systematic evaluation of Monica, who reported to be severely handicapped in the musical domain despite her efforts to learn it, largely confirms the presence of an under-developed system for processing music. Her musical impairment cannot be explained by hearing loss, since she has normal audiometry and is proficient in the recognition of other auditory non-musical material, such as voices, spoken lyrics and common environmental sounds. Her musical disorder cannot be explained by a lack of exposure, since she had music lessons during childhood and was raised in a family in which a few siblings are musically normal. Finally, the musical deficit cannot be ascribed to some general cognitive slowing since she scores normally on cognitive tests and has reached a high level of education. The musical disorder appears as an isolated deficit in an otherwise fully normal cognitive and affective system.

Monica's learning disability may arise from a basic problem in pitch discrimination. For Monica, music must sound monotonous since she cannot detect pitch variations that are smaller than 2 semitones. Thus, she cannot perceive the pitch differences -- the tone and semitone-- that are the building blocks of the musical scales of most cultures. This pitch defect is construed as the origin of congenital amusia.

Reducing congenital amusia to a single deficient mechanism in pitch perception requires elaboration. First, ascribing musical impairments to a low-level pitch discrimination problem is not trivial. Acquired amusia resulting from brain lesions usually arise from failures specific to higher-level aspects of music processing, by degrading tonal knowledge for example, and is not necessarily associated with a pitch discrimination defect (e.g., Peretz, 1993). Conversely, brain lesions that disturb pitch discrimination do not necessarily affect music processing to the same degree as seen in Monica (Tramo, Bharucha and Musiek, 1990). For instance, excision of cortical tissue within the right Heschl's gyrus produces an elevation in the threshold for discriminating pitch direction (Johnsrude, Penhune and Zatorre, 2000) on the order of two semitones, which is better than Monica's performance. Such lesions do cause perceptual and memory disturbances for melodies, but patients with such lesions cannot be called amusic since they can discriminate above chance (Zatorre, 1985) and can also learn new melodies (Samson and Zatorre, 1992), albeit less well than controls.

Secondly, the fine-grained pitch discrimination disorder was not the only impaired musical ability. Monica was also impaired in discriminating melodies by their rhythm. Her difficulty with temporal patterns seems to hold even when all pitch variations are removed, although the nature and extent of this impairment could not be documented. There are several possible explanations for the presence of this myriad of musical deficits. The explanation that we presently favour is that the ensemble of musical deficits are cascade effects of a faulty pitch processing system. In effect, in our ongoing study of other congenital amusic participants we have observed that they systematically fail on tests that probe their ability to process musical pitch variations whereas most of them are able to process musical temporal variations as normal listeners. Thus, the presence of a pitch defect appears diagnostic of the existence of a music learning disability whereas the association of a rhythmic problem appears optional. Hence, we construe that a faulty perception of pitch might bring the development of the entire musical system to a halt. In this perspective, fine-grained pitch perception might be an essential component around which the musical system develops in a normal brain.

Further investigation of multiple single cases such as this one, and the joint study of musical pitch and time structure in infants, will be necessary to test the above account. In turn, this research endeavor should provide serious indications as to which processing component is crucial for normal musical development and in what way music might be special. More generally, knowledge of every aspect of this music-specific disorder should enrich current views of other forms of learning disabilities. For instance, one may consider congenital amusia as a mirror image of some developmental disorders of language, whereby pitch would be to music what time is to speech.

Experimental procedures

Battery of musical tests

The musical battery uses a pool of 30 *novel* two-phrase monodies that obey the rules of the Western tonal system. Half were written in a duple meter, half in a triple meter. The two-phrase sequences were used in the metric subtest. For all the other subtests, only the second phrase of each sequence served as stimuli. These second phrases were 4 bars long, lasted about 4 s. and contained from 8 to 19 tones (mean= 10.7). The stimuli were generated on computer controlling a Yamaha TX-81Z synthesizer. The chosen tempo was fixed at 120 crochets per minute and the voice was the approximation of a piano sound. The analog output was recorded on a digital DAT SONY recorder, which was also used to play melodies to the subjects.

In the contour and interval subtests, two types of manipulation were applied to the second phrase of 15 original sequences. These manipulations consisted of creating a contour-violated alternate melody by modifying the pitch direction of one tone and an interval-violated alternate melody by modifying the pitch of same critical tone to the same extent in terms of semitone distance but in keeping with the original contour. The changed pitch remained in the key of the melody. Its serial position varied across melodies; half fell in the beginning of the melody and half fell in the end, while avoiding the first and last tone positions. Average pitch interval changes were 4.3 and 4.2 semitones apart from the original pitch, in the contour-violated and interval-violated condition, respectively. The minimal pitch change was set to three semitones and the maximal pitch change was set to 7 semitones. The changes generally fell into the frequency range of the melody. Two sets, each consisting of 2 practice trials and 30 experimental trials, were constructed with these melodies. Each trial consisted of a warning signal and a target melody followed, after a 2 s. silent interval, by a comparison melody. Duration of the inter-trial interval was 5 s. long. Each set consisted of 15 pairs that were made of identical melodies and 15 trials of different melodies, presented in a random order.

