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Research report

Integration of lexical and sublexical processing in the spelling of regular words: A multiple single-case study in Italian dysgraphic patients

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ABSTRACT

In this study we investigated 12 cases of “mixed dysgraphia”, a spelling impairment where regular words are spelt better than either ambiguous words or regular non-words.

Two explanations of mixed dysgraphia were formerly offered by Luzzatti et al. (1998): (i) a double functional lesion of the orthographic output lexicon (or damage to its access) and of the acoustic-to-phonological conversion; and (ii) some kind of interaction/summation between lexical and sublexical spelling routes when processing regular words.

We first analysed whether a double functional lesion was sufficient to explain the mixed dysgraphia, checking acoustic-to-phonological conversion by means of the repetition of words and non-words: the answer was positive in five cases and uncertain in three. We tested the remaining four cases to see if there was an interaction between lexical and sublexical processing of regular words, quantifying for each patient, on a probabilistic basis, the separate contribution of the residual lexical and sublexical resources. We investigated whether the processing along these routes was simultaneous but independent (“independent cooperation”) or if instead there was “interaction”, i.e., the simultaneous activity led to an added increase of efficiency over and above the mere combination of separate success probabilities. For one case the processing along the two routes was independent, in the other three cases an interaction resulted. Following the same approach, we found that for the five cases with a double functional lesion, the observed success on regular word spelling was higher than that expected on a probabilistic basis, but the interpretation of this finding was different.

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1. Introduction

Contemporary cognitive models of spelling assume at least two routes for the translation of phonology to orthography: the lexical route by which familiar words are processed as a whole, and the sublexical phonological-to-orthographic conversion routine by which unfamiliar words are assembled¹ (Baxter and Warrington, 1985; Beauvois and Dérouesné, 1981; Coltheart et al., 1980; Harris and Coltheart, 1986; Patterson et al., 1985; Patterson, 1986; Shallice, 1981).

In this study we refer to the dual-route model of spelling developed by Patterson (1986). The model assumes two independent routes of spelling: a lexical route and a sub-word-level procedure; the latter is based on two conversion processes: acoustic-to-phonological and phoneme-to-grapheme. The lexical route and the sub-word-level spelling routine have been considered independent and modular as they can be impaired in isolation after brain damage: some patients show selective damage of the sub-word-level spelling routine (phonological dysgraphia); others show the opposite dissociation, i.e., predominant impairment of the lexical spelling procedure (surface dysgraphia).

The two spelling routes may have a different weight in different languages. English speaking subjects rely mainly on the lexical route of spelling, whereas an opposite pattern applies for languages with shallow orthography (Zoccolotti et al., 2005). For instance, in Italian, sublexical processes almost always allow an adequate transcription of the phonological strings by means of phoneme-to-grapheme correspondence based on either one-sound-to-one-letter or syllabic conversion (i.e., when the orthographic realisation of a phone is determined by the following vowel: [ka], [ko], [ku] = CA, CO, CU; [ki], [ke] = CHI, CHE; [t/a], [t/o], [t/u] = CIA, CIO, CIU; and [t/i], [t/e] = CI, CE). However, even Italian orthography is not completely shallow: a shift from the sub-word-level to the lexical route is necessary whenever a phoneme can be transcribed by more than one orthographic rule, only one of which bears the correct lexical form. For instance the sound [kwo] can be spelt both as QUO and as CUO and only stored lexical knowledge allows correct spelling of the words CUORE (heart) and QUOTA (quota). When learning to write, Italian children proceed from a segmentation of the phonological input string to its translation into the corresponding orthographic sequence. Only the accustomed use of written language will shape the later acquisition of orthographic lexical representation. In languages where the sub-word-level routine is more extensively used, such as Italian, orthographic lexical representations are acquired later, and this makes surface developmental reading and spelling disorders (surface dyslexia and dysgraphia) more likely than in languages with predominantly irregular spelling, i.e., lexical spelling, such as English or French, where both phonological and surface disorders may emerge.

For what concerns the sub-word-level spelling routine, standard information processing models usually distinguish between two levels of conversion (Patterson, 1986; Patterson and Shewell, 1987; Bub and Chertkow, 1990; Ellis and Young, 1988; Howard and Franklin, 1988; Kay et al., 1992). An effective use of the sub-word-level routine (i.e., of the phonological-to-orthographic conversion rules) requires first that the subject segment and identify the phonological string to be converted. Such analysis takes place in the acoustic-to-phonological conversion. Primary damage to the acoustic-to-phonological conversion will thus cause incorrect spelling even if the phonological-to-orthographic conversion is unimpaired. In fact, the phonological-to-orthographic conversion skill (writing of single letters) of dysgraphic patients is often intact, while the isolation and identification of single phonemes to be converted may be dramatically impaired (Luria et al., 1969; Hatfield and Weddell, 1976; Luzzatti et al., 2000).

Luzzatti et al. (1998) reported that 21% of Italian aphasic patients were significantly better at spelling regular words than ambiguous/irregular words or non-words, and called this pattern *mixed dysgraphia* as this spelling disorder cannot be classified as surface or deep/phonological dysgraphia. In this study we investigated 12 further cases of “mixed dysgraphia”.

A crucial point for the understanding of mixed dysgraphia is the functional relationship between the lexical and sublexical routes: while most accounts assume that they are independent, data from normal subjects (Barry, 1988) and dysgraphic patients (Hillis and Caramazza, 1991; Rapp et al., 2002; McCloskey et al., 2006) indicate that lexical and sublexical processes may interact in different ways.

For instance, in their analysis of the possible mechanism(s) that allow the integration of lexical and sublexical information, Rapp et al. (2002) report the “fill in the gap” account, according to which first the lexical system generates a partial orthographic representation, and later the sublexical process provides a plausible content for these gaps (e.g., Miceli et al., 1987). Evidence has already been reported in English that the sublexical spelling system helps to reinforce the lexical system by strengthening the graphemes making up a target word and by creating a competitive advantage for the target over orthographically and/or phonologically similar word neighbours, which compete with it for output (Folk et al., 2002; Folk and Jones, 2004). Further evidence from lexical priming experiments indicates that lexical and sublexical processes activate a shared set of graphemic representations and, in particular, that lexical processing influences the spelling of non-words (Barry, 1988; Folk and Rapp, 2004). However, given the underlying shallow orthography, the order of processes might be the opposite in the Italian language. Rapp et al. (2002) reported an alternative explanation, called “simultaneous activation account”, assuming that both the lexical and sublexical processes are simultaneously engaged in the retrieval of an orthographic string from phonological input, and that they simultaneously activate candidate graphemic elements for output. They developed a version of the “simultaneous activation” account based on a “computational” model and, assuming that in the undamaged system the lexical source of activation prevails, suggested that integration of lexical and sublexical information would occur at the graphemic (buffer) layer.