As mentioned previously, in the metric test, the 30 entire two-phrase sequences served as stimuli. Each sequence was recorded in a random order with an inter-trial interval of 5 s. These experimental trials were preceded by four practice trials. For the rhythmic test, the stimuli were derived from the second phrase of the 30 sequences used in the metric task, in order to correspond to the target melodies used in the contour and interval subtests. One manipulation was applied to these isolated phrases to create different comparison patterns. These temporal alternates involved a temporal grouping change by changing the durational values of two adjacent tones. This local change was such that the size of each temporal group defined by temporal proximity was changed, while keeping the meter and the total number of sounds identical. The serial positions of these changes varied across patterns. Thus, the only cue available for discrimination was the temporal grouping of the tones (i.e. the rhythm). A set of 2 practice and 30 experimental trials was constructed with these temporal patterns in the same way as the contour and interval conditions.

For the contour and interval subtests and the rhythmic test, subjects were required to perform a "same-different" classification task. They had to judge, on each trial, whether the target sequence and the comparison sequence were the same or not. Prior to each condition, two practice trials were presented. For the metric task, subjects were informed that they would be hearing waltzes and marches, which they had to discriminate along that dimension. Subjects were encouraged to tap along with what they perceived to be the underlying beat of each sequence. Feedback on the response was only provided on the practice trials.

Psychophysical tests of pitch discrimination

A standard, psychophysical forced-choice procedure was used in successive blocks of 40 trials each, starting with the largest pitch distance. In each block, half the stimuli contained no change and half contained a rising or falling pitch change; trials were presented in a random order. Monica was first tested with a 5-tone sequence in which the fourth tone could vary in pitch. We started with the largest changes of 11-semitones and with piano tones of different durations. The following blocks used smaller pitch distances and pure tones of 700 ms duration only, since these tones were reported to be intelligible by Monica. In a subsequent session, Monica was presented with the same pure 700 ms tones but within a pair of stimuli such that the second tone was either the same or different than the first. Monica was invited to press a "yes" key when she detected a change and to press a "no" key when she detected no change. Feedback was provided on the screen of the computer and the task was self-paced.

All stimuli were generated digitally using the MITSYN signal-processing software at a sampling rate of 44.1 kHz. A sequence consisted of five successive tones and a pair consisted of two successive tones. The duration of each tone was 700 msec, unless noted otherwise, with the sound envelope of a piano tone. The frequency spectrum of each tone either corresponded to the one of a piano tone or to a single frequency with a similar amplitude counter (referred to here as "pure tones"). All target tones corresponded to C5 (with a fundamental frequency of 524 Hz). In half the stimuli, either the fourth tone of the sequence or the second tone of the pair could take a different pitch value. These different pitch values correspond to B5 (988 Hz) or C4 sharp (277 Hz) for the 11 semitone distance, to E5 (660 Hz) or A4 flat (416 Hz), D5 sharp (623 Hz) or A4 (440Hz), D5 (588 Hz) or B4 flat (466 Hz), and C5 sharp (555 Hz) or B4 (494 Hz) for the 4, 3, 2, 1 semitone distance, respectively. The stimuli were presented bilaterally through Sennheiser HD450 headphones, in a quiet room, at an intensity level of 70 dB SPL-A, via the Media Control Functions program.

Pitch variation discrimination in speech

Thirty-two sentence pairs naturally spoken by a native female French speaker were recorded. Each pair represented two lexically identical versions of a sentence, but differed in intonation. Twelve pairs constituted statement-question pairs (e.g. "Il parle Français." versus "Il parle Français?"). Twelve pairs constituted focus-shift pairs, involving a shift in the word which bore the focus of the sentence (e.g. "Allez DEVANT la banque, j'ai dit." versus "Allez devant la BANQUE, j'ai dit."; capitals indicates stress). All pairs were acoustically adjusted by computer editing to equalize patterns of syllable timing and loudness within each pair, yielding natural-sounding sentence pairs in which pitch variations was the only salient cue for intonation interpretation. The computer-editing procedure is described in detail in Patel et al. (1998). The sentences were presented one at a time, in a random order. For the final pitch change sentences, the task of the subject was to judge whether the utterance was a question or an assertion. For the internal pitch change sentences, the task of the subject was to indicate which word bore the stress. Each task was performed on 32 trials. No feedback was provided to the subject.

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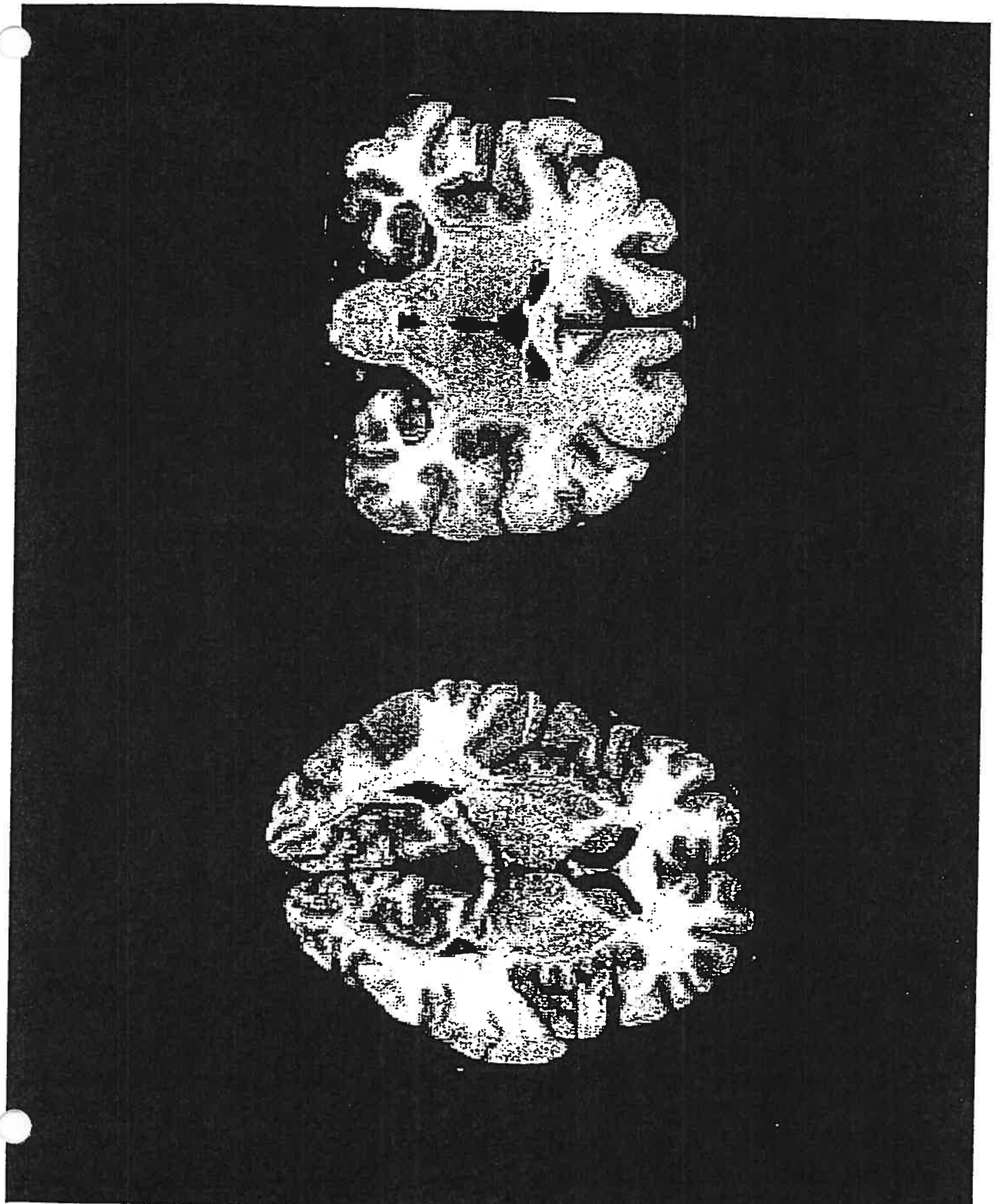
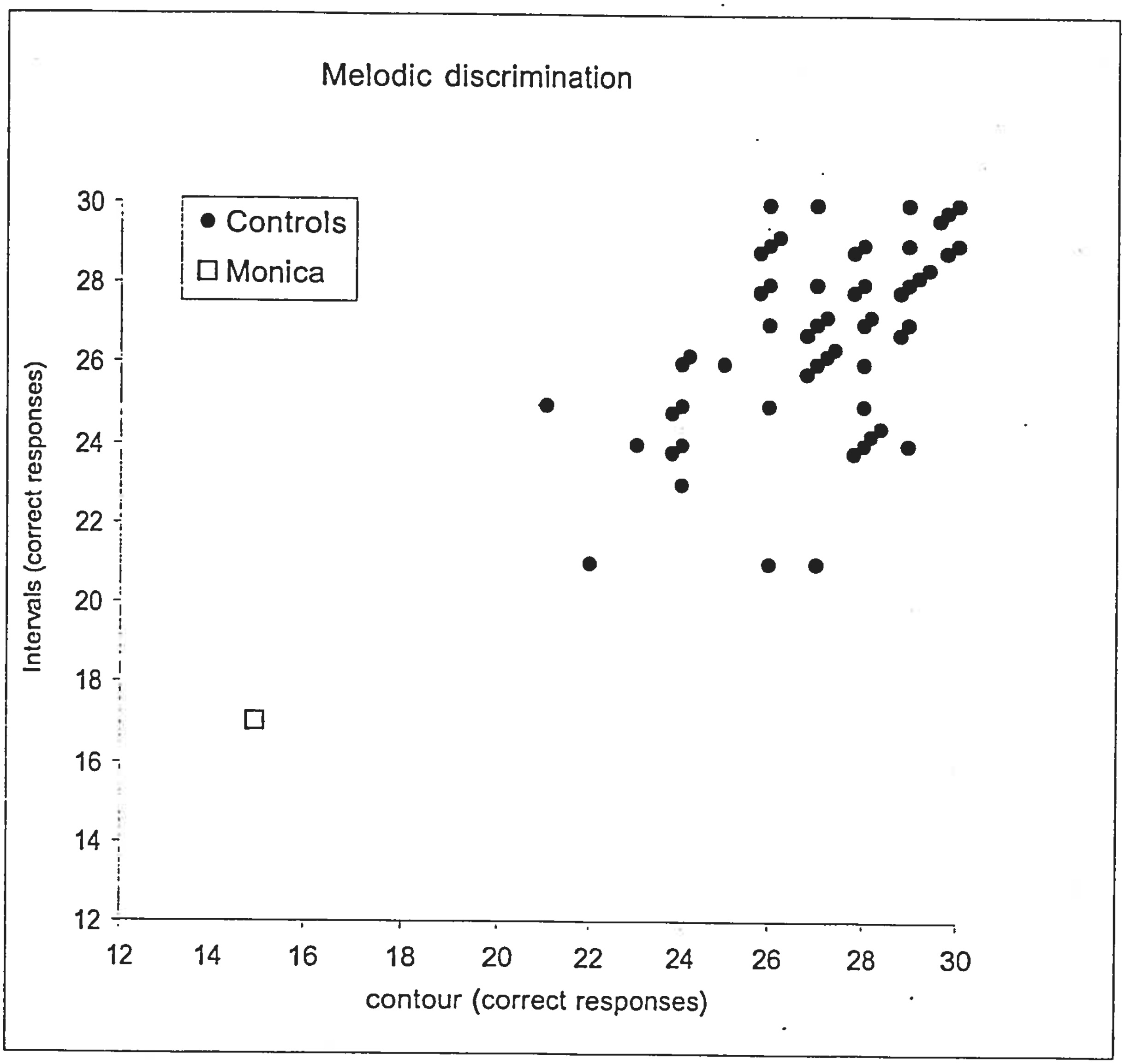
Figure legends