¹ We will not consider here a further fractionation of the lexical route into a semantic lexical and a direct phonological/orthographic lexical route, and the consequent possibility of a spelling deficit deriving from the isolated impairment of these subroutines (direct dysgraphia).

More recently, McCloskey et al. (2006) suggested that activation of the graphemic layer feeds back to the lexical level, exerting an influence on lexical selection: the graphemic-to-lexical feedback provides a mechanism for the interaction of lexical and sublexical information. Other authors suggested that integration of lexical information with that arising from the sub-word-level phonological-to-orthographic conversion routine takes place at an earlier stage. For instance, Barry and Seymour (1988) found that in normal subjects, non-word spelling is influenced by the spelling of previously heard rhyming words. The interaction between lexical and sublexical processes was further corroborated by Folk et al. (2002) who described a dysgraphic patient whose weakened lexical system stood isolated: the deactivation of the sublexical process after articulatory suppression meant that it no longer contributed to spelling success.

According to Luzzatti et al. (1998), the origin of mixed dysgraphia might be twofold.

- (i) Two separate functional lesions, one of the acoustic-to-phonological conversion routine, the other of the orthographic output lexicon (or of the access to the lexicon) (Fig. 1), could account for the worse performance on both non-words and ambiguous words. As Luzzatti et al. (1998) suggested, regular words could still be spelt through an indirect circuit connecting the phonological output lexicon, the phonological buffer and the sound-to-spelling routines (a path that does not allow to spell either irregular words or non-words). Damage to the sound-to-spelling routines is another possible mode of sublexical spelling disorder, though this would not explain mixed dysgraphia. In fact, damage to the latter routines would no longer give regular words an advantage over non-words, as not even regular words could overcome damage to the orthographic output lexicon by using the sublexical route.
- (ii) Alternatively, there might be a “mutual interaction” of the residual resources of both the partially impaired sub-word-level routine and the lexical route, causing regular words to be better spelt as they can be processed along both routes. In other words, some kind of mutual interaction occurs in the

latter case, in the presence of impairment to both lexical and sublexical processing.

The aim of the present paper is to further explain the origins of mixed dysgraphia and to provide a principled account for its occurrence in Italian patients. We will first consider the hypothesis of a double functional lesion and afterwards that of interaction between the sublexical and lexical routes. We shall eventually try to throw light on the definition and meaning of the interaction itself.

(a) *Double deficit hypothesis*

This hypothesis implies two separate functional lesions: one of the orthographic output lexicon and the other of the acoustic-to-phonological conversion routine (Fig. 1).

- (i) Damage to the *orthographic output lexicon* implies a poor spelling of irregular/ambiguous words.
- (ii) Damage to the *acoustic-to-phonological conversion* can be ascertained contrasting the repetition of regular words with that of non-words (matched for length and phonological complexity). Damage to this process signifies poorer repetition of non-words than regular words because words can access the phonological output buffer for repetition via the phonological input and output lexicons. If there is no difference in the repetition of words and non-words, a selective damage to the acoustic-to-phonological conversion procedure can be excluded.

(b) *Interaction between lexical route and sublexical routine*

Alternatively, some kind of interaction between the two partially impaired writing routes could explain the pattern of a relative sparing of regular word spelling. Rapp et al. (2002) hypothesized a simultaneous activation of the two routes and integration of the products of these separate processes at the orthographic output buffer level. They suggested that a damaged orthographic output lexicon is replaced, or integrated, by sound-to-spelling conversion routines, giving rise to high probability phoneme-to-grapheme mappings. In addition, McCloskey et al. (2006) recently proposed that the

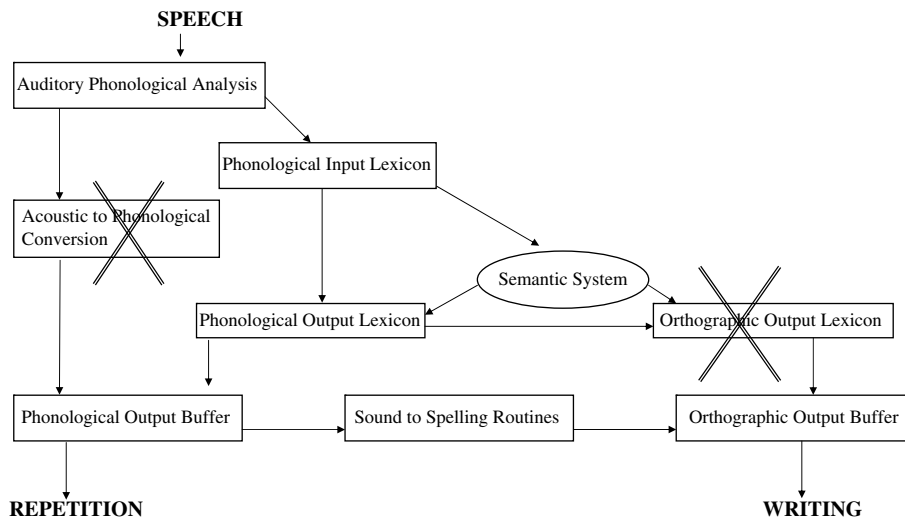


Fig. 1 – Two possible separate functional lesions that can associate in determining mixed dysgraphia.

interaction would act through a feedback of information from the phonological output buffer to the orthographic output lexicon. In Italian, however, orthography is substantially regular, the sublexical processes are far more effective, and the phonologically plausible alternatives are generally two or exceptionally three (e.g., CIELO (sky)/*CELO; BIGLIA (marble)/*BILIA/*BILLIA [plausible but non-lexical alternatives are marked with*]). Therefore, the likelihood of a correct spelling of ambiguous/irregular words by mere chance is far higher than in English, and the corpus of observed errors is therefore less informative. Due to this fact, the integration of the lexical and sublexical routes is less easily demonstrated in Italian by the emergence of phonologically plausible errors. Therefore, our inquiry into the functional relationship between orthographic output lexicon and phoneme-to-grapheme conversion routine will follow a different approach.

The core of our hypothesis is that in mixed dysgraphia, while writing regular words, the two spelling routes simultaneously process the phonological input and their respective (partially impaired) efficiency is combined so that the eventual spelling has a *higher success* than if it were processed by either route alone.

The above hypothesis will be referred to as a “cooperation” between two distinct processes. It is important to note that, in our definition, the presence of cooperation is defined on the basis of an *increased chance of success* with regular words, and not on the basis of detection of any particular error type, such as high probability of phoneme-to-grapheme mappings.