Figure 1. Magnetic resonance images of Monica's brain. The left and right image show coronal and horizontal sections, respectively, through the temporal lobes. The MRI has been linearly transformed and rotated into the standardized stereotaxic space of Talairach. Heschl's gyri are labelled in color.

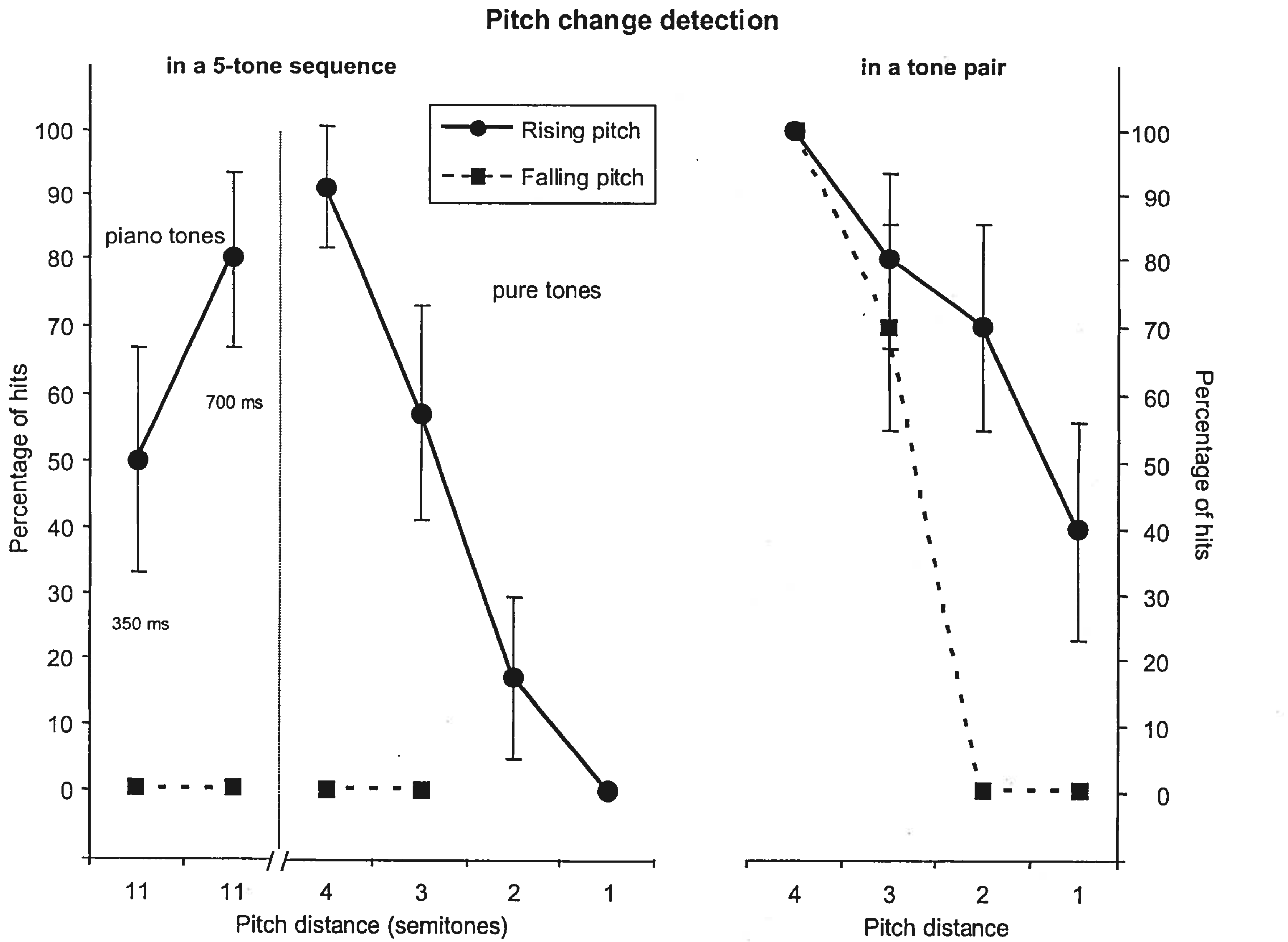
Figure 2. Melodic discrimination. Monica's scores in the contour and interval subtests of the musical battery relative to the individual scores of 57 nonmusicians (Age range: 14-74 yrs. Range in years of education: 7-20 yrs), who have no known impairment for music. Chance level is 15.