If we accept the existence of cooperation, a further question arises: are these simultaneous processes (lexical route and sublexical routine) independent, or do they reinforce each other? If the combination were able to reinforce the respective efficiency of each spelling route, we could speak in terms of a real “interaction” between them. For the sake of clarity, in the remainder of the paper we will denote as *independent cooperation* the condition where the simultaneously working processes do not further reinforce their efficiency (although they associate their separate success probabilities and increase the overall chances of correct spelling of regular words). We will use the alternative term of *interaction*

whenever the simultaneous activity of these processes implies an intrinsic increase of efficiency that is over and above the combination of their separate success probabilities.

How can we empirically contrast *independent cooperation* and *interaction*? Let us assume the case of independent cooperation: when writing regular words, the two spelling routes are activated simultaneously but operate separately and independently (“*independence assumption*”). In this account, if one of the two procedures is successful and the other fails, the former will feed the correct response to the output system. In this case, the probability of spelling success *at least* on one route will be calculated following a probabilistic rule. This mathematical procedure has been adopted in neurophysiology to decide between probabilistic and coactivation models in reaction times with redundant signals (e.g., Miller, 1982; Savazzi and Marzi, 2004). If, on the other hand, we observe a spelling success higher than that calculated on the basis of probability, we would have to reject the independence assumption, implying that interaction occurs (see Fig. 2).

In this study we will undertake a multiple single case study of the origins of mixed dysgraphia carried out on patients affected by this type of spelling disorder.

2. Materials and methods

2.1. Subject classification

In this study we analysed the spelling performance of a series of aphasic patients examined at the Aphasia Units of the Rho-Passirana General Hospital (Milano), and of the Montescano Medical Center (Pavia). The first step was to screen the cases affected by *mixed dysgraphia*.

Mixed dysgraphic patients were defined operationally on the basis of significantly better ability to spell regular words than words with unpredictable transcription (ambiguous/irregular words) and non-words. Logistic regression analysis (McCullagh and Nelder, 1983; Aitkin et al., 1989) was applied to the profile of each individual subject. The regression units were the stimuli of the Test and the dependent variable for

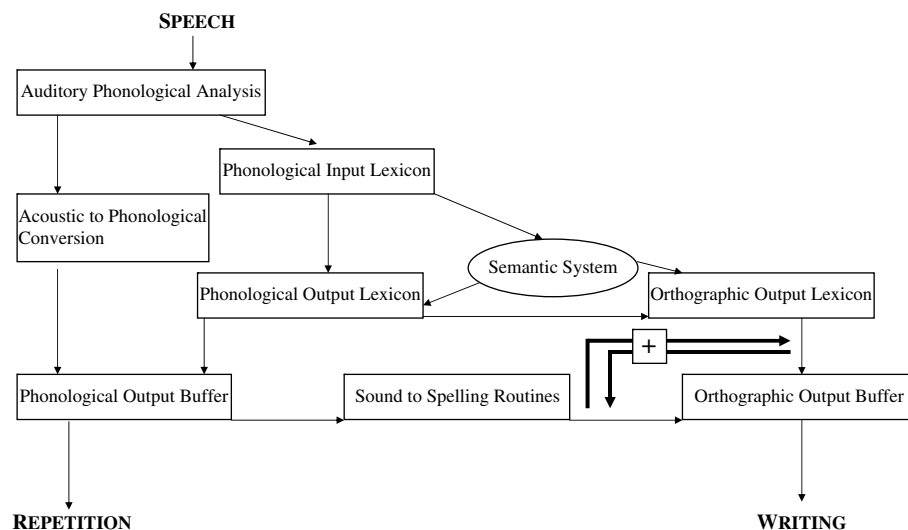


Fig. 2 – A possible account of the advantage of regular words spelling: interaction between lexical- and sub-lexical routes.

each stimulus was dichotomous (passed = 1, failed = 0). The model included both categorical (regular words versus non-words and regular words versus ambiguous words) and continuous variables (written word frequency according to Bortolini et al., 1972 and item difficulty, i.e., the number of subjects out of 110 controls who wrote each item correctly). As the statistical design also included covariance by item difficulty, any dissociation still significant after this adjustment would be stronger than – and not simply proportional to – the difference observed among controls. As a single case diagnostic procedure was used, the significance level of each comparison was set at .05, without an overall protection for the whole set of patients (Willmes, 1985).

In 12 patients (nine males and three females) the pattern of impairment corresponded to the operational criterion of *mixed dysgraphia*. Mean age was 48.1 years (SD = 11.3) and mean length of education 9.4 years (SD = 4.1).² Ten patients were affected by stroke, two by focal traumatic brain injury. Type and severity of aphasia was assessed by means of the Italian version of the Aachen Aphasia Test (Luzzatti et al., 1994b). Five patients suffered from non-fluent language impairment (Broca's aphasia). Seven patients suffered from a fluent language disorder: two from Wernicke's aphasia, four from conduction aphasia and one from anomia. Patients with pre-morbid hearing loss were not admitted to the study. Table 1 shows the demographic and clinical data of the sample and Table 2 the spelling profile of each patient.

2.2. Writing task

The writing task used in this study is extensively reported in Luzzatti et al. (1994a). In the present study we considered three of the five sections of the task: (1) 80 regular words with complete one-sound-to-one-letter correspondence; the stimuli set was stratified by lexical (*written word frequency*) and phonetic-phonological complexity (presence of *continuant* versus *plosive* consonants, *stimulus length*, presence of *consonant clusters*, presence of *geminate consonants*); (2) 55 ambiguous/irregular words, i.e., words with unpredictable transcription using sound-to-spelling routines; and (3) 25 non-words with one-sound-to-one-letter correspondence; the length and phonological/graphemic structure of these stimuli were parallel to that of the 80 regular words (see above).

The two sets of lexical stimuli (regular words and ambiguous words) were matched for word frequency (47.4 ± 69.6

² As three subjects (DR, LMi, MR) did not complete the minimum level of education in force in our country until 1969 (8 years) but attained only primary school-leaving certificate (5 years), their orthographic output lexicon may not have been fully developed and the mixed dysgraphia diagnosis might not be fully justified. In order to overcome this hindrance, we compared their spelling performance with irregular/ambiguous words to that of an additional control group of 22 healthy subjects with similar educational level. Mean correct controls performance was 80% (SD = 11%) and the worst score obtained was 62%. The observed correct spelling of all three patients under scrutiny (47%, 51%, 40% respectively) was lower than that (mean – 2 SD = 58%) of the control sample and was lower than the worst observed control score. Thus, we can be confident that their poor performance with ambiguous words is not exhaustively explained by low educational level.

Table 1 – Demographic and clinical data of the 12 mixed dysgraphic patients

Name	Gender	Age	Education	Aetiology	Aphasia type
DR	M	69	5	CVA	Broca's
MG	M	24	11	TBI	Broca's
LZ	F	36	16	CVA	Broca's
EG	M	53	8	CVA	Broca's
RL	F	51	8	CVA	Broca's
AL	M	49	8	CVA	Wernicke's
LMi	M	56	5	TBI	Wernicke's
CG	M	45	18	CVA	Conduction
LMo	M	43	8	CVA	Conduction
UB	M	48	8	CVA	Conduction
MR	F	62	5	CVA	Conduction
PV	M	41	13	CVA	Anomic

M = male; F = female; CVA = cerebro-vascular accident; TBI = traumatic brain injury.

and 47.6 ± 96.5 respectively, Luzzatti et al., 1994a). Both regular and ambiguous words included bisyllabic and three to four syllable stimuli. (see Appendix A for the complete list of stimuli).

Words and non-words were given separately and the examiner read each item aloud in a neutral tone, i.e., without underlining the presence of possible orthographic ambiguities. No feedback was provided on the adequacy of the responses. Spontaneous repairs and latencies longer than 5 s were scored as errors.

As is the case with the majority of languages using an alphabetic script, also in Italian a given phonological string has sometimes more than one possible orthographic solution, though only one is correct for a certain lexeme:

(1) The syllable [ʃe], for instance, is regularly transcribed by the sequence SCE. However, there are some words in which it is conveyed by the sequence SCIE (e.g., [ʃentsa] = SCIENZA/science instead of *SCENZA). The same applies to the syllables [tʃe] and [dʒe] which in certain cases are transcribed by the sequence CIE and GIE instead of CE and GE (e.g., [tʃelo] = CIELO/sky instead of *CELO and [idʒene] = IGIENE/hygiene instead of *IGENE).

(2) The phonological group [kw] may be transcribed by the orthographic sequences QU, CU, CQU. The general rule is that the syllables [kwa], [kwɪ] and [kwe] are transcribed with the sequences QUA, QUI and QUE (QUALE/which, QUINDI/thus, QUESTO/this), while the syllable [kwo] is transcribed by the sequence CUO (CUOCO/cook); also here, however, exceptions exist (e.g., QUOTA/quota instead of *CUOTA).

(3) The segments [lj]–[λ] and [nj]–[ɲ] that are phonologically distinct in Tuscan and central-southern pronunciation, are homophonous in north-western Italian pronunciation, so much so that the spelling of words like [balja]/[baʎa], nurse, (BALIA and not *BAGLIA) or [dʒeranjo]/[dʒeraɲo], geranium, (GERANIO and not *GERAGNO) is not predictable along the phonological-to-orthographic conversion routine.

(4) Plosive phones that are followed by the liquid consonants ([r] and [l]) or by the semiconsonant [j] are homophones to their doubled pairs, thus rendering uncertain the transcription of words such as [libro], book, (LIBRO and not *LIBBRO),

Table 2 – Spelling profile of each patient (percentages of correct responses)

	DR (%)	MG (%)	LZ (%)	EG (%)	RL (%)	AL (%)	LMi (%)	CG (%)	LMO (%)	UB (%)	MR (%)	PV (%)
Regular words	83	91	69	80	64	81	80	66	79	84	79	91
Ambiguous words	47	75	27	36	35	40	51	40	40	49	40	60
Non-words	40	40	32	32	8	52	16	28	32	32	44	72

[febre], fever, (FEBBRE and not *FEBRE), [biblico], biblical, (BIBLICO and not *BIBBLICO) and [publico], public, (PUBBLICO and not *PUBBLICO).

2.3. Repetition task

To detect possible damage to the acoustic-to-phonological conversion routine patients were also given a repetition task of words and non-words. The task was made up of 30 concrete words and 15 non-words matched for length (syllable number) and phonological complexity (presence of consonant clusters). Words and non-words were randomised into a single list and the examiner read each item aloud in a neutral tone. No feedback was provided on the adequacy of the responses. Spontaneous repairs and latencies longer than 5 s were scored as errors.

2.4. Searching for a possible impairment of the acoustic-to-phonological conversion routine (required for the co-occurrence of two cognitive deficits hypothesis)

As anticipated above, performance on the repetition task is of relevance to the hypothesis of two separate functional lesions (i.e., acoustic-to-phonological conversion routine and orthographic output lexicon). For each patient we analysed whether the success rate in repeating concrete words was different from that of non-words. The comparison was carried out using Fisher's exact test, and the hypothesis was considered one-sided. Because the power of this analysis was rather limited due to the relatively small number of stimuli, we also considered performances with a p-value between .05 and .10 as candidates for a discrepancy in processing words and non-words.

2.5. Interaction and verification of the independence model

The *independence assumption* predicts that regular words can be processed by either the lexical or sublexical route with independent success probability. In order to check for *independence*, we must estimate the separate capacity of these routes, and check whether the hit rate on regular words, that can be processed along both routes, corresponds to the combination of the separate capacities. According to a simplified model, the lexical and sublexical processes can be viewed as gates that are open only a given percentage of times and independently, and these percentages can be estimated, respectively, from the success rates of ambiguous words and non-words. As regular words can be processed along either of these pathways, the success rate with regular words expected with the independence hypothesis corresponds to the probability that at least one gate is open when regular words are being spelt. In

this case, the *expected* rate of hits with regular words is obtained summing the following three probabilities: hit on the lexical route and failure on the sublexical, failure on the lexical and hit on the sublexical, hit on both routes (see Table 3). The above model may appear somewhat simplistic, because it assumes that, for regular words, a hit on one route only is sufficient to yield correct spelling ("sufficiency assumption"). In other words, this model implies that, whenever the two channels feed contrasting responses, the system is able to decide which is the right response. This is not granted, and perhaps the marred process might feed a faster response, so that a spelling error could actually emerge: in such cases, the independence assumption would overestimate the expected hit rate. Notwithstanding this possibility, our statistical approach holds full validity: overestimation of the expected hit rate implies underestimation of the size of the real interaction. Therefore, any interaction detected under the independence assumption would be even stronger if the sufficiency assumption were violated.