Figure 3. Pitch change detection. Monica's percentages of correct detection of a pitch change either on the fourth tone in a five tone sequence (left panel) or on the second tone of a pair (right panel) as well as the corresponding standard error, for each pitch distance. False alarms are not reported because Monica mistakenly detected a change in an identical tone sequence only once.





Peretz et al. Figure 2



Peretz et al. Figure 3

DISCUSSION GÉNÉRALE

Découverte de cas d'amusie acquise et congénitale

Les trois études composant la présente thèse apportent une importante contribution au domaine scientifique. Elles ont d'abord permis la découverte de nouveaux cas d'amusie manifestant un trouble spécifique de perception et de reconnaissance musicale, ce qui n'est pas trivial car de tels cas demeurent rares et relativement peu étudiés. L'examen de ce désordre musical s'est fait, lors de la première étude de cette thèse, chez des patients ayant subi une lésion cérébrale (amusie acquise). Il ne s'agissait pas d'une première recherche avec une telle population car quelques cas d'amusie acquise avaient déjà été rapportés dans la littérature, dont trois cas d'agnosie musicale récemment étudiés dans notre laboratoire (Peretz, 1996; Peretz et al., 1994, 1997). Il était donc déjà connu qu'une lésion au cerveau pouvait causer des troubles spécifiques de reconnaissance musicale. Par contre, cette étude a permis d'apppuyer notre hypothèse voulant que les dommages causés au cerveau suite à une ligature d'anévrisme rupturé sur l'artère cérébrale moyenne constituent un bon moyen de détecter et d'étudier l'agnosie musicale. En effet, les résultats indiquent une haute incidence de troubles de perception et de mémoire musicale chez les patients ayant subi des opérations pour réparer des anévrismes situés sur l'artère cérébrale moyenne. Sept patients sur 20 manifestent un trouble plus ou moins sévère de reconnaissance musicale; la probabilité de découvrir des patients avec un trouble de reconnaissance musicale est donc de 35% dans un échantillon non sélectionné de patients ayant subi une chirurgie afin de ligaturer un anévrisme sur l'artère cérébrale moyenne. De plus, cette approche a mené à la découverte de deux nouveaux patients clairement agnosiques pour la musique (N.R. et R.C.), ce qui porte à cinq le nombre de cas ici étudiés avec la même étiologie neurologique. Ce pourcentage est élevé étant donné la rareté de cas similaires d'agnosie musicale dans la littérature (Dalla Bella et Peretz, 2000). Ainsi, la première étude confirme que les régions irriguées par l'artère cérébrale moyenne sont déterminantes dans la reconnaissance musicale.

Alors que l'amusie acquise avait déjà fait l'objet de recherches expérimentales, il en est autrement pour les déficits musicaux présents sans atteinte cérébrale connue (l'amusie congénitale). Quelques cas anecdotiques avaient été rapportés dans la littérature, mais ceux-ci ne fournissaient aucune base empirique solide à l'existence de l'amusie congénitale. Les résultats obtenus dans notre deuxième étude sont donc importants car ils suggèrent que l'amusie congénitale n'est pas un mythe, mais bien un trouble d'apprentissage pour la musique. En effet, l'évaluation systématique de 11 adultes, qui s'estimaient eux-mêmes sévèrement handicapés dans le domaine musical en dépit des efforts pour apprendre la musique, confirme la présence d'un sous-développement du système en jeu dans le traitement musical. Ce déficit musical s'inscrit dans la catégorie des troubles spécifiques d'apprentissage, comme la dysphasie ou la dyslexie. En effet, le trouble n'est pas la conséquence directe d'un déficit d'acuité auditive ou d'une lésion cérébrale. De plus, il ne provient pas d'un manque de stimulation musicale car tous les participants amusiques ont suivi des cours de musique durant leur enfance. Finalement, les participants avaient atteint un haut niveau de scolarité, ce qui exclut un possible retard d'apprentissage général.

Au-delà de son caractère nouveau et spectaculaire, l'amusie congénitale indique que la spécialisation de certaines régions du cerveau pour la musique ne découlerait pas uniquement d'une exposition musicale fréquente et précoce puisque, même avec une stimulation adéquate, nos amusiques congénitaux n'ont pas réussi à développer normalement les habiletés musicales élémentaires. De plus, comme il n'y a pas eu de lésion cérébrale chez ces personnes, d'autres facteurs à l'origine de l'amusie congénitale sont à considérer. Une hypothèse plausible est celle d'une origine génétique. En effet, on observe une concentration familiale chez les amusiques congénitaux car plusieurs membres de chaque famille amusique rapportent un problème musical. De plus, au moins un des membres de la fratrie n'éprouve aucun trouble musical, ce qui indique que cette concentration est davantage susceptible d'être d'origine génétique que de découler de facteurs environnementaux (p.ex. manque d'exposition à la musique).