The lexical route capacity can be reliably estimated by the hit rate in spelling irregular/ambiguous words. Estimating the sub-word-level capacity is less straightforward for the following reason: when acoustic-to-phonological conversion is impaired, the phonological output buffer can still be

Table 3 – Example of the expected success rate on regular words

Status of the system	Corresponding percentage
1. Lexical route open, sublexical process closed	$47\% \times 60\% = 28\%$ (expected success rate)
2. Lexical route closed, sublexical process open	$53\% \times 40\% = 21\%$ (expected success rate)
3. Both lexical route and sublexical process open	$47\% \times 40\% = 19\%$ (expected success rate)
4. Both lexical route and sublexical process closed	$53\% \times 60\% = 32\%$ (expected failure rate)

The lexical and sublexical processes are viewed as gates that are open only on a given percentage of occasions; these percentages can be estimated, respectively, from the success rates on ambiguous words and irregular words. As a regular word can be processed along either of these pathways, the success rate on regular words expected with the independence hypothesis corresponds to the probability that at least one gate is open. This is equal to the sum of the percentages corresponding to conditions 1, 2 and 3 (see below). Only in condition 4 does the percentage correspond to the expected failure rate, because in conditions 2 and 3 the "sufficiency assumption" (see text) implies that the response should be correct. Subject DR (see Table 2). Success rate on ambiguous words = 47%. The gate of the lexical route is open 47% and closed 53% of the occasions. Success rate on non-words = 40%. The gate of the sublexical process is open 40% and closed 60% of the occasions. The overall expected success rate on regular words is $28\% + 21\% + 19\% = 68\%$.

accessed by regular words through a connection between the phonological input and output lexicons, but this alternative route is not available for non-words. Consequently, a spared phonological output buffer could feed forward more information about regular words than non-words. The acoustic-to-phonological processing unit may be impaired when a patient repeats regular words better than non-words. In this case, hits on non-words spelling would underestimate the sub-word-level contribution to regular word spelling.³

Therefore, in order to check the *independence assumption*, we picked out the patients for whom repetition of regular words was equivalent to the repetition of non-words. For these patients, the probability that regular words are spelt correctly at least along one route (lexical or sublexical) can be calculated, yielding an *expected* success rate, that can be compared to the *observed* performance. In other words, by analysing patients in whom repetition of words and non-words is similarly affected, the efficiency of the sublexical route in regular word spelling can be estimated from their performance with non-words.

In practice, we verified whether the expected success rates on regular word spelling were within the 95% confidence limits of the observed success rates, using the method of Blyth, Still and Casella (Mehta and Patel, 2005). This statistical comparison enabled us to test inferentially our main question: i.e., *whether the performance observed on regular words is significantly better than that expected assuming independence. In this case, the independence assumption would be rejected with an error risk corresponding to the significance level* and it could be concluded that the lexical and sublexical routes do actually interact. As said earlier, any interaction detected with the suggested procedure would be even stronger if the marred process feeds a faster, though wrong response to the output system. In this case, a spelling error could be actually observed in some cases where one route works and the other is defective, but our formula would overestimate the hit rate expected under the independence assumption. In any case, it is easy to see that the sufficiency assumption maximises the hit rate expected under the independence assumption. A similar mathematical approach has been recently proposed to predict regular words reading and spelling accuracy on the basis of how well subjects perform on irregular words and non-words (Rapcsak et al., 2007).

3. Results

In a preliminary analysis we compared each subject's ability to repeat words and non-words. This procedure aimed at discriminating those patients for whom the isolated impairment of acoustic-to-phonological conversion could be reliably excluded from those where this impairment could be supposed. Table 4 reports the results obtained on the repetition task.

Statistical analysis (Fisher's exact test) showed that in three of the 12 mixed dysgraphic patients (DR, EG, LZ) repetition of non-words was significantly more impaired than that

of concrete regular words ($p < .05$), while in two other cases (MR and UB) this pattern was presumable ($.05 < p < .10$). In these patients a possible impairment of the acoustic-to-phonological conversion routine could be hypothesised.

In three other cases (LMi, CG and MG), the difference between the repetition of regular words and non-words was about 20%, and the p -value between .10 and .20. For these latter patients, considering the risk of type II error, a possible impairment of the acoustic-to-phonological conversion routine could not be reliably excluded.

Three patients (AL, LMo, RL) repeated very few stimuli, the observed difference between words and non-words on the repetition task was less than 20%, and no statistical difference emerged (p -value higher than .30). The remaining patient (PV) was almost flawless on the repetition task.

For the three patients with a poor overall repetition regardless of the stimulus type, we can presume that the phonological output buffer could not feed forward more information about regular words than non-words. For the patient with flawless repetition we are confident that the acoustic-to-phonological conversion routine was spared. Therefore, for these four patients (AL, LMo, RL and PV) we can assume that non-word spelling is a valid estimate of the sublexical route contribution to the spelling of regular words.

Table 5 shows the comparison between observed and expected hit rates of regular word spelling in the 12 patients of this study. Patients are grouped according to the theoretical questions that were at the basis of their analysis.

The first section of Table 5 reports the five patients whose pattern of performance is compatible with a *double deficit* (acoustic-to-phonological conversion and orthographic output lexicon, see Fig. 1). For these patients, better repetition of regular words than non-words implies that the repetition of regular words can bypass the acoustic-to-phonological sublexical conversion. Also the spelling process might follow a lexical (direct or semantic) route, along a connection between the phonological input and output lexicons. Such a bypass would obviously be impossible when spelling non-words. Therefore, the success rate on the spelling of non-words underestimates the possibility of spelling regular words without the intervention of the orthographic output lexicon, granted by a shunt along the phonological output lexicons and the sound-to-spelling routine. Due to this underestimation, the observed rate of correct regular word spelling should be higher than that estimated on the basis of probability. For all five patients with a double deficit, the observed hit rate of regular words was significantly higher than that expected on the basis of probability combination. This outcome is similar to the pattern of a real interaction, but its meaning might be different and could be defined as a "pseudo-interaction".

In the second section of Table 5, we report the data of the three patients (LMi, CG and MG) whose repetition performance did not allow a clear classification of the spelling impairment (see above and Table 4). For these patients we cannot make any predictions based on theoretical grounds. For one case (LMi) the limits of the observed hit rate did not include the rate expected on the basis of the independence assumption, whereas for the other two they did.

Table 5 (last section) reports the four patients without selective deficit of acoustic-to-phonological conversion. Only

³ Better repetition of words compared to non-words could be also due to limited verbal working memory (Bisiacchi et al., 1989) and/or an impaired processing in the phonological output buffer.

Table 4 – Repetition task (percentages of correct responses)

Patients	Repetition of non-words worse than repetition of words (patients with double deficit)					Non-conclusive comparison			Equivalent repetition of words and non-words			
	DR	EG	LZ	MR	UB	LMi	CG	MG	PV	AL	LMO	RL
Words (n = 30)	83%	100%	97%	63%	83%	80%	73%	53%	97%	17%	30%	13%
Non-words (n = 15)	53%	60%	60%	33%	60%	60%	53%	33%	100%	27%	20%	7%
Fisher test, p (one-tailed)	0.039	0.0006	0.003	0.056	0.090	0.142	0.157	0.171	0.667	0.339	0.367	0.453

The grouping criterion was the p-value (see text).

for these cases can spelling data help to discriminate between *independent cooperation* and *interaction*. In one patient (PV) spelling of regular words was not significantly better than that through at least one of the two spelling routes. The performance of case PV points to an *independent cooperation*. For the remaining three patients (AL, LMO and RL) the success rate with regular words was higher than that expected, indicating an *interaction* between the lexical and sublexical spelling routes.

Table 6 shows for all mixed dysgraphia patients the distribution of error types across the different sets of stimuli. These data are reported for the sake of completeness, but at this stage establishing any correspondence between the type of errors and the type of mixed dysgraphia would be premature. Misspellings can include more than one error type at a time and, consequently, for each patient the grand total of

error types may be higher than the overall number of misspelled words (spontaneous repairs and latencies longer than 5 s were scored as errors).

Three major error types are considered:

- (1) Single or multiple grapheme deletions, additions, substitutions not involving more than half of the orthographic string, resulting therefore in a phonologically similar response.
- (2) Phonologically plausible errors: orthographic substitution leading to phonologically plausible errors.
- (3) Other errors: e.g., no response, perseverations, fragments, errors in the realisation of regular digrams, written sequences without any phonological or lexical similarity to the target words.

In none of the 12 patients did the observed errors include any instances of either semantic or allographic deficits.

In principle, a dysfunction of the graphemic output buffer could have given rise to some of the single or multiple graphemic errors observed: this error type was observed in all 12 patients, irrespective **whether or not they presented any interaction**. Length effect could be viewed as a marker of a buffer deficit, and its analysis might be informative about the role played by a buffer dysfunction.

Table 5 – Expected hit rates of regular word spelling, observed hit rates, and confidence limits of observed hit rates

(a) Patients with a double deficit (acoustic-to-phonological conversion and orthographic output lexicon)	
DR	Expected 68%, observed 83% (95% confidence limits from 73% to 90%)
EG	Expected 57%, observed 80% (95% confidence limits from 70% to 88%)
LZ	Expected 50%, observed 69% (95% confidence limits from 58% to 79%)
MR	Expected 66%, observed 79% (95% confidence limits from 69% to 87%)
UB	Expected 65%, observed 84% (95% confidence limits from 74% to 91%)
(b) Patients whose repetition performance does not allow a clear classification	
LMi	Expected 59%, observed 80% (95% confidence limits from 70% to 88%)
CG	Expected 57%, observed 66% (95% confidence limits from 55% to 76%)
MG	Expected 85%, observed 91% (95% confidence limits from 83% to 96%)
(c) Patients with equivalent repetition of words and non-words	
PV	Expected 89%, observed 91% (95% confidence limits from 83% to 96%) ⇒ Independent cooperation
AL	Expected 71%, observed 81% (95% confidence limits from 72% to 89%) ⇒ Interaction
LMO	Expected 59%, observed 79% (95% confidence limits from 69% to 87%) ⇒ Interaction
RL	Expected 40%, observed 64% (95% confidence limits from 53% to 74%) ⇒ Interaction

4. Discussion

In this study we used a novel statistical approach to explain the occurrence of mixed dysgraphia, and found that this neuropsychological picture can be traced back to a variety of causes. For a first group of five patients (“double deficit” patients) the spelling performance could be explained (see Fig. 1) merely by a concurrent damage to the orthographic output lexicon (causing impaired spelling of irregular words) and to the auditory-to-phonological conversion (causing impaired spelling of non-words). Following this account, the relatively spared performance on regular words would be explained as the effect of a possible bypass of the acoustic-to-phonological conversion procedure via a shunt along the phonological input and output lexicons. In principle, this pattern of damage would be sufficient to give an advantage to the spelling of regular words over non-words and irregular words, without needing to introduce an interaction phenomenon.

Mixed dysgraphia could be explained also by an alternative type of double functional deficit, i.e., the acoustic-to-phonological conversion and a disconnection between conceptual

Table 6 – Error distribution

Group	Patient	Errors in 80 regular words				Errors in 55 ambiguous words				Errors in 25 non-words				
		Ph	Other	Total errors	Wrong responses	Ph	Ph-pl	Other	Total errors	Wrong responses	Ph	Other	Total errors	Wrong responses
Two deficits	DR	9	5	14	14	15	13	5	33	29	9	6	15	15
Two deficits	EG	8	8	16	16	18	9	13	40	35	13	4	17	17
Two deficits	LZ	21	6	27	25	18	14	13	45	40	19	0	19	17
Two deficits	MR	11	6	17	17	14	13	10	37	33	9	7	16	14
Two deficits	UB	13	1	14	13	14	11	3	28	28	16	3	19	17
Unclassified	LMi	14	3	17	16	18	11	7	36	27	17	4	21	21
Unclassified	CG	21	7	28	27	17	3	14	34	33	6	12	18	18
Unclassified	MG	6	1	7	7	12	5	0	17	14	8	7	15	15
Independent cooperation	PV	4	3	7	7	10	9	5	24	22	6	1	7	7
Interaction	AL	6	9	15	15	11	6	16	33	33	6	6	12	12
Interaction	LMo	13	4	17	17	23	7	7	37	33	10	7	17	17
Interaction	RL	18	12	30	29	21	5	11	37	36	12	12	24	23

Patients are grouped for: (i) possible double cognitive deficit (i.e., acoustic-to-phonological conversion plus orthographic output lexicon); (ii) unclassifiable status of the acoustic-to-phonological conversion routine; (iii) independent cooperation between the two spelling routes; and (iv) interaction between the two spelling routes. Ph = single or multiple graphemic errors; Ph-pl = phonologically plausible errors; other = other errors.

knowledge and the undamaged orthographic output lexicon. In this case, the spelling of regular words would be granted along an alternative (lexical) path, i.e., from auditory analysis to phonological input lexicon to phonological output lexicon to orthographic output lexicon, thus bypassing the lexical-semantic access. Spelling along a direct lexical path would imply a prevalence of lexical homophone but non-homograph substitutions (blue ⇒ blew, or write ⇒ right). However, such couples are represented by less than five occurrences in Italian, and more generally this type of error can only be detected by means of sentence spelling. Over and above this argument, the vast majority of misspellings observed in double lesion patients (see Table 6) were phonologically plausible substitutions, i.e., non-lexical errors that cannot derive from compensation yielded by a 10 direct lexical shunt.