Trouble de reconnaissance musicale: problème de discrimination des variations de hauteur dans une mélodie

Tel que prévu, les troubles de reconnaissance observés chez les amusiques (acquis ou congénitaux) s'accompagnent d'une déficience au niveau perceptif et font donc partie de la catégorie des agnosies apperceptives. L'agnosie associative, reflétant des habiletés perceptives préservées malgré l'incapacité de reconnaître un air musical, est sûrement un phénomène rare car aucun cas n'a été observé dans la présente thèse. En effet, tous les patients agnosiques suite à une atteinte cérébrale ont une performance inférieure à celle du plus faible des participants contrôles (point de césure) pour au moins un des trois tests de discrimination des variations de hauteur dans une mélodie, à l'exception d'un patient qui manifeste un problème perceptif seulement pour la discrimination rythmique. De la même façon, tous les amusiques congénitaux ont une performance inférieure à celle du plus faible des participants contrôles dans la plupart des tests demandant une analyse des hauteurs de son (discrimination de deux mélodies, détection de fausses notes dans une mélodie, jugement sur la consonance d'airs musicaux). Un trouble de discrimination de deux mélodies différant par le rythme est également observé chez la moitié des amusiques congénitaux, mais il s'accompagne toujours d'un problème de perception des variations de hauteur. Ainsi, les habiletés à discriminer des variations de hauteur dans une mélodie sont déterminantes dans l'apparition de l'agnosie musicale acquise ou congénitale. Ces résultats ne sont pas surprenants car une étude effectuée avec des sujets normaux a démontré que la variation des hauteurs dans une mélodie est l'élément le plus déterminant dans la reconnaissance musicale, tandis que le rythme constitue un indice moins important (Hébert et Peretz, 1997).

Sur la base des résultats comportementaux obtenus dans la deuxième étude, nous proposons que l'ensemble des problèmes musicaux observés chez les personnes avec une amusie congénitale originerait d'une déficience pour le traitement des variations de hauteur. Bien que le lien causal soit évident entre une atteinte du traitement des variations de hauteur et un déficit de perception mélodique ou de reconnaissance musicale, certaines relations sont plus difficiles à concevoir. Par exemple, les incapacités à discriminer deux mélodies par le rythme et à garder la pulsation rythmique semblent, à première vue, peu reliées aux habiletés de la perception des hauteurs. Quelques explications sont possibles, mais pour lesquelles nous n'avons pas encore d'appui empirique. D'abord, le problème de discrimination des hauteurs peut interfèrer sur l'exécution des tâches rythmiques. Par exemple, le « bruit » produit par les variations mélodiques peut perturber l'attention que les amusiques portent au changement rythmique. Une tâche de discrimination du rythme sur des notes d'une même hauteur serait une façon de vérifier l'interférence produite par les variations de hauteur. De plus, les variations mélodiques (p.ex. la tonique arrive souvent sur un temps fort et donne un appui à la musique) et non seulement le rythme permettent de suivre la pulsation d'une musique, et encore ici, un problème de discrimination mélodique peut perturber l'extraction de la pulsation d'une musique. Enfin, une autre hypothèse est que la perception fine des variations de hauteur serait une composante essentielle au développement de l'ensemble du système musical, mélodique et rythmique, dans un cerveau normal.

Cette dernière hypothèse ne s'applique par contre pas aux amusies acquises. En effet, un cas d'agnosie associative suite à une lésion cérébrale (C.N.) a déjà été décrit dans la littérature (Peretz, 1996; Peretz et al., 1994), ce qui démontre que les troubles de reconnaissance musicale ne découlent pas toujours d'un problème perceptif. De plus, un des patients agnosiques de la première étude ne démontrait aucun problème de perception des variations de hauteur, mais éprouvait de la difficulté à percevoir les variations rythmiques. L'atteinte de la perception rythmique n'origine donc pas d'un problème au niveau de la discrimination des hauteurs chez ce patient.

Ces deux visions de l'amusie congénitale et acquise ne sont par contre pas incompatibles. En effet, comme les patients cérébrolésés ne manifestaient aucun problème de traitement des variations de hauteur avant leur accident vasculaire-cérébral, les différentes fonctions et structures musicales se sont développées correctement chez eux. Ces fonctions ont par la suite été atteintes, de façon sélective ou concomitante, selon le lieu et l'étendue de la lésion cérébrale. Par contre, chez les amusiques congénitaux, le problème de discrimination des variations de hauteur empêcherait le développement adéquat de l'ensemble des habiletés musicales, même celles reliées au rythme.

Spécificité du trouble au domaine musical

L'intérêt d'étudier ces cas d'amusie n'est pas seulement à cause de la nature et l'ampleur du déficit musical, mais également à cause de la préservation des fonctions non musicales. La spécificité du trouble au domaine musical suggère que les habiletés musicales sont indépendantes des autres fonctions cognitives. D'abord, l'administration d'une batterie d'évaluation intellectuelle et mnésique a fait la preuve du bon fonctionnement cognitif général de tous les cas d'amusie (acquise ou congénitale). Aucun trouble au niveau du langage réceptif n'a été observé chez les sujets amusiques évalués. De plus, ce qui est particulièrement remarquable c'est que la perception et la reconnaissance des différents événements auditifs non musicaux étaient généralement préservées.

En effet, les deux patients cérébrolésés ayant été identifiés comme clairement agnosiques pour la musique (N.R. et R.C.) étaient capables de distinguer entre des paroles provenant de chansons familières et celles provenant d'expressions familières. Ils réussissaient également, lorsqu'on leur lisait les paroles, à fournir les titres de chansons qu'ils n'étaient plus capables de reconnaître par la mélodie. Cependant, le désordre auditif de R.C. n'est pas entièrement spécifique à la musique car il éprouvait de légères difficultés à reconnaître des stimuli non verbaux, tels que les voix humaines ou certains sons de l'environnement (Faïta et al., 1996). Ainsi, selon la localisation et l'étendue de la lésion dans le cerveau d'un amusique acquis, des régions cérébrales traitant des stimuli auditifs non musicaux peuvent également être perturbées à cause de leur proximité avec les régions musicales, ce qui n'exclut pas l'autonomie des fonctions musicales.

On constate également que tous les amusiques congénitaux ont préservé leur capacité à traiter le matériel non musical, aussi bien que les sujets contrôles. En effet, ils démontrent de bonnes habiletés pour ce qui est d'interpréter l'intonation du langage, identifier des personnalités connues à partir de leur voix uniquement et reconnaître des sons environnementaux tels que les cris d'animaux et les sonneries. À l'exception d'un sujet, Monica, ils ont tous réussi à identifier et à reconnaître des chansons familières à partir des paroles seules. Cette dernière constatation nous permet de s'assurer que les amusiques n'ont pas été privés de musique dans leur environnement et qu'ainsi leur déficit ne découle pas d'un manque de stimulation musicale. La faible connaissance de paroles de chansons chez Monica pourrait par contre découler du peu d'intérêt qu'elle porte aux chansons. En effet, elle évite la musique car celle-ci lui est désagréable à l'oreille, tandis que la plupart des amusiques, même s'ils avouent apprécier peu la musique, la supporte et en écoute à l'occasion. Par ailleurs, même si la plupart des amusiques congénitaux se disent peu touchés par la musique, la perception des émotions gai et triste en musique est préservée chez eux, possiblement parce qu'ils peuvent utiliser le tempo et non seulement la mélodie pour baser leur jugement. Par contre, leurs faibles habiletés pour la perception mélodique les empêcheraient d'apprécier la musique à sa juste valeur.

Bien que les déficits des amusiques congénitaux soient spécifiques au domaine musical, il est important de noter que leur trouble de discrimination des hauteurs ne s'observe pas exclusivement lors d'un traitement musical. D'abord, le groupe d'amusiques congénitaux a obtenu une performance significativement inférieure au groupe contrôle lors d'une tâche de discrimination des variations de hauteur à l'intérieur de phrases déclaratives ou interrogatives où tous les indices linguistiques ont été enlevés. De plus, l'utilisation de tests psychophysiques chez un sujet amusique congénital (Monica) dans la troisième étude démontre que le problème de discrimination des variations de hauteur ne se manifeste pas seulement dans une mélodie, mais également dans un contexte non musical. En effet, le seuil de détection des changements de hauteur dans une séquence de cinq sons est beaucoup plus élevé que les sujets normaux : Monica ne peut détecter des variations de hauteur inférieures à un ton, tandis que la plupart des gens peuvent détecter des différences aussi minimes qu'un quart de ton (Luce, 1993). Il s'agit donc d'une atteinte à un traitement de bas niveau des hauteurs.

Ces observations suggèrent que le déficit de discrimination des hauteurs manifesté par les amusiques congénitaux n'est pas spécifique à la musique mais plutôt significatif à la musique, c'est-à-dire que ce problème a des répercussions particulièrement importantes dans le domaine musical, mais non dans les autres domaines. En effet, la discrimination fine des variations de hauteur est probablement plus pertinente et nécessaire à la musique que dans n'importe quel autre domaine. Par exemple, les mélodies utilisent généralement des changements de hauteur de l'ordre de un demi-ton ou un ton (Vos et Troost, 1989), tandis que les variations de hauteur pour l'intonation de la parole tendent en moyenne à être plus grandes (en moyenne, 5 demi-tons) (Patel et al., 1998). Ainsi, une dégradation du système de traitement des hauteurs peut compromettre la perception musicale, mais laisser les autres domaines, tels que l'intonation de la parole, intacts. Le même mécanisme de traitement des hauteurs peut néanmoins servir aux différents domaines auditifs car il s'agit d'un système de traitement auditif de bas niveau.

Cette « significativité » à la musique peut aussi s'expliquer par l'utilisation d'autres indices sonores que la hauteur pour discriminer les stimuli auditifs non musicaux. Par exemple, on sait que les changements temporels suffisent à décoder le langage (Shannon et al., 1995). Par contre, l'utilisation de paramètres sonores autres que les variations de hauteur (p.ex. durée, timbre, intensité) ne serait pas suffisante pour reconnaître la musique occidentale (Hébert et Peretz, 1997 ; Terhardt, 1978). Il pourrait en être autrement de certaines musiques non occidentales (p. ex. musique africaine) où le rythme a un rôle dominant, souvent même plus important que la mélodie. On peut s'attendre à ce que les amusiques congénitaux, malgré leur trouble de discrimination des hauteurs, aient davantage de facilité à traiter et à reconnaître ces extraits musicaux.