In any case, for four patients the two above types of double deficit could be ruled out. For these cases, we assumed that the information fed forward by the phonological buffer was not biased in favour of regular words, and it was worth analysing whether the two spelling routes showed some type of cooperation. For one patient the observed hit rate on regular words could be traced back to simple cooperation between the two routes, i.e., these routes only combine their independent success probabilities without any intrinsic advantage from a trade-off between the two spelling routes. For three cases our analysis argued in favour of a real reinforcement of regular word spelling due to some interplay between the lexical and the sublexical spelling routes.

For the three remaining cases, the pattern could not be clearly specified.

These findings suggest that spelling in Italian cannot be oversimplified as ‘non-lexical’. Indeed, the Italian shallow orthography allows spellers mainly (but not only) to rely on the sub-word-level routine. English speaking subjects, on the other hand, have to rely far more extensively (though not exclusively) on the lexical route. Hence, at a more general

level, one can say that the two spelling routes are used in languages with shallow and opaque orthography with a different weight both during acquisition (Zoccolotti et al., 2005) and later. This is analogous to the phenomenon observed in the field of acquired dyslexia, where Italian reading is not simply ‘non-lexical’ (Gerhand et al., 2000).

The approach followed here for deciding whether the lexical and sublexical routes operate independently resembles the calculation of the race-model violation (Miller, 1982, 1986; Savazzi and Marzi, 2004). In an earlier study, Luzzatti et al. (1998) offered this explanation to account for mixed dysgraphia; hence we think it useful to discuss in more detail the similarities and differences between the method followed in the present study and Miller’s approach. The race-model applies to reaction time (RT) studies, where RTs to a single stimulus are typically slower than to two identical stimuli (for instance, one in the right and one in the left visual field). The race-model assumes that RTs are distributed with a certain dispersion, and that on each trial the stimulus that, by chance, has a shorter processing time arrives first and is the registered one. Even with an independent processing of the two simultaneous stimuli the model implies that on each trial the faster process would be registered, so that the mean time observed after the simultaneous stimuli will be shorter than that with a single presentation. The analogy with our case might be that: (i) the two writing routes (lexical and sublexical) correspond to the two sides of presentation of the RT studies; (ii) non-words and irregular words are viewed as the single stimulus in RT experiments; and (iii) regular words correspond to the multiple stimulus condition.

At closer scrutiny, it is clear that the race-model does not apply to our study. In RT experiments the stimulus always “arrives” at the post (in terms of the horse race) and what matters is time, whereas in our case a stimulus often does not “arrive” (whenever spelling of a given word is wrong). As such, race-model experiments can work on the continuous

cumulative distributions of the RTs, whereas, with discrete data, we are confined to the study of a binomial model. Consequently, we verified the independence hypothesis with the study of a discrete probability process.

As anticipated in Section 1, due to the peculiarity of the Italian language, ambiguous/irregular words have a considerable chance of being spelled correctly by mere chance, because the alternative spellings of an ambiguous/irregular word are only two, or exceptionally three.⁴ On this basis, the number of errors observed with ambiguous/irregular words is probably underestimated, and the efficiency of the lexical route overestimated. This potential overestimation, in turn, would artifactually increase the expected rate of correct regular words for each subject, and makes it more difficult to detect an interaction. Therefore, the proof that interaction exists will not be marred by this artefact. On the contrary, whenever no interaction is found, we are left with the doubt that this might be due to an overestimation of the lexical route efficiency. By this line of reasoning, the reality of the interaction effect has a stronger support than the reality of the simple cooperation pattern.

The case for the existence and the relevance of a simple cooperation pattern looks not very convincing also for other reasons. Firstly, it rests on a failed rejection of the statistical null hypothesis (see statistical methods and Table 5). Secondly, as explained earlier, our calculation of the expected hits for regular words yields the maximum among a set of possible values, but a lower estimate might be more realistic if the wrong spelling arising from one route sometimes reaches the output system earlier than the correct spelling yielded by the other route. Clearly, the latter possibility strengthens the data supporting the existence of an interaction. Thirdly, patient PV, whose profile suggests only an independent cooperation, was very mild in comparison to the other patients, and small effects might be more difficult to detect.

At this stage, we are far from proposing an exhaustive model that exactly predicts the level of spelling impairment for each type of stimuli. Starting from the evidence that some patients suffer from mixed dysgraphia, we excluded the cases with a possible “double deficit” because the latter pattern makes them unsuitable for a sound study of interaction; however we do not exclude that even “double-deficit patients” might show some degree of interaction. The relative performance with non-words and ambiguous words probably depends on the relative impairment of the sublexical routine and lexical route caused by the original brain damage. Interaction might be a general mechanism acting upon the residual resources of both spelling routes, independently of the level of their impairment.

The question for future inquiries is whether or not the presence of interaction, whose existence has been now demonstrated also for Italian patients, is the rule in mixed dysgraphia, or whether independent cooperation might be

a possible alternative mode of operating for the defective spelling system.

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Appendix A

List of stimuli (WF = written word frequency according to Bortolini et al., 1972)

Section 1

80 regular words	WF	Syllables	Clusters	Doubled consonants
sole (<i>sun</i>)	173	2	no	no
muro (<i>wall</i>)	93	2	no	no
mano (<i>hand</i>)	196	2	no	no
mare (<i>sea</i>)	223	2	no	no
vino (<i>wine</i>)	78	2	no	no
filo (<i>thread</i>)	5	2	no	no
mulo (<i>mule</i>)	10	2	no	no
lume (<i>lamp</i>)	10	2	no	no
faro (<i>light</i>)	9	2	no	no
remo (<i>oar</i>)	0	2	no	no
lavoro (<i>work</i>)	267	3	no	no
numero (<i>number</i>)	149	3	no	no
nuvola (<i>cloud</i>)	27	3	no	no
fenomeno (<i>phenomenon</i>)	21	4	no	no
minerale (<i>mineral</i>)	26	4	no	no
semaforo (<i>traffic lights</i>)	0	4	no	no
sinonimo (<i>synonymous</i>)	0	4	no	no
sifone (<i>siphon</i>)	0	3	no	no
visone (<i>mink</i>)	0	3	no	no
somaro (<i>donkey</i>)	0	3	no	no
senso (<i>sense</i>)	123	2	yes	no
forma (<i>form</i>)	119	2	yes	no
frase (<i>sentence</i>)	34	2	yes	no
marmo (<i>marble</i>)	33	2	yes	no
nervo (<i>nerve</i>)	19	2	yes	no
sfera (<i>sphere</i>)	0	2	yes	no
frana (<i>landslide</i>)	8	2	yes	no
farsa (<i>farce</i>)	0	2	yes	no
selva (<i>forest</i>)	0	2	yes	no
salsa (<i>sauce</i>)	0	2	yes	no
ferro (<i>iron</i>)	69	2	no	yes
valle (<i>valley</i>)	83	2	no	yes
messa (<i>Mass</i>)	47	2	no	yes
sonno (<i>sleep</i>)	41	2	no	yes
villa (<i>large house</i>)	30	2	no	yes
muffa (<i>mould</i>)	5	2	no	yes
molla (<i>spring</i>)	7	2	no	yes
masso (<i>rock</i>)	0	2	no	yes
rullo (<i>roll</i>)	0	2	no	yes

⁴ Current models of sublexical processing propose that the phonological-to-orthographic correspondence is weighted according to occurrence or alternatively that it is rule-based. With respect to “ambiguous” words in the Italian language, the choice between alternatives is very limited (two or seldom three alternatives).

Section 1 (continued)				
80 regular words	WF	Syllables	Clusters	Doubled consonants
renna (<i>reindeer</i>)	0	2	no	yes
capo (<i>head</i>)	145	2	no	no
tipo (<i>type</i>)	128	2	no	no
dito (<i>finger</i>)	45	2	no	no
coda (<i>tail</i>)	29	2	no	no
buco (<i>hole</i>)	15	2	no	no
tubo (<i>pipe</i>)	3	2	no	no
baco (<i>worm</i>)	8	2	no	no
buca (<i>hole</i>)	9	2	no	no
diga (<i>dam</i>)	0	2	no	no
topo (<i>mouse</i>)	11	2	no	no
clima (<i>climate</i>)	88	2	yes	no
festa (<i>holiday</i>)	78	2	yes	no
prato (<i>meadow</i>)	49	2	yes	no
monte (<i>mountain</i>)	144	2	yes	no
spesa (<i>expense</i>)	50	2	yes	no
fusto (<i>stalk</i>)	13	2	yes	no
lardo (<i>bacon fat</i>)	0	2	yes	no
mirto (<i>myrtle</i>)	0	2	yes	no
trave (<i>beam</i>)	7	2	yes	no
stelo (<i>stem</i>)	0	2	yes	no
letto (<i>bed</i>)	176	2	no	yes
notte (<i>night</i>)	228	2	no	yes
matto (<i>mad</i>)	87	2	no	yes
babbo (<i>dad</i>)	27	2	no	yes
latte (<i>milk</i>)	32	2	no	yes
tappo (<i>cork</i>)	0	2	no	yes
ratto (<i>rat</i>)	0	2	no	yes
lutto (<i>mourning</i>)	10	2	no	yes
fetta (<i>slice</i>)	9	2	no	yes
mappa (<i>map</i>)	0	2	no	yes
strada (<i>road</i>)	311	2	yes	no
fronte (<i>forehead</i>)	120	2	yes	no
nastro (<i>band</i>)	8	2	yes	no
stampa (<i>print</i>)	28	2	yes	no
sponda (<i>bank</i>)	11	2	yes	no
crosta (<i>scab</i>)	8	2	yes	no
fulcro (<i>fulcrum</i>)	0	2	yes	no
filtro (<i>filter</i>)	0	2	yes	no
frusta (<i>whip</i>)	3	2	yes	no
tromba (<i>trumpet</i>)	6	2	yes	no

Section 2 (continued)		
55 ambiguous/irregular words	WF	Syllables
paglia (<i>straw</i>)	8	2
olio (<i>oil</i>)	74	2
balia (<i>nurse</i>)	8	2
vigilia (<i>eve</i>)	10	3
milione (<i>million</i>)	145	3
Italia (<i>Italy</i>)	256	3
ognuno (<i>everybody</i>)	35	3
segno (<i>sign</i>)	215	2
compagno (<i>partner</i>)	59	3
pugnale (<i>dagger</i>)	9	3
ragno (<i>spider</i>)	0	2
niente (<i>nothing</i>)	588	2
genio (<i>genius</i>)	13	2
geranio (<i>geranium</i>)	7	3
paniere (<i>basket</i>)	0	3
cerniera (<i>zipper</i>)	0	2
libro (<i>book</i>)	86	2
fibra (<i> fibre</i>)	9	2
zebra (<i>zebra</i>)	0	2
litro (<i>litre</i>)	12	2
cetra (<i>cithara</i>)	0	2
febbre (<i>fever</i>)	16	2
labbro (<i>lip</i>)	34	2
quattro (<i>four</i>)	193	2
fabbro (<i>smith</i>)	0	2
spettro (<i>ghost</i>)	0	2
cuoco (<i>cook</i>)	9	2
scuola (<i>school</i>)	102	2
cuore (<i>heart</i>)	136	2
cuoio (<i>leather</i>)	12	2
scuotere (<i>shake</i>)	19	3
squalo (<i>shark</i>)	0	2
aquila (<i>eagle</i>)	9	3
quarto (<i>fourth</i>)	51	2
quarzo (<i>quartz</i>)	0	2
squama (<i>scale</i>)	0	2
quota (<i>quota</i>)	0	2
liquore (<i>liqueur</i>)	11	3
equo (<i>fair</i>)	0	2
iniquo (<i>iniquitous</i>)	0	3
obliquo (<i>oblique</i>)	0	3

Section 2

55 ambiguous/irregular words	WF	Syllables
scienza (<i>science</i>)	31	2
coscienza (<i>conscience</i>)	34	3
igiene (<i>hygiene</i>)	5	3
società (<i>society</i>)	110	3
usciera (<i>usher</i>)	0	3
ascella (<i>armpit</i>)	0	3
gente (<i>people</i>)	202	2
scena (<i>scene</i>)	45	2
macello (<i>slaughterhouse</i>)	0	3
conoscenza (<i>knowledge</i>)	20	4
aglio (<i>garlic</i>)	0	2
caviglia (<i>ankle</i>)	0	3
vaniglia (<i>vanilla</i>)	0	3
migliaia (<i>thousand</i>)	42	3

Section 3

25 non-words	WF	Syllables	Doubled consonants
nise	0	2	no
nifo	0	2	no
ralo	0	2	no
vona	0	2	no
relo	0	2	no
vimàne	0	3	no
forèla	0	3	no
rinàfo	0	3	no
ramàsola	0	4	no
lesònimo	0	4	no
fella	0	2	yes
seffa	0	2	yes
sovva	0	2	yes
nissa	0	2	yes
sinno	0	2	yes

(continued on next page)

Section 3 (continued)			
25 non-words	WF	Syllables	Doubled consonants
tido	0	2	no
pabo	0	2	no
bita	0	2	no
dute	0	2	no
puda	0	2	no
nitta	0	2	yes
loppa	0	2	yes
rebba	0	2	yes
mippo	0	2	yes
satto	0	2	yes

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