Réseaux neuronaux intervenant dans la reconnaissance musicale

Une des contributions de cette thèse est de préciser quels sont les réseaux neuronaux intervenant dans la perception et la reconnaissance musicales. Comme H.V. (Griffiths et al, 1997) et le patient de l'équipe de Piccirilli (2000), les deux patients clairement agnosiques de la première étude, N.R. et R.C., démontrent des atteintes du côté droit sur la radiographie cérébrale, ce qui indique qu'une lésion à cet hémisphère semble suffisante pour produire une agnosie musicale apperceptive. Il est de plus intéressant de mentionner que ces quatre cas d'agnosie apperceptive pour la musique ont subi des lésions à des aires similaires à l'hémisphère droit (ou non dominant). Tous les quatre montrent des indices de lésions dans les régions postérieures du gyrus temporal; l'insula est également touchée chez trois d'entre eux. Ces régions interviendraient dans la reconnaissance musicale car elles seraient déterminantes dans le traitement des variations de hauteur d'une mélodie et donc dans l'encodage adéquat de l'information musicale. D'ailleurs, un déficit perceptif pour le traitement des variations des hauteurs est typiquement associé à une atteinte de l'hémisphère droit (Liégeois-Chauvel et al., 1998; Peretz, 1990). Cet effet de latéralisation est aussi observé dans la première étude car la grande majorité des patients qui manifestaient un déficit pour la discrimination de mélodies par les variations de hauteur avaient subi une chirurgie à l'hémisphère droit. Ainsi, la première étude permet d'observer une certaine constance entre la nature du déficit musical (agnosie apperceptive) et le site de la lésion (régions postérieures du gyrus temporal supérieur de l'hémisphère droit). Par contre, l'étendue des dommages survenant suite à un accident vasculaire-cérébral ne permet pas de préciser davantage les structures impliquées dans la reconnaissance musicale.

Contrairement à celui des personnes manifestant une amusie acquise, le cerveau des amusiques congénitaux n'est pas altéré par de vastes lésions. L'analyse de leur cerveau offre donc la possibilité de préciser davantage les structures cérébrales traitant les stimuli musicaux. Par contre, le cerveau d'un seul amusique congénital (Monica) a été analysé jusqu'à maintenant. L'analyse volumétrique quantitative de ce cerveau, effectuée avec l'imagerie par résonance magnétique, n'indique aucun changement ou atrophie dans la région du cortex auditif. Ces découvertes n'excluent cependant pas la possibilité de l'existence d'anomalies au niveau microstructurel, mais elles indiquent qu'il n'y a pas d'atrophie ou pathologie corticale évidente. Ainsi, nous ne pouvons présentement confirmer l'hypothèse selon laquelle l'amusie congénitale résulte d'une légère anomalie lors du développement du système auditif, probablement à l'hémisphère droit.

Appuis aux fondements biologiques de la musique

Les différentes informations recueillies dans les trois études de cette thèse soulèvent la possibilité que la musique soit biologique. En effet, la spécificité des troubles musicaux observés chez les amusiques (acquis et congénitaux) indique que le système musical est indépendant des autres fonctions cognitives et que certaines régions cérébrales sont consacrées au traitement musical. De plus, l'existence de l'amusie congénitale suggère que la spécialisation de régions cérébrales pour la musique ne découle pas uniquement d'une stimulation musicale précoce. L'hypothèse avancée, selon laquelle les problèmes de perception des hauteurs des amusiques congénitaux ne sont pas spécifiques mais plutôt significatifs à la musique, pourrait par contre indiquer que les réseaux neuronaux ne sont pas dédiés au traitement exclusif de la musique, mais à celui de tous les stimuli demandant une analyse fine des hauteurs. Par contre, la présence de tels réseaux a sa raison d'être et n'est pas fortuite. Comme la musique serait le seul domaine auditif où le traitement fin des hauteurs joue un rôle important, on peut penser que ces structures ont été dédiées au traitement fin des hauteurs afin de permettre une perception et une reconnaissance adéquate de la musique. D'ailleurs, une grande variété de musiques à travers le monde utilisent de petits intervalles de hauteur (Vos et Troost, 1989), ce qui démontre l'importance, chez tout être humain, d'une capacité à traiter finement les hauteurs pour reconnaître la musique.

Un autre appui aux fondements biologiques réside dans l'existence d'arguments en faveur d'une origine génétique à l'amusie congénitale. En effet, on observe une concentration familiale chez les amusiques congénitaux qui serait davantage d'origine génétique qu'environnementale. À cet argument s'ajoute une étude effectuée chez des jumeaux monozygotes et dizygotes qui conclut que la perception des hauteurs (dans un test de détection de fausses notes similaire à celui utilisé dans notre étude) est hautement héréditaire et fortement influencée par les gènes (Drayna et al., 2001). L'amusie congénitale pourrait donc être d'origine génétique car l'ensemble des problèmes musicaux des amusiques congénitaux découlerait, selon notre hypothèse, d'un trouble de perception des hauteurs.

En conclusion, les trois études de cette thèse ont fourni un cadre permettant la découverte et le diagnostic de l'amusie acquise ou congénitale, ce qui devrait faciliter les recherches futures sur cette pathologie. L'ensemble des tests neuropsychologiques utilisés ont également permis de préciser la nature et la spécificité des déficits musicaux chez ces amusiques, et à confirmer l'importance de la discrimination des hauteurs dans la perception et la reconnaissance musicale. Bien que les résultats rapportés dans cette thèse suggèrent qu' à la base de l'amusie congénitale il y a une atteinte de bas niveau pour le traitement des hauteurs, ceux-ci demeurent préliminaires. Par contre, une étude psychophysique menée récemment dans nos laboratoires auprès de neuf sujets amusiques congénitaux a confirmé leurs difficultés à discriminer les changements de hauteur inférieurs à deux demi-ton, en comparaison des contrôles qui ont affiché un effet plafond pour un intervalle de la moitié d'un demi-ton (Hyde et Peretz, sous presse). La poursuite des recherches permettra de préciser la contribution d'un tel déficit aux problèmes musicaux observés chez les amusiques congénitaux. L'emploi de la résonance magnétique fonctionnelle chez ces sujets lors de l'écoute de séquences sonores aidera également à déterminer les réseaux neuronaux responsables du traitement des variations de hauteur. Cette étude est présentement en cours dans nos laboratoires.

